Ann Dermatol. 2024 Apr;36(2):120-122
https://doi.org/10.5021/ad.21.101
pISSN 1013-9087·eISSN 2005-3894

AD ANNALS of DERMATOLOGY

Brief Report

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Rare Cutaneous Soft Tissue Sarcomas Treated With Slow Mohs Micrographic Surgery: Ten Years' Experience at a Single Institution

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Dear Editor:

With the widespread adoption of Mohs micrographic surgery (MMS) for the management of skin cancer, dermatologic surgeons are increasingly being consulted in cases of cutaneous soft tissue sarcoma. Soft tissue sarcoma is a relatively uncommon malignancy of mesenchymal origin[1]. Although there are limited data in the literature, surgery appears to be the procedure of choice for all patients with localized cutaneous soft tissue sarcomas. The guidelines of the European Society for Medical Oncology recommend excision with a wide surgical margin for primary, low-grade, deep, soft tissue sarcomas ≤5 cm, while surgery with or without adjuvant radiotherapy is suggested for primary, high-grade, deep tumors >5 cm.[2] At present, there is no consensus regarding the surgical margin, but the National Comprehensive Cancer Network Soft Tissue Sarcoma Guidelines recommend 1 cm of normal tissue or intact fascia layer[3]. Although MMS has been suggested to be a useful method for treatment of cutaneous malignant tumors due to its advantages of margin control with preservation of normal tissue, there is a paucity of published data on the optimal surgical margin and outcomes of slow MMS. Here, we describe a variety of cutaneous soft tissue sarcomas diagnosed at a dermatologic surgery unit, and aimed to determine the optimal surgical margin for slow MMS to minimize tumor recurrence.

This retrospective study was conducted at the Dermatologic Surgery Clinic, Severance Hospital, which is a tertiary referral teaching hospital affiliated with Yonsei University in Seoul, South Korea. The study conformed to the ethical guidelines of the 1975 Declaration of Helsinki and was approved by the ethics committee of Yonsei Severance Hospital (No. 2019-2564-001). The medical records of patients who underwent surgery for rare cutaneous soft tissue sarcomas at the clinic between January 2009 and December 2018 were reviewed. Kaposi's sarcoma and dermatofibrosarcoma protuberans, which were the two most common cutaneous soft tissue sarcomas, and angiosarcomas treated primarily with radiotherapy or chemotherapy, were excluded.

During the 10-year period from 2009 to 2018, 12 patients with cutaneous soft tissue sarcomas (8 males and 4 females; mean age at diagnosis, 57 years) were treated using MMS. The two most common types of cutaneous soft tissue sarcoma were undifferentiated pleomorphic sarcoma (3/12; 25.0%) and angiosarcoma (3/12; 25.0%), followed by one case each (1/12; 8.3%) of clear cell sarcoma, leiomyosarcoma, myxofibrosarcoma, Ewing's sarcoma (extraskeletal Ewing's sarcoma), rhabdomyosarcoma, and dendritic cell sarcoma. The extremities were the most common tumor

Received: Jun 14, 2021
Revised: Dec 13, 2022
Accepted: May 9, 2023
Published online: Feb 16, 2024

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Table 1. Clinical information on the patients with various soft tissue sarcomas treated with slow MMS

Patient No.	Diagnosis	Age/ Sex	Site	Size (cm)	Surgical treatment	No. of stages for slow MMS	Total surgical margin (cm)	Reconstruction method	Metastasis (organ)	Local recurrence	Adjuvant treatment	Follow-up period	Status at last follow-up
1	Dendritic cell sarcoma	56/F	Back	6.5×4.0	Slow MMS	1	0.5	Primary closure	POY 1 (not certain)	POM 6		1.5 yr	Deceased
2	Leiomyosarcoma	82/M	Back	7.5×7.5	Slow MMS	2	1.5	FTSG	POY 3 (lung)	-		4.5 yr	Deceased
3	Ewing sarcoma	28/M	Buttock	1.5×1.9	Slow MMS	1	1.0	Multilayered purse string suture	POY 1 (lung)	-		1 yr	Alive
4	Myxofibrosarcoma	72/F	Elbow	2.0×1.9	Slow MMS	1	0.5	Rhomboid transposition flap	-	-		4 yr	Alive
5	Undifferentiated pleomorphic sarcoma	77/M	Buttock	3.5×2.8	Slow MMS	1	1.0	Multilayered purse string suture	-	-		3.5 yr	Alive
6	Undifferentiated pleomorphic sarcoma	69/M	Arm	0.9×0.6	Slow MMS	1	1.0	Multilayered purse string suture	-	-		1 yr	Alive
7	Undifferentiated pleomorphic sarcoma	69/M	Arm	0.8×0.6	Slow MMS	1	1.0	Multilayered purse string suture	-	-		2 yr	Alive
8	Rhabdomyosarcoma	5/F	Cheek	0.4×0.4	Slow MMS	1	0.5	Multilayered purse string suture	-	-		3 yr	Alive
9	Clear cell sarcoma	33/F	Arm	1.0×1.2	Slow MMS	1	1.0	Multilayered purse string suture	-	-		7 mo	Alive
10	Angiosarcoma	75/M	Scalp	8.5×9.8	Slow MMS	2	1.0	Secondary intention healing	POY 1 (lung)		Radiotherapy/ chemotherapy	3.5 yr	Deceased
11	Angiosarcoma	69/M	Scalp	3.0×3.0	Slow MMS	2	1.5	O to Z flap	POM 6 (lung)		Radiotherapy/ chemotherapy	8 mo	Alive
12	Angiosarcoma (well-differentiated)	51/M	Ankle	1.7×1.3	Slow MMS	1	0.5	Secondary intention healing	-	-	Radiotherapy	9 yr	Alive

F: female, M: male, MMS: Mohs micrographic surgery, FTSG: full thickness skin graft, POM: post-operative month, POY: post-operative year.

site (5/12; 41.7%), followed by the trunk (4/12; 33.3%) and head and neck (3/12; 25%). Three patients diagnosed with Ewing's sarcoma, rhabdomyosarcoma, and clear cell sarcoma were <40 years old at the time of presentation, while the patient with rhabdomyosarcoma was 5 years old. The remaining patients were >50 years old. All 12 patients were immunocompetent, with no history of other malignancies.

All patients were treated with slow MMS, with the aim of preserving normal tissue. The tumor was excised and precisely mapped, and placed in 10% formaldehyde solution. Histologic evaluation for the formalin-fixed paraffin-embedded horizontal sections of the tumor was performed. In detailed process of the horizontal section, the tumor was excised along designed margin in 45-degree angle, creating beveled sloping specimen comparable to bowl-shape configuration. This method of excision allows the histotechnician to flatten the undersurface of the specimen more easily into an even horizontal plane. In this horizontal section technique, both lateral and deep margin can be demonstrated in every sections[4]. The process was repeated with tissue excised only at positive margins, allowing complete tumor removal while sparing normal tissue.

The mean follow-up period was 2.85 years (range: 7 months to 9 years) after the first stage of slow MMS. Only 1 (8.3%) of the 12 patients treated with slow MMS experienced local recurrence within 6 months of surgery (patient 1; 6.5-cm large dendritic cell sarcoma on the lower back). Although no metastasis was found in any patient at the first visit, distant metastasis occurred in five patients treated with slow MMS, including advanced angiosarcoma in two patients (patients 10 and 11) and dendritic cell sarcoma, leiomyosarcoma, and Ewing's sarcoma in one patient each (patients 1, 2 and 3, respectively). The detailed characteristics and clinical information of the patients are summarized in **Table 1**.

In this study, we showed that slow MMS might be a useful modality for treating cutaneous sarcoma in a certain special circumstance; even with a surgical margin of 0.5–1 cm, most patients did not experience local recurrence and satisfactory cosmetic outcomes were achieved (**Fig. 1**). In particular, cutaneous sarcomas ≤2 cm were treated successfully with a surgical margin of 0.5 cm, with neither local recurrence nor distant metastasis (patients 4, 8, and 12). However, we found that the tumors >5 cm (patients 1, 2, and 10) metastasized to other organs after surgery. Therefore, we could not determine the optimal surgical margin, especially for larger tumors (>5 cm in diameter). In addition, metastasis to the lung was observed in a patient with Ewing's sarcoma <5 cm. With regard to tumor subtypes, distant metastasis occurred in two patients with angiosarcoma, which is known to be highly aggressive[5]. Although primary cutaneous leiomyosarcoma and Ewing's sarcoma rarely metastasize[6-8], distant metastasis to the lungs was also observed in patients with these two subtypes of cutaneous soft tissue sarcoma in our cohort. Therefore, soft tissue sarcomas require individualized management based on the tumor subtype, size, depth, and lymph node involvement.

https://doi.org/10.5021/ad.21.101

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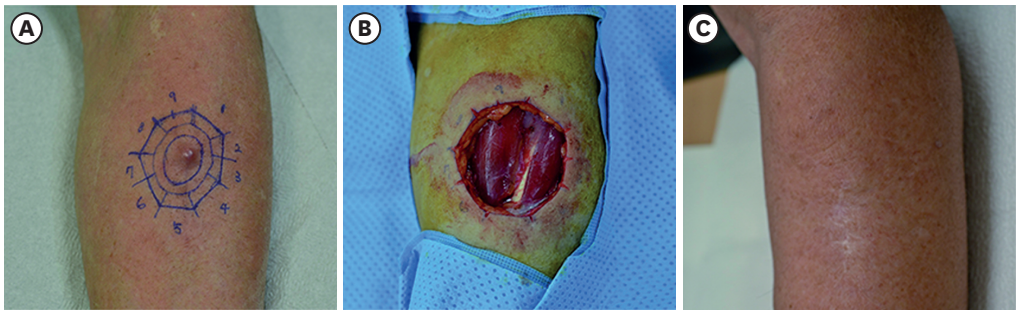

Fig. 1. Clinical pictures and surgical outcomes of patients with pleomorphic sarcoma. (A) A 1-cm surgical margin was taken for slow Mohs micrographic surgery in a patient with pleomorphic sarcoma (patient 7). (B) The tumor was excised completely. Multilayered purse-string sutures with subsequent pulley buried dermal sutures were applied. (C) At 1 year later, the patient had a minimal surgical scar on his arm, which showed satisfactory functional and cosmetic outcomes.

This study had some limitations. Due to the small number of cases and lack of a control group, we could not perform statistical analysis to determine whether slow MMS was more beneficial than wide excision for managing soft tissue sarcoma. The extremely low prevalence of cutaneous sarcoma in Korea and lack of previous reports precluded further statistical analysis of the data. In addition, specific subtypes were not determined in some tumors, such as dendritic cell sarcoma, because immunohistochemical staining was positive for only CD68 and negative for D2-40, CD21, p53, smooth muscle actin, S-100, and anaplastic lymphoma kinase in atypical spindle cells. Moreover, the duration of follow-up period was not long in some patients.

Nevertheless, this retrospective review had some strengths. To our knowledge, this is the first slow MMS case series demonstrating good clinical and cosmetic outcomes for a variety of unusual cutaneous soft tissue sarcomas. We suggest that slow MMS may be a useful modality for treating patients with cutaneous sarcoma. In addition, this study provided valuable information related to the treatment of rare cutaneous soft tissue sarcomas based on real-world experience.

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FUNDING SOURCE

This work was supported by the Soonchunhyang University Research Fund.

CONFLICTS OF INTEREST

The authors have nothing to disclose.

DATA SHARING STATMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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