Flexor Digitorum Superficialis to Flexor Digitorum Profundus Tendon Transfer after Wide Excision of Myxoinflammatory Fibroblastic Sarcoma: A Case Report

Bo Hyun Lee¹, Lan Sook Chang¹, Youn Hwan Kim¹, Chang Hun Lee², Seong Oh Park³

¹Department of Plastic and Reconstructive Surgery, Hanyang University College of Medicine, Seoul, Korea
²Department of Orthopedic Surgery, Hanyang University College of Medicine, Seoul, Korea
³Department of Plastic and Reconstructive Surgery, Seoul National University Hospital, Seoul National University College of Medicine, Seoul, Korea

Abstract

Myxoinflammatory fibroblastic sarcoma (MIFS) is a rare, low-grade soft tissue neoplasm that typically arises in the extremities. A 72-year-old woman presented with recurrent masses on her left forearm. Four years prior, she underwent an excisional biopsy, diagnosed as MIFS, followed by wide excision and split-thickness skin graft coverage. Preoperative magnetic resonance imaging was conducted for the recurrent mass, revealing multiple tumors invading the flexor digitorum profundus (FDP) muscle and fascia. The tumors including most of the FDP muscle were therefore excised. The third flexor digitorum superficialis (FDS) tendon was harvested, divided in half, and connected to the second and fifth FDP tendons. The same procedure was performed for the third and fourth FDP tendons with fourth FDS tendon. An anterolateral thigh free flap was used to reconstruct the skin and soft tissue defect of the left forearm. Adjuvant radiation therapy was performed. At 12 weeks postoperatively, the patient exhibited no wound complications and achieved spontaneous partial flexion of the metacarpophalangeal and proximal interphalangeal joints.

Keywords: Sarcoma; Free tissue flaps; Tendon transfer; Case reports

Introduction

Myxoinflammatory fibroblastic sarcoma (MIFS) is a rare, low-grade soft tissue neoplasm characterized by a mixture of myxoid and fibroblastic components, varying degrees of inflammation, and a distinctive immunohistochemical profile [1-3]. While MIFS typically originates in the distal extremities [1], cases arising in the proximal extremities have also been reported [2,4-6]. Though a rare affliction, recognizing MIFS is important because of its potential for local recurrence and distant metastasis [1]. In this report, we present the case of a patient with recurrent MIFS in the upper extremity treated with wide excision of cancer, repair of the flexor digitorum profundus (FDP) using a flexor digitorum superficialis (FDS) tendon transfer, and coverage with an anterolateral thigh (ALT) free flap. Though MIFS has been mentioned in several pathology journals, there has been little exploration in journals dealing with clinical management. We introduce this case to highlight the process of functional defect management after MIFS tumor ablative surgery and the progression of recurrent tumor. The report was approved for exemption by the Institutional Review Board of Hanyang University Hospital (IRB exemption no. 2022-12-038). The patient provided written informed consent for the publication and use of images.
Case

A 72-year-old woman on hemodialysis for end-stage renal disease and a medical history of hypertension and diabetes mellitus, presented with recurrent masses on her left forearm (Fig. 1). Four years prior, she underwent an excisional biopsy and was diagnosed with MIFS, followed by wide excision and coverage with a split-thickness skin graft.

Preoperative magnetic resonance imaging (MRI) revealed multiple nodules in the left forearm with invasion of the FDP muscle and muscular fascia, suggesting a recurrent tumor (Fig. 2). No distant metastasis was found on bone scan or whole-body positron emission tomography-computed tomography. Preoperative brachial angiography revealed patent radial and ulnar arteries, with no evidence of stenosis (Fig. 3).

In consideration of the size and extent of the lesion, we performed a longitudinal elliptical-shaped incision to encompass it (Fig. 4). The excision extended through the skin, subcutaneous tissue, fascia, and flexor muscles, resulting in near-total resection of the FDP and partial resection of the flexor carpi ulnaris, reaching the periosteal layer of the ulna in an en-bloc fashion (Fig. 5A). A frozen biopsy confirmed margin-free resection.

Subsequently, a longitudinal skin incision was made along the volar aspect of the left wrist. After identifying the median nerve, further dissection was performed with careful attention to prevent nerve injury. Intraoperatively, we found the second digit compartment of the FDS ruptured at the wrist level, and severe adhesion in the soft tissue around the flexor tendons.

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**Fig. 1.** The patient’s preoperative photograph. A 72-year-old woman presented with a recurrent mass of the left forearm.

**Fig. 2.** Preoperative magnetic resonance imaging (MRI). Preoperative MRI revealed multiple nodules in the left forearm of approximately 8 cm in longitude, with invasion of the flexor digitorum profundus muscle and muscular fascia, suggestive of a recurrent tumor. (A) Coronal view. (B, C) Axial view.

**Fig. 3.** Preoperative angiography. Both the radial and ulnar arteries were found to be patent, with no evidence of stenosis in brachial angiography.
due to previous surgeries. Adhesiolysis was performed to address these adhesions.

The third FDS tendon was identified, cut distally at an appropriate length, and divided into two halves. One half was connected to the second FDP, and the other to the fifth FDP using the Pulvertaft method (Fig. 5B) [7]. While applying passive extension and flexion to the fingers, the tension of the FDS was carefully adjusted for optimal functionality during FDP reconstruction using Prolene 4-0 loop sutures at the proximal and distal anastomosis sites. The same method was employed using the fourth FDS tendon to reconstruct the third and fourth FDP.

To cover the skin and soft tissue defect with exposed bone, a 17×5.5 cm-sized ALT flap was harvested from the right thigh (Fig. 6). The flap’s pedicle was an approximately 10 cm-long descending branch of the lateral circumflex femoral artery (LCFA). An end-to-side anastomosis was performed between the descending branch of the LCFA and the ulnar artery. Additionally, end-to-end anastomosis was conducted between the vena comitantes.

The final biopsy results confirmed recurrent MIFS with clear resection margins. The mitotic rate was less than 1/10 high-

![Fig. 4. Preoperative photograph. The previous surgical scar and the tumor’s location were marked with dotted lines, and the incision line was marked with a solid line.](image)

![Fig. 5. Intraoperative photographs. (A) Wide excision of tumor including most of the flexor digitorum profundus (FDP) with a periosteal layer of the ulna at forearm level was performed. The recipient vessel was the radial artery (tagged with a vessel loop). (B) The third flexor digitorum superficialis (FDS) was divided in half and connected to the second FDP and fifth FDP tendon in the Pulvertaft method. The same procedure was performed for the third FDP and fourth FDP with fourth FDS.](image)

![Fig. 6. Photograph of harvested anterolateral thigh (ALT) flap. An 18×6 cm-sized ALT flap was harvested for left forearm reconstruction.](image)
power fields (HPFs), and no necrosis was observed, consistent with the previous biopsy findings. The patient recovered well and was discharged without complications (Fig. 7).

Following discharge, adjuvant radiation therapy was performed with a total dose of 60 Gy in 30 fractions. At 12 weeks postoperatively, the patient exhibited no wound problems and achieved spontaneous partial metacarpophalangeal joint flexion, with approximately 70° of active range of motion (ROM), and proximal interphalangeal joint flexion, with approximately 40° of active ROM in the outpatient follow-up (Fig. 8).

One year after discharge, a new mass lesion was observed around the surgical site, raising suspicion of local recurrence (Fig. 9). Subsequently, another round of wide excision and full-thickness skin graft was performed. Biopsy results showed an increase in the mitotic count to 8/10 HPFs compared to the previous count. ROM remained unchanged at 12 weeks post-surgery. About 1 month later, local recurrence was confirmed once again. Although above-elbow amputation was recommended, the patient declined, leading to another local excision. Unfortunately, subsequent progression to axillary lymph node metastasis and bone metastasis occurred. The patient is currently under consideration for systemic chemotherapy.

Fig. 7. The patient’s postoperative photographs. (A) An immediate postoperative view and (B) 14 days after reconstruction.

Fig. 8. Photographs of postoperative week 12. The patient performed spontaneous partial metacarpophalangeal joint and proximal interphalangeal joint flexion in the outpatient follow-up. (A) Full active flexion in anteroposterior view. (B) Full active extension in anteroposterior view. (C) Full active flexion in lateral view. (D) Full active extension in lateral view.
Discussion

MIFS was initially characterized in 1998 as a relatively rare slow-progressing low-grade soft-tissue sarcoma by Meis-Kindblom and Kindblom [1], Montgomery et al. [2], and Michal [3]. While typically exhibited as painless small masses, MIFS can occasionally cause pain or paresthesia [1,5,6]. Such tumors commonly arise in the distal extremities, including the hands, feet, and lower legs, and a few cases have been reported in the upper arm or thigh, among other proximal locations [2,4-6]. A literature review by Tejwani et al. [6] revealed that 95.6% (174/182) of cases originated in the distal extremities. As sarcomas in general are known to rarely occur in the distal extremities, diagnosing MIFS at an early stage is difficult.

MIFS manifests with diverse MRI findings, ranging from poorly circumscribed multinodular masses with tendon sheath involvement to well-defined lesions, sometimes subcutaneous, with potential muscle or bone invasion [8,9]. Heterogeneity in MIFS varies based on its components, with the myxoid component showing T2 hyperintensity and T1 hypointensity [10]. A recent study by Gaetke-Udager et al. [11] further elucidated this spectrum, highlighting osseous destruction and peripheral enhancement in high-grade areas, alongside diffuse enhancement in the low-grade component.

The pathological features of MIFS include varied proportions of myxoid areas, marked inflammatory infiltration, and the presence of diverse atypical cells such as virocyte-like cells, Reed Sternberg-like cells, and lipoblast-like vacuolated cells [1-3,5,12,13]. Lucas [13] illustrated these diverse histological forms in detail.

Despite only four cases of metastatic MIFS reported to date [1,12,14], the high local recurrence (31.3%) after surgical excision [6] necessitates vigilant postoperative surveillance. Lang et al. [4] recommended quarterly evaluations for the initial 2-year period, followed up every 6 months up to 5 years as is the case for other low-grade sarcomas.

Achieving favorable oncological, functional and aesthetic outcomes often involves radical excision with soft tissue reconstruction techniques like free vascularized flaps or local fasciocutaneous flaps. Additionally, radiation therapy regimens, as seen in Tejwani et al. [6] with preoperative doses of 50 Gy in 25 fractions and postoperative doses ranging from 60 to 63 Gy in 30 to 25 fractions, have demonstrated local control rates exceeding 80%.

In our case, the recurrence occurred in the forearm, a less common location compared to the other distal extremities (1.6%, 2/128) [4]. While neoplastic progression with increased atypical cells compared to primary tumors can occur in recurrent MIFS [1], no significant difference in mitotic activity was observed in the first local recurrence. However, in subsequent instances of local recurrence, an increase in mitotic count was observed. Radiologic and histologic findings aligned with previous studies.

Given the critical role of various forearm muscles in hand movement, reconstructing flexor tendons was crucial for improving the patient’s quality of life. The simultaneous FDS to FDP tendon transfer and ALT free flap yielded excellent aesthetic and functional outcomes, demonstrating the effectiveness of multi-modal approaches in such cases. Considering the locally aggressive nature of MIFS, continuous surveillance remains essential. Moreover, because MIFS predominantly oc-
curs in distal extremities, there may come a point where recurrence warrants considering amputation. It is crucial to engage in thorough discussions with the patient to determine further treatment plans. Additionally, given the potential progression to metastasis as seen in this patient, establishing regimens of systemic chemotherapy through further research is necessary.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

ORCID iDs

Bo Hyun Lee https://orcid.org/0000-0003-1989-0026
Lan Sook Chang https://orcid.org/0000-0003-4725-772X
Youn Hwan Kim https://orcid.org/0000-0003-3365-1232
Chang Hun Lee https://orcid.org/0000-0003-4330-7726
Seong Oh Park https://orcid.org/0000-0001-8990-0635

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