CASE REPORT

Rapid progression of solitary fibrous tumor after induction of hemodialysis

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Abstract
A 42-year-old male patient presented in 2002 with a solitary fibrous tumor (SFT) arising from the visceral pleura of the right lung. Thoracic surgery was performed to remove the tumor. A second operation to remove a recurrent tumor on the parietal pleura of the right thorax was performed in 2010. A follow-up computed tomography (CT) scan revealed local recurrence in the chest wall. And then a third operation involving en bloc resection of chest wall was performed in 2012. Thereafter, a CT scan in 2015 revealed slow-growing local recurrence. In 2016, he was started on hemodialysis. Two months later he was hospitalized because of chest pain and dyspnea. Imaging showed bilateral massive pleural effusion and dissemination along with left pulmonary metastasis. We report a case of SFT recurrence, which rapidly worsened after induction of hemodialysis. Induction of hemodialysis is potentially challenging that may lead to be in a tumor-bearing condition.

INTRODUCTION
Solitary fibrous tumor (SFT) is a rare tumor that arises from mesenchymal cells. While the majority of SFTs have a benign clinical course, the tumor may recur and metastasize after surgical resection, often >10 years after surgery. Approximately 10–20% of SFTs have malignant potential, eventually leading to death [1]. Herein, we present a case of recurrence SFT. To the best of our knowledge, this is the first report of recurring SFT with rapid progression after induction of hemodialysis.

CASE REPORT
A 42-year-old male patient presented in 2002 with a SFT, 5.5 cm in diameter, arising from the visceral pleura of the right upper lung. Video-assisted thoracic surgery was performed to remove the tumor. Histologically, the tumor was composed of spindle cells randomly arranged in fascicle with variable hyalinous collagen fibers. Four mitoses per 10 high-power fields were found, cellularity was not high, cytological atypia and necrosis were not apparent (Fig. 1A and B). MIB-1 immunostaining was
performed retrospectively, it could not be evaluated for deterioration of the paraffin section. Seven years later, in 2009, the patient was asymptomatic but a follow-up computed tomography (CT) scan revealed a pleural tumor with enlargement (Fig. 2A). A second operation to remove the recurrent tumor was performed in 2010, 8 years after the initial surgery. A nodule on the parietal pleura of the right thorax, which was inferred the port-site recurrence in the first operation, was resected completely. Histologic finding was similar to the first specimen, and mitotic index was the same, 4 per 10 high-power fields (Fig. 1C). The tumor cells were immunohistologically positive for CD34 and STAT6. The percentage of MIB-1-positive nuclei was 9.0% (Fig. 3A). After the operation the patient was monitored closely without further treatment. A follow-up CT scan revealed local recurrence in the chest wall (Fig. 2B). A third operation involving en bloc resection of chest wall was performed in 2012, 10 years after the initial procedure. Histology showed 4 mitoses per 10 high-power fields (Fig. 1D). The tumor cells were immunohistologically similar to those in the second operation. The percentage of MIB-1-positive nuclei was 17.0% (Fig. 3B). Thereafter, we believed that the disease had been brought under control, but a follow-up CT scan in December 2015 revealed slow-growing local recurrence again (Fig. 2C).

Meanwhile the patient had been diabetic since 2002, and his poor control of the diabetes led to diabetic nephropathy. In January 2016, 14 years after the initial surgery, he was started on hemodialysis for end-stage renal disease. Two months later he was hospitalized because of chest pain and dyspnea. Imaging showed bilateral massive pleural effusion and left pulmonary metastasis (Fig. 2D). The patient died in April 2016.

**DISCUSSION**

Surgical resection is the mainstay of treatment, and recurrence-free survival generally exceeds 90% after complete resection. However, ~10% of SFTs will recur locally or distantly, often >10 years after surgery. While many local recurrences can be controlled by resection, widespread disease is fatal because of the poor response of SFTs to chemotherapy [2]. Furthermore, several studies have indicated that in frequently recurring SFTs, the length of time between the first and second recurrence is shorter than the interval between the initial diagnosis and the first recurrence, regardless of whether surgery was performed [3]. Frequent surgical resection for recurrent tumors causes aggressive adhesion, making complete surgical resection a considerable challenge [3]. In our case, each recurrence was shorter than the previous one, and worse in immunohistologic malignancy. The percentage of MIB-1-positive nuclei of local recurrence tumor had increased from 9.0 to 17.0% between second operation and third operation. The SFT probably had potential gradually indicating a rise in the aggressiveness of the recurrence in our case.

Long-term clinical follow-up for all patients with SFTs is recommended because of the potential adverse biological behavior of this tumor. In addition, during long-term follow-up patients’ concomitant diseases, such as diabetes and atherosclerosis, can progress and perhaps affect the clinical course of SFTs. In general, it is thought that hemodialysis may affect tumors. Patients with end-stage renal disease who require renal maintenance hemodialysis have a high risk of malignancy induced by depressed host immunity [4]. Moreover, the risk is considered to be progressively higher in patients with a longer duration of dialysis [4]. However, Liu et al. [5] showed...
that the highest standardized incidence ratio of urothelial carcinoma of the bladder occurred in the first 3 years of hemodialysis and that the risk gradually declined with prolonged hemodialysis. Moreover, Ricci [6] reported a significantly lower incidence of metastases in patients on hemodialysis compared with those not on hemodialysis. Given these results, he hypothesized that the dialysis membrane plays important roles in blocking or inactivating tumor cells [6]. More specifically, it is generally thought that circulating tumor cells (CTCs) in cancer patients that disseminate from the primary tumor through the circulatory system, some of which are ultimately capable of forming distant metastases, lead to recurrence.

So, we conjecture that hemodialysis for cancer patients leads to adverse effects, immune suppression and CTCs reduction. In the present case, the patient was started on hemodialysis 14 years after the initial operation, but after only 2 months of hemodialysis was referred to our hospital with dissemination and distant metastasis of SFT, which were different from the heretofore local recurrence. Needless to say, it is possible that the induction of hemodialysis for a coincidence that the rapid progression of tumor is a consequence of the worsening aggressiveness of tumor in our case. But, the change of micro-environments such as immunosuppression mechanism might play an important role for the rapid progression in our patient after induction of hemodialysis. As clinicians we must take into account the immune dysfunction associated with hemodialysis therapy when we introduce hemodialysis to patients in a situation of cancer immune escape or in a similar situation, even in the state of slow-growing local recurrence of SFT. In such a situation, induction of hemodialysis is potentially challenging that may lead to be in a tumor-bearing condition.

![Figure 3: Immunohistochemistry findings. (A and B) Representative examples of immunohistochemistry on formalin-fixed, paraffin-embedded tumor tissues resected in 2010 (A) and 2012 (B) with MIB-1-positive cells. (A, B: anti-MIB-1 immunohistochemical staining; ×40).](https://academic.oup.com/omcr/article-abstract/2017/7/omx037/3926769)

![Figure 2: Histological findings. (A and B) Microscopic view showing that the tumor resected in 2002 was composed of spindle-shaped cells with pattern and a few mitotic cells. (C and D) The recurrent tumors resected in 2010 (C) and 2012 (D). The mitotic counts had been increasing. (Hematoxylin and eosin stain; original magnification A ×10, B-D ×40).](https://academic.oup.com/omcr/article-abstract/2017/7/omx037/3926769)
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CONFLICT OF INTEREST STATEMENT
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ETHICAL APPROVAL
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CONSENT
We obtained written consent from the family of the patient for the publication of this case report and any accompanying images.

GUARANTOR
Motoaki Yasukawa.

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