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CASE REPORT

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Report of two cases of myxoinflammatory fibroblastic sarcomas with preceding hematolymphoid neoplasms: Is there any association?

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Abstract

Myxoinflammatory fibroblastic sarcoma (MIFS) is an uncommon soft tissue sarcoma. We present two cases of MIFS: A known case of Hodgkin's lymphoma presented with hand mass; a recurrence of MIFS with a history of chronic lymphocytic leukemia.

KEYWORDS

case report, hematolymphoid neoplasms, myxoinflammatory fibroblastic sarcoma

1 | INTRODUCTION

Myxoinflammatory fibroblastic sarcoma (MIFS) is an uncommon soft tissue sarcoma, which was described for the first time in 1998.[1] MIFS is a slow-growing, painless subcutaneous mass in the distal extremities of young to middle-aged adults.[2] In histology, MIFS is composed of mixed inflammatory cell infiltrate, Reed-Sternberg-like cells, lipoblast-like cells, and atypical spindle to epithelioid cells in the myxoid and hyaline background.[1] This tumor is a low-grade sarcoma with a high rate of local recurrence and low rate of metastasis; therefore, correct diagnosis and follow-up are important.[3] We present two cases of MIFS: A known case of Hodgkin's lymphoma presented with hand mass; a recurrence of MIFS with a history of chronic lymphocytic leukemia.

2 | CASE PRESENTATION 1

A 25-year-old man was admitted on July 17, 2019 due to a left second metacarpal mass. He had a history of Hodgkin lymphoma and had received 13 courses of chemotherapy. Hodgkin lymphoma was presented 6 years ago with malaise and weight loss for 1 month and fever with dry cough for 3 weeks. Computed Tomography (CT) scanning of lung and mediastinum at that time showed multiple massive lymphadenopathies in the superior and middle mediastinum, subcarina, and azygo-esophageal spaces encasing mediastinum vessels. A moderate amount of pericardial effusion, two small nodules in the lungs, and left side pleural thickening suggested malignant lymphoma. CT scan of the abdomen showed mild hepato-splenomegaly without abdominal

lymphadenopathy. The whole body bone scan was also normal. The patient left the hospital with his consent but continued workup in another center with a biopsy and final diagnosis of Hodgkin lymphoma. The pathology report was not available at the visit in July 2019. At this visit, a physical examination showed soft tissue mass in the dorsum of the hand with mild tenderness without movement restriction. The patient underwent surgery and the specimen was sent to the pathologist in two containers. The first container consisted of two gray pieces measured 4×2.5×0.8 cm and 0.5×0.5×0.5 cm, and the report was compatible with myxoinflammatory fibroblastic sarcoma. The second container consisted of several fragments of gray bony tissue measured 1.5×1×0.5 cm, and the report was fibro-osseous tissue. The pathologist recommended immunohistochemistry (IHC) staining to rule out recurrence of Hodgkin lymphoma due to the presence of inflammatory background and Reed-Sternberg-like cells. IHC was done. CD15 and CD30 were negative in Reed-Sternberg-like cells. CD31 and CD34 were positive in vessels. Meanwhile, CD45 and EMA were negative. IHC pattern was in favor of myxoinflammatory fibroblastic sarcoma (Figure 1).

3 | CASE PRESENTATION 2

A 78-year-old man was admitted on June 20, 2020 due to a hemorrhage from a right forearm mass. The mass measured 25×5×5 cm. In past medical history, he had a history of a similar tumor at the same site which underwent surgery with a pathology diagnosis of well-differentiated liposarcoma, but immunohistochemistry (IHC) had shown S100 negativity. He mentioned a history of chronic lymphocytic leukemia (CLL) but without further documents. Also, a history of ischemic heart disease and coronary artery graft (5 years ago) with nodular hyperplasia of the prostate, renal cortical cyst (32 mm) with renal and urinary bladder stones, and prostatectomy (last year) was noticed. The patient was hypertensive. Drug history included ASA, losartan, amlodipine, temozolomide, chlorambucil, and captopril. The patient underwent surgery on June 25, and a brownish piece of tissue including skin and underlying hemorrhagic gelatinous mass was sent to the pathologist. The total tissue size was 20×15×5 cm and the mass measured 13×12.5×4 cm. In pathologic examination, the presence of frequent Reed-Stenberg-like cells was noticed. IHC panels including CD15, CD30, HMB-45, Melan-A, SMA, Desmin, NSE, CK, EMA, CD34, CD68, and S100 were negative in Reed-Sternberg-like cells. Vimentin was positive and Ki-67 was positive in 10% of cells. The slides of the first tumor (of the last year) were re-examined. There were few Reed-Sternberg-like cells in that slides. In recurrent tumors, the cells were more prominent. The pathologist suggested myxoinflammatory fibroblastic sarcoma (Figure 2). Mitotic figures were <2 mitoses/50 HPF, the tumor's greatest diameter was 13 cm and one peripheral surgical margin was involved. The patient claimed a mass in the chest wall at the last visit of the pathologist on July 12. Written informed consents were obtained from both patients for reporting the cases.

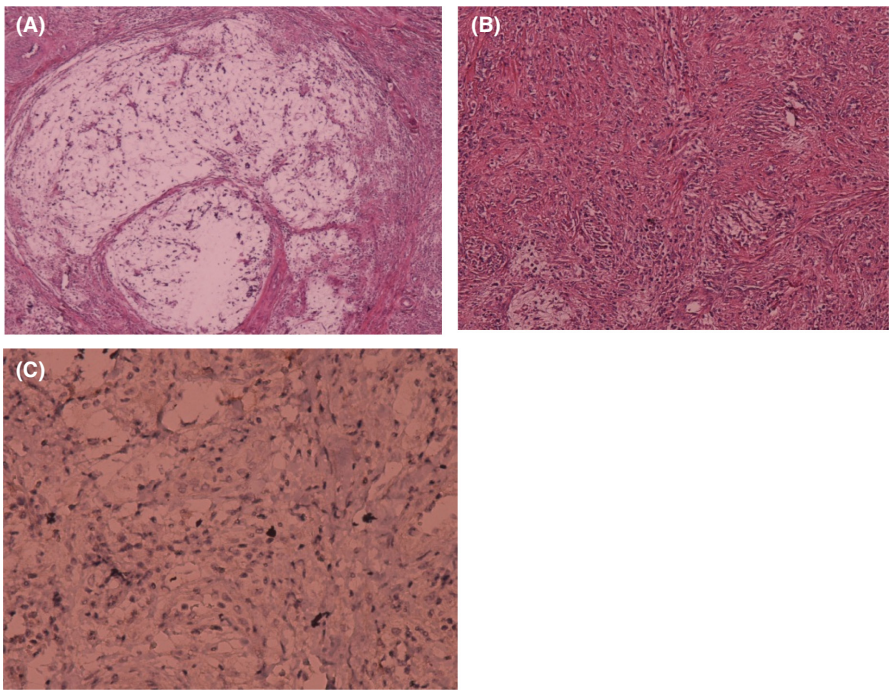

FIGURE 1 Myxoinflammatory fibroblastic sarcoma after Hodgkin lymphoma. (A) Hematoxylin–Eosin Stain ×40. (B) Hematoxylin–Eosin Stain ×100. (C) Immunohistochemistry negativity for CD30 ×200 magnifications

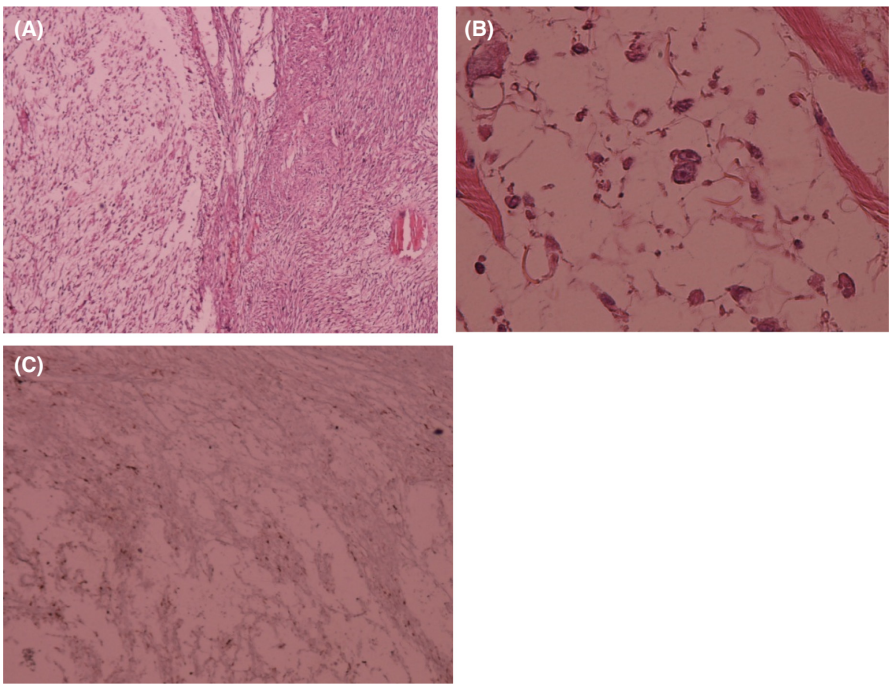

FIGURE 2 Myxoinflammatory fibroblastic sarcoma after chronic lymphocytic leukemia. (A) Hematoxylin–Eosin Stain ×40. (B) Hematoxylin–Eosin Stain ×400. (C) Immunohistochemistry negativity for S100 ×40 magnifications

4 | DISCUSSION

Myxoinflammatory fibroblastic sarcoma is a rather new entity in soft tissue tumors. MIFS is a slow-growing, subcutaneous mass in the distal extremities, especially the upper extremities.[2] Clinical symptoms resemble benign tumors, ganglion cyst, lipoma, fibroma, and inflammatory processes.[3–5] Some cases of MIFS have been reported in uncommon sites including the upper back, scapula, head, and cervicothoracic region.[2,4,6,7] Etiologic causes and risk factors are unknown. Only one study has reported a history of renal transplantation in one case.[8] In the macroscopic examination, consistency is myxoid to hard in subcutaneous tissue with well-defined or infiltrative borders.[1,3,7] MIFS has variegated and heterogeneous microscopic findings and consists of a multinodular appearance with three components: inflammatory cells, areas of fibrosis or hyalinized stroma, and areas of the extracellular myxoid matrix with tumor cells.[2,8] Tumor cells are atypical epithelioid and spindle cells resembling Reed-Sternberg cells, multivacuolated cells resembling lipoblast cells, and ganglion cells without mitosis.[2,3,8] Heterogeneous components of MIFS in small biopsy specimens may be the cause of misdiagnosis. A prominent myxoid component in biopsy specimens may be seen in myxoid malignant fibrous histiocytoma, myxofibrosarcoma, superficial acral fibrosarcoma, myxoid liposarcoma, and myxoma. The prominent presence of inflammatory cells in incomplete excision may be misdiagnosed as an inflammatory myofibroblastic tumor, Hodgkin's lymphoma, and reactive inflammatory processes.[3,5–8] The presence of lipoblast-like cells and epithelioid cells in biopsy may be mistaken as liposarcoma and epithelioid sarcoma respectively.[5–8] Michael Michel et al. reported 23 cases of high-grade MIFS and considered the diagnosis, as a spectrum from low- to high-grade neoplasms.[9] Immunohistochemistry studies of MIFS are variegated but it is important to rule out other differential diagnoses. MIFS is often positive for Vimentin, CD68, and focally for SMA but negative for EMA, Melan-A, HMB-45, S100, CD30, and CD15.[2] Immunostaining positivity of emperipolesis for cyclin D1 is emphasized.[9] MIFS has been associated with t (1,10)(p22, q24) resembles hemosiderotic fibrolipomatous tumor and pleomorphic hyalinizing angiectatic tumor; therefore, MIFS has no characteristic immunohistochemistry and FISH.[2,4,8]MIFS is a postoperative diagnosis and clinicopathologic correlation is necessary for diagnosis.[2,6] MIFS has a high rate of local recurrence of about 22%–67%, a low rate of metastasis, about 2%, and a low mortality rate, but some cases of MIFS with inguinal lymph node involvement, liver, lung, and vertebra metastasis has been reported.[3,4,7] The patient needs wide local excision with a clear margin, follow-up for 5 years after surgery for local recurrence and radiological examination for metastasis evaluation.[3] We presented two cases of challenging MIFS. One case with the history of Hodgkin's lymphoma accompanied by Reed-Sternberg-like cells in microscopic examination of hand mass. We ruled out Hodgkin's lymphoma by immunohistochemistry, and the final diagnosis was made as MIFS following Hodgkin's lymphoma. In another case with a history of liposarcoma and Chronic lymphocytic leukemia presented with limb amputation and wide local recurrence of the previous tumor, the final diagnosis became recurrence of MIFS. So far, no risk factors and etiologic causes have been suggested for MIFS.

Whether there is a relationship between this entity and hematolymphoid neoplasms or this is merely an association by chance must be elucidated in the future.

5 | CONCLUSION

Myxoinflammatory fibroblastic sarcoma is a rare tumor with a diagnostic challenge for Pathologists. It may be associated with hematolymphoid neoplasms or the therapy-related etiology.

AUTHOR CONTRIBUTION

Z.A. conceived of the presented idea, provided the microscopic photographs, and carried out the experiment. M.R. signed out the cases and confirmed the diagnosis. Z.A. and M.R. wrote the manuscript. M.S. revised the manuscript. All authors discussed the results and contributed to the final manuscript.

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CONFLICTS OF INTERESTS

The authors declare that there is no conflict of interest.

ETHICAL APPROVAL

This case report is ethical according to the world medical association declaration of Helsinki.

CONSENT

Written informed consent is obtained from patients to publish this report.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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