

Case Report

Whether inflammatory myofibroblastic tumor of the thigh relapses after surgical excision?

Hao Liu*, Jun Lin*, Peng Yang, Hao Shen, Huilin Yang

*Department of Orthopaedic Surgery, First Affiliated Hospital of Soochow University Suzhou, Jiangsu, China. *Equal contributors.*

Received May 2, 2015; Accepted June 23, 2015; Epub July 15, 2015; Published July 30, 2015

Abstract: As reported previously, the 61-year-old male Chinese patient suffering from the inflammatory myofibroblastic tumor of the right thigh without bone involvement was performed a surgical excision and a local radiotherapy two years ago. However, a moderately soft and painful 200 × 100 mm mass of the posterior thigh, where magnetic resonance imaging (MRI) scans revealed an inhomogeneously hyperintense on T1-weighted imaging (T1WI), T2-weighted imaging (T2WI) and diffusion-weighted imaging (DWI), was found forty days after the operation. Two years into follow-up, we intend to judge whether the tumor relapses by regular imaging examination including thoracic computed tomography (CT) scans and local MRI scans of lower extremities. The diagnosis of postoperative cyst is suggested.

Keywords: Inflammatory myofibroblastic tumor, radiotherapy, magnetic resonance imaging

Introduction

Inflammatory myofibroblastic tumor (IMT) primarily affects children and young adults, whose mean age at diagnosis is 10 years, although the age range extends throughout adulthood [1, 2]. IMT occurs throughout the body, most frequently in the mesentery, omentum, retroperitoneum, pelvis, and abdominal soft tissue in 73% of cases, followed by the lung, mediastinum, and head and neck [3].

Primary treatment of IMTs is complete surgical excision. Meanwhile, chemotherapy, radiotherapy, and anti-inflammatory therapy have been used as an adjuvant therapy with surgery to deal with residual sites [4, 5]. Approximately 25% of extrapulmonary IMT recur in anatomical site and multinodularity [1, 6]. Furthermore, metastasis occurs in <2% of cases [3]. Specially, anaplastic lymphoma kinase (ALK)-negative IMTs may have a higher likelihood of metastasis [7].

We illustrate an unusual case of an ALK-negative IMT of the thigh without bone involvement treated with surgical excision and radio-

therapy. Also, we analyse whether the IMT relapses by regular MRI scans for two years into follow-up.

Case presentation

A 61-year-old male Chinese patient was referred to our service two years ago. According to pre-operative MRI scans and histopathologic examination as previously described [8], the diagnosis of an IMT of the thigh without bone involvement which shows negative immunoreactivity for ALK is specific.

Postoperatively, the patient was treated with a three-dimensional conformal radiotherapy with a total dose of 50Gy/25F (2Gy/F, 5F/week). Pelvic lymphatic drainage area was not included in the irradiation.

The patient have been followed up for two years. Forty days after the operation, MRI scans showed a local lesion in the posterior thigh with an inhomogeneously hyperintense on T1WI, T2WI and DWI (**Figure 1A-C**), and an inhomogeneously hypointense on contrast-enhanced imaging (**Figure 1D**). Nine months postopera-

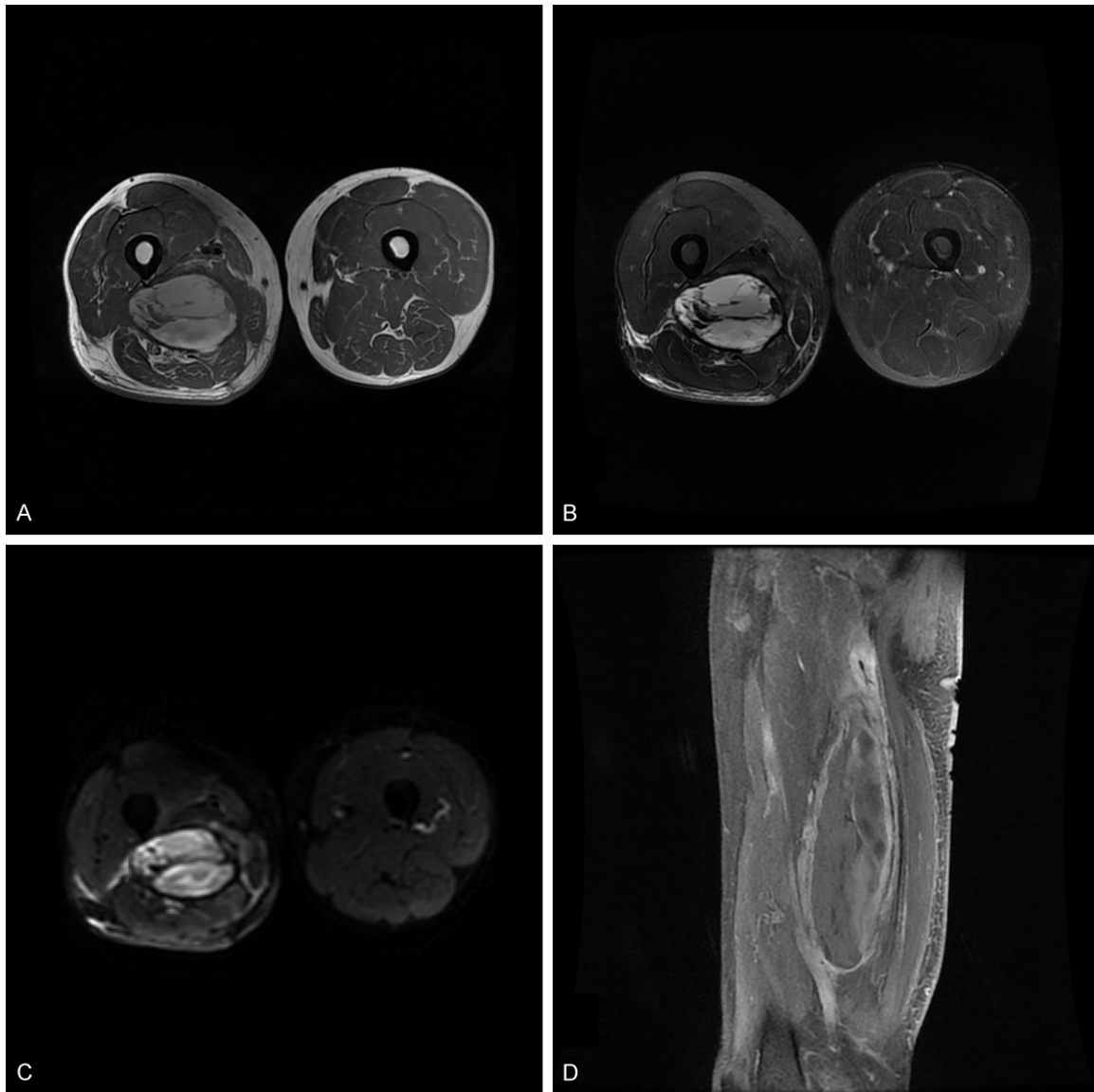


Figure 1. T1WI, T2WI, DWI and contrast enhanced imaging for the lesion forty days postoperatively. MRI scans show the local lesion with an inhomogeneously hyperintense on axial T1WI, T2WI and DWI (A-C). Sagittal contrast-enhanced imaging (D) shows an inhomogeneously hypointense mass in the posterior thigh.

tively, MRI scans showed the lesion with a homogeneously hypointense on T1WI (**Figure 2A**), and an inhomogeneously hyperintense on T2WI and DWI (**Figure 2B, 2C**). The contrast-enhanced imaging (**Figure 2D**) showed an inhomogeneously hypointense. Eighteen months postoperatively, MRI scans showed the lesion with a homogeneously hypointense on T1WI (**Figure 3A**), and a homogeneously hyperintense on T2WI and DWI (**Figure 3B, 3C**). The contrast-enhanced imaging (**Figure 3D**) showed a homogeneously hypointense. Specially, the volume of mass minished obviously in MRI con-

ventional sequences. Futher examination of thoracic CT scans showed no evidence of metastases.

The patient is currently disease-free.

Discussion

Complete surgical resection has been performed as a primary treatment for the IMT. Considering the aggressive nature of the tumor, additional radiotherapy was recommended as an adjuvant treatment. Radiotherapy is rarely discussed in the treatment of IMTs. It is neces-

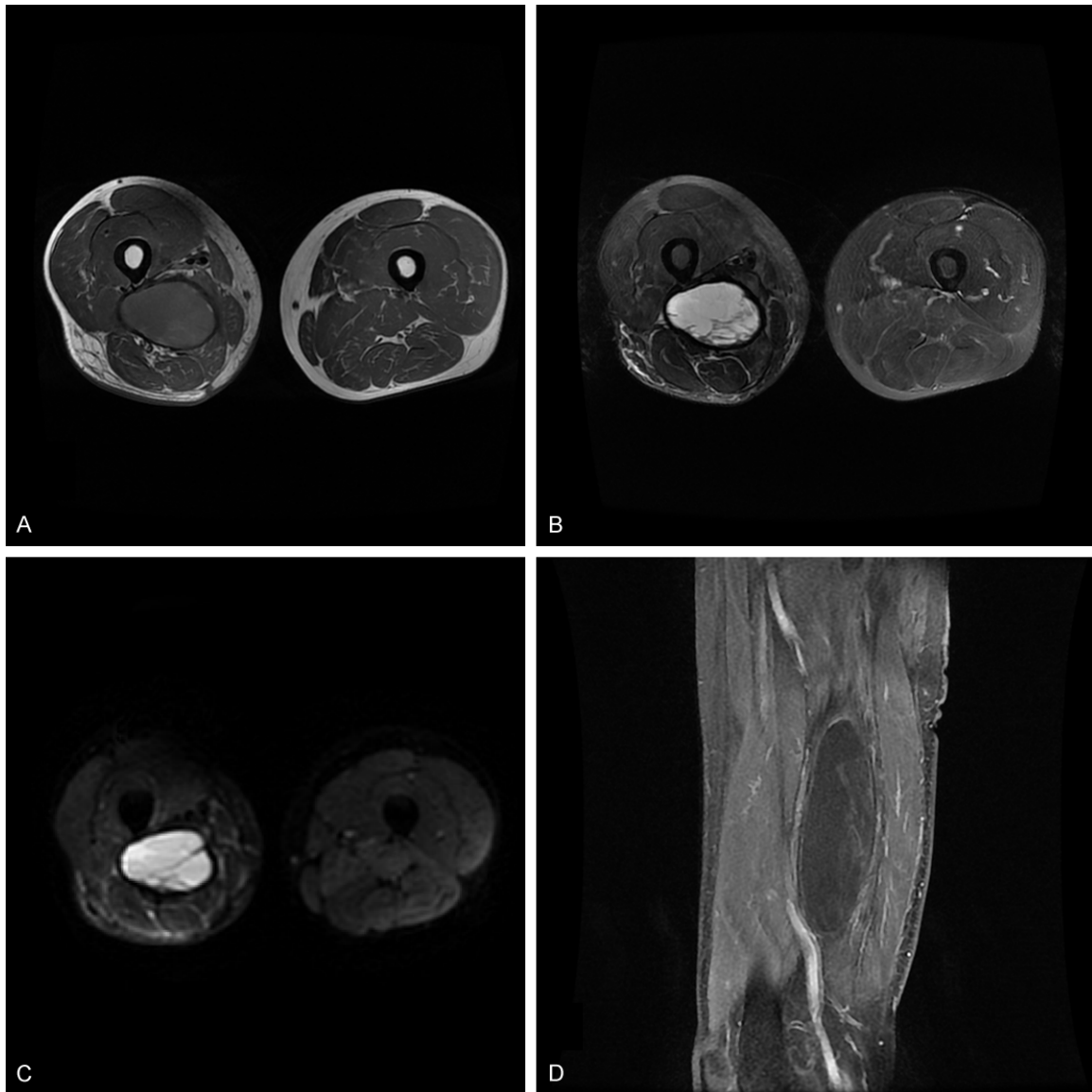


Figure 2. T1WI, T2WI, DWI and contrast enhanced imaging for the lesion nine months postoperatively. MRI scans show the local lesion with a homogeneously hypointense on T1WI (A), and an inhomogeneously hyperintense on T2WI and DWI (B, C). Sagittal contrast-enhanced imaging (D) shows an inhomogeneously hypointense mass in the posterior thigh.

sary to determine the appropriate treatment volume and radiation dosage. Based on nearly no macroscopically residual tumor after the operation in this case, we determined to perform the radiotherapy with a total dose of 50Gy (2Gy/F, 5F/week) and a local postoperative site irradiation [9]. The efficacy of radiotherapy and much effective irradiation plans for IMTs require further research.

IMTs are restless to metastasize to other sites of body and relapse after treatments, and even rarely transform into a true sarcoma [10, 11].

Considering distant metastasis could primarily turn to the lungs [3], we performed thoracic CT of the patient postoperatively to certify no metastatic site in lungs.

Radiologically, MRI is often used to assess the extent of soft tissue disease and the inherent structure of the lesion. In most reported cases of IMT, MRI shows hypointense or isointense on T2WI and homogeneous contrast enhancement due to a relative lack of mobile protons within fibrous lesions [12, 13]. In this case, postoperative MRI scans showed the lesion

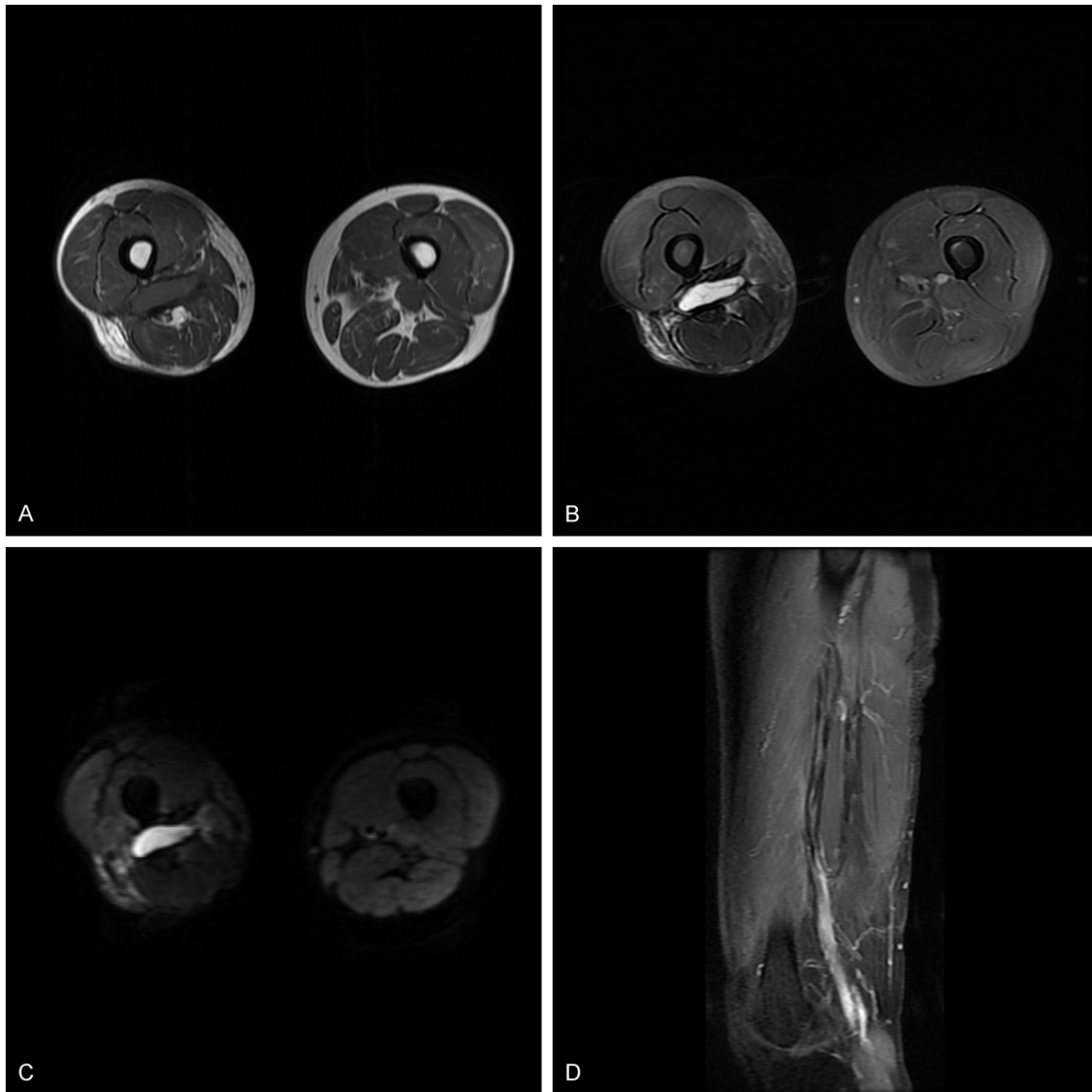


Figure 3. T1WI, T2WI, DWI and contrast enhanced imaging for the lesion eighteen months postoperatively. MRI scans show the local lesion with a homogeneously hypointense on T1WI (A), and a homogeneously hyperintense on T2WI and DWI (B, C). Sagittal contrast-enhanced imaging (D) shows a homogeneously hypointense. Specially, the volume of mass minishes obviously in the MRI conventional sequences.

with hyperintense on T2WI and hypointense on contrast enhanced imaging, so we don't consider the postoperative lesion as recurrence of IMT. In addition, based on hypointense on T1WI and hyperintense on DWI, we tend to regard the lesion as a cyst. Eighteen months after the operation, MRI scans still showed the lesion with a homogeneously hypointense on T1WI, and a homogeneously hyperintense on T2WI and DWI. The contrast-enhanced imaging still showed a homogeneously hypointense. What's more, the volume of mass minished obviously

in MRI conventional sequences. The above evidence makes the diagnosis of a cyst for the lesion more convinced, however, persistently close follow-up of the patient is requisite.

Conclusion

In summary, we have firstly reported an additional case of ALK-negative IMT of the lower extremities without bone involvement treated with surgical excision and radiotherapy. During two-year follow-up after the treatment, MRI scans including T1WI, T2WI, DWI and contrast

enhanced imaging revealed no local recurrence of IMT. Furthermore, based on MRI scans, the diagnosis of a cyst for postoperative mass is suggested.

Acknowledgements

Funded by Jiangsu Provincial Special Program of Medical Science (BL2012004); Supported by the National Natural Science Foundation of China (Grant No. 81401768); Supported by the Natural Science Foundation of Jiangsu Province (Grant No. BK20140289); Supported by the Specialized Research Fund for the Doctoral Program of Higher Education of China (Grant No. 20123201120018); Supported by China Postdoctoral Science Foundation on the 53rd general program (Grant No. 2013M531404).

Disclosure of conflict of interest

None.

Address correspondence to: Dr. Huilin Yang, Department of Orthopaedic Surgery, The First Affiliated Hospital of Soochow University, 188 Shizi Street, Suzhou 215006, Jiangsu, China. Tel: 0086-512-67780101; Fax: 0086-512-67780999; E-mail: hly-angsuzhou@gmail.com

References

- [1] Coffin CM, Watterson J, Priest JR, Dehner LP. Extrapulmonary inflammatory myofibroblastic tumor (inflammatory pseudotumor). A clinicopathologic and immunohistochemical study of 84 cases. *Am J Surg Pathol* 1995; 19: 859-872.
- [2] Coffin CM, Hornick JL, Fletcher CD. Inflammatory myofibroblastic tumor: comparison of clinicopathologic, histologic, and immunohistochemical features including ALK expression in atypical and aggressive cases. *Am J Surg Pathol* 2007; 31: 509-520.
- [3] Fletcher CD, Bridge JA, Hogendoorn PC. WHO Classification of Tumours of soft Tissue and Bone. Lyon: International Agency for Research on Cancer; 2013.
- [4] Hoover SV, Granston AS, Koch DF, Hudson TR. Plasma cell granuloma of the lung, response to radiation therapy: report of a single case. *Cancer* 1977; 39: 123-125.
- [5] Hakoziaki Y, Katou M, Nakagawa K, Shirahama T, Matsumoto T. Improvement of inflammatory pseudotumor of the liver after nonsteroidal anti-inflammatory agent therapy. *Am J Gastroenterol* 1993; 88: 1121-1122.
- [6] Alaggio R, Cecchetto G, Bisogno G, Gambini C, Calabro ML, Inserra A, Boldrini R, De Salvo GL, G d'Amore ES, Dall'igna P. Inflammatory myofibroblastic tumors in childhood: a report from the Italian Cooperative Group studies. *Cancer* 2010; 116: 216-226.
- [7] Debelenko LV, Arthur DC, Pack SD, Helman LJ, Schrupp DS, Tsokos M. Identification of CARS-ALK fusion in primary and metastatic lesions of an inflammatory myofibroblastic tumor. *Lab Invest* 2003; 83: 1255-1265.
- [8] Lin J, Liu H, Zhuang Y, Yang P, Zheng Y, Yang Y, Yang H. Inflammatory myofibroblastic tumor of the thigh without bone involvement: a case report. *World J Surg Oncol* 2014; 12: 208.
- [9] Cox JD, Ang KK. Radiation Oncology: Rationale, Technique, Results. Philadelphia, PA: Mosby; 2009.
- [10] He CY, Dong GH, Liu HG. Recurrent laryngeal inflammatory myofibroblastic tumor with positive anaplastic lymphoma kinase mimicking recurrent respiratory papillomatosis: a case report. *World J Surg Oncol* 2014; 12: 54.
- [11] Donner LR, Trompler RA, White RT. Progression of inflammatory myofibroblastic tumor (inflammatory pseudotumor) of soft tissue into sarcoma after several recurrences. *Hum Pathol* 1996; 27: 1095-1098.
- [12] Sasagawa Y, Akai T, Itou S, Iizuka H. Multiple intraosseous inflammatory myofibroblastic tumors presenting with an aggressive clinical course: case report. *Neurosurgery* 2011; 69: E1010-E1015, E1015-E1016.
- [13] Gasparotti R, Zanetti D, Bolzoni A, Gamba P, Morassi ML, Ungari M. Inflammatory myofibroblastic tumor of the temporal bone. *AJNR Am J Neuroradiol* 2003; 24: 2092-2096.