Human Pathology (2014) **45**, 2355–2358

ELSEVIER

Human PATHOLOGY

www.elsevier.com/locate/humpath

Case study

Multiple metastases from histologically benign intraarticular diffuse-type tenosynovial giant cell tumor: a case report☆,☆☆

Naofumi Asano MD[a,b,*], Akihiko Yoshida MD, PhD[c], Eisuke Kobayashi MD, PhD[a], Takehiko Yamaguchi MD, PhD[d], Akira Kawai MD, PhD[a]

[a]*Department of Musculoskeletal Oncology, National Cancer Center Hospital, Tokyo 104-0045, Japan*
[c]*Department of Pathology and Clinical Laboratories, National Cancer Center Hospital, Tokyo 104-0045, Japan*
[d]*Department of Pathology, Jichi Medical University, Tochigi 329-0498, Japan*
[b]*Department of Orthopedic Surgery, Keio University, Tokyo 160-8582, Japan*

Received 20 May 2014; accepted 17 June 2014

Keywords:
Diffuse-type tenosynovial giant cell tumor;
Intraarticular D-TGCT;
PVNS;
Metastasis;
Implantation phenomenon

Summary Diffuse-type tenosynovial giant cell tumor (D-TGCT) is a relatively rare mesenchymal tumor. It is a locally aggressive but virtually nonmetastasizing neoplasm and thus regarded as benign. Only a few D-TGCTs with benign histology have been reported to metastasize. We report an extremely rare case of benign D-TGCT in which multiple metastases developed 9 years after surgery for the primary tumor. The present case suggests that conventional D-TGCT has the potential to form distant metastases, albeit exceptionally rarely, and that this probable implantation phenomenon can be managed conservatively.

1. Introduction

Tenosynovial giant cell tumor (TGCT) is a mesenchymal tumor typically arising in close association with the synovial lining of the joint space. Localized-type TGCT, also known as giant cell tumor of the tendon sheath, is a well-demarcated lesion predominantly located in the synovium or tendon sheath. In contrast, diffuse-type TGCT (D-TGCT) is characterized by infiltrative growth and has a propensity for local recurrence in up to 50% of cases [1-4]. D-TGCT manifests as 2 forms—intraarticular and extraarticular—and historically, the intraarticular form has been known as pigmented villonodular synovitis (PVNS). Initially believed to be an inflammatory process, D-TGCT is currently recognized as a neoplasm genetically characterized by fusion genes involving *CSF1* at 1p13 encoding colony stimulating factor 1 [4,5]. Despite its locally aggressive nature, D-TGCT is unlikely to develop distant metastasis, unless associated with malignant transformation characterized by anaplastic cytology [1-4]. In this report, we present an extremely rare case of D-TGCT from which metastases developed in the lung, soft tissues, and lymph nodes despite its entirely benign histology.

☆ This study was presented in part at the 18th Annual Meeting of the Connective Tissue Oncology Society in Prague, Czech Republic, November 2012.
☆☆ Competing interests: No conflict of interest.
* Corresponding author at: Department of Musculoskeletal Oncology, National Cancer Center Hospital, 5-1-1 Tsukiji, Chuo-ku, Tokyo, 104-0045 Japan.
E-mail address: asanonaofumi@yahoo.co.jp (N. Asano).

http://dx.doi.org/10.1016/j.humpath.2014.06.025
0046-8177/

2. Case report

A 44-year-old Japanese man presented with a 7-month history of left knee swelling. Physical examination showed a large soft tissue mass in the lateral aspect of the left knee. Magnetic resonance imaging (MRI) demonstrated an ill-defined, heterogeneous mass measuring 9.8 cm, which was located around the fibular head and infiltrated the tibia and fibula. The tumor showed low intensity on both T1-weighted (Fig. 1A) and T2-weighted images (Fig. 1B), with peripheral contrast enhancement (Fig. 1C). After needle biopsy, the lesion was curetted. Histologically, the specimen showed diffuse proliferation of mononuclear cells admixed with histiocytes and multinucleated giant cells (Fig. 1D and E). Xanthomatous cells and hemosiderin deposition were focally conspicuous. Cleftlike spaces were occasionally present, and no necrosis was evident. The tumor cells lacked nuclear atypia and spindling. Mitoses were occasionally noted (up to 2 per 10 high-power fields), and they were not atypical. The tumor primarily involved the proximal tibiofibular joint and had infiltrated the adjacent bone and soft tissue structures. The findings were diagnostic of conventional intraarticular D-TGCT (PVNS).

Four years after the initial operation, local recurrence was detected by MRI as a 13-cm soft tissue mass. Above-knee amputation was performed because the tumor involved the neurovascular structures and bones. The resected tumor showed an identical histology to the primary and was diagnosed as a recurrent conventional intraarticular D-TGCT.

Four years after the amputation, the patient presented with a 3-week history of left back pain. Computed tomography (CT) revealed a 1.5-cm mass in the left lung (Fig. 2A), and the nodule showed a high standardized uptake value (8.61) on positron emission tomography. As a primary lung cancer was suspected, this lung nodule was resected by thoracoscopic surgery. Unexpectedly, however, it demonstrated the histology of D-TGCT, resembling the primary tumor in the knee (Fig. 2B and C). Mononuclear tumor cells were diffusely immunoreactive for clusterin (clone41D, 1:1000; Millipore, Temecula, CA) [6], which highlighted characteristic cytoplasmic dendritic processes (Fig. 2C, inset). The tumor was diagnosed as a pulmonary metastasis of histologically benign D-TGCT.

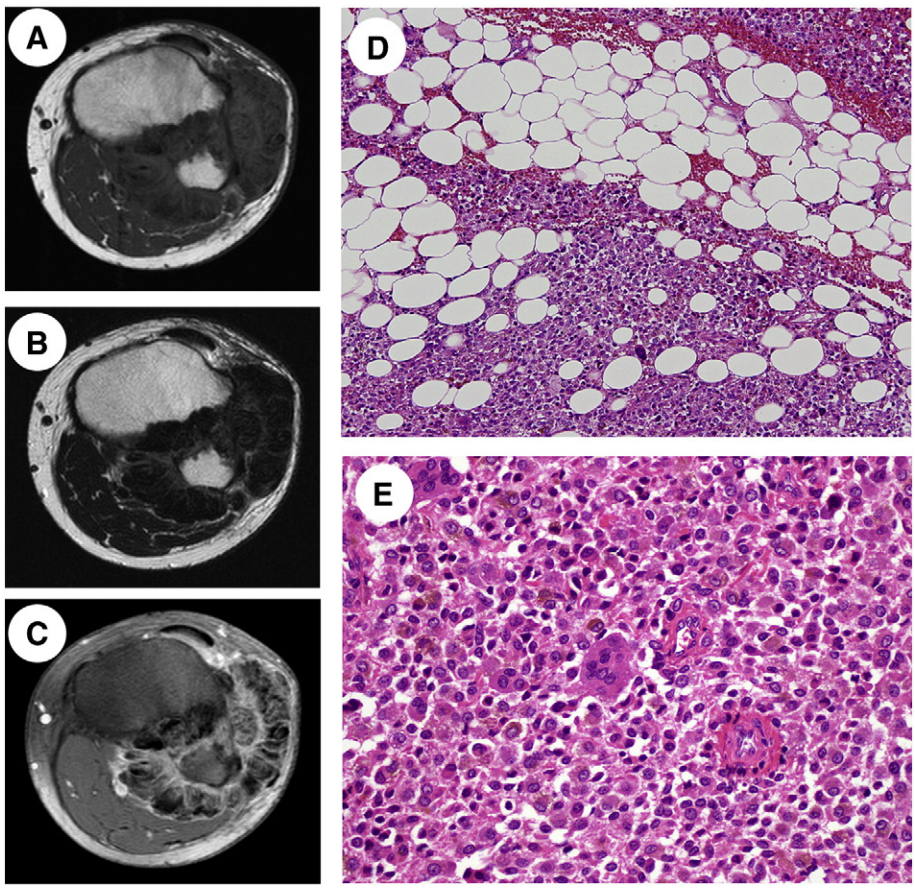

Fig. 1 MRI at presentation demonstrated a large ill-defined and heterogeneous mass surrounding the fibular head and infiltrating the tibia and fibula. The tumor showed low intensity on T1-weighted (A) and T2-weighted images (B), with peripheral contrast enhancement (C). D, The curetted specimen showed a diffuse TGCT consisting of diffusely infiltrating sheets of mononuclear cells (original magnification ×40). E, The tumor cells were nonatypical round cells with hemosiderin deposition and admixed with multinucleated giant cells (×400).