Radiology Case Reports 15 (2020) 1256–1260

Available online at www.sciencedirect.com

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journal homepage: www.elsevier.com/locate/radcr

Case Report

Case report of intra-articular synovial sarcoma in the hip joint☆,☆☆

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ARTICLE INFO

Article history:
Received 8 April 2020
Revised 14 May 2020
Accepted 14 May 2020

Keywords:
Intra-articular mass
Synovial sarcoma
Musculoskeletal tumor
MRI
Hip joint

ABSTRACT

Synovial sarcoma is a type of soft tissue sarcoma that commonly occurs around large joints. However, primary intra-articular synovial sarcoma is considered an extremely rare variant of synovial sarcoma. We present a case of synovial sarcoma arising from the hip joint in a 17-year-old female patient. Clinical, radiographic, magnetic resonance imaging and histopathology findings are described. The patient underwent neoadjuvant chemoradiation therapy followed by right hemipelvectomy, and there was no local recurrence in the subsequent follow ups. Unfortunately, the patient had pleural metastasis, and she was treated with palliative chemotherapy. To the best of our knowledge, there is only one published case report in the English literature on intra-articular synovial sarcoma from the hip joint.

Introduction

Synovial sarcoma is a relatively common soft tissue sarcoma that was first reported in 1893 and has variable degrees of differentiation [1,2]. It accounts for 2.5%-10% of soft tissue sarcomas [1,2]. Synovial sarcoma has been described in the literature to commonly occur near joints rather than intra-articular regions. These lesions are frequently located in the lower extremities, particularly around the knee joint [1,3,4]. Rarely, the tumor has an intra-articular origin, with such occurrences accounting for less than 5% of cases [5]. Synovial sarcomas are commonly observed in adolescents and young adults aged 15-40 years [6].

The imaging features suggestive of synovial sarcoma are characterized by the presence of periarticular, multiloculated or lobulated cystic masses with heterogeneous septal and/or peripheral nodular enhancement near a large joint. Calcification and bone erosions can be observed [1,5]. Synovial sarcoma is considered an intermediate- to high-

☆ Acknowledgment: No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this paper. No funds were received in support of this study.
☆☆ Conflicts of Interest: There are no conflicts of interest.
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https://doi.org/10.1016/j.radcr.2020.05.026

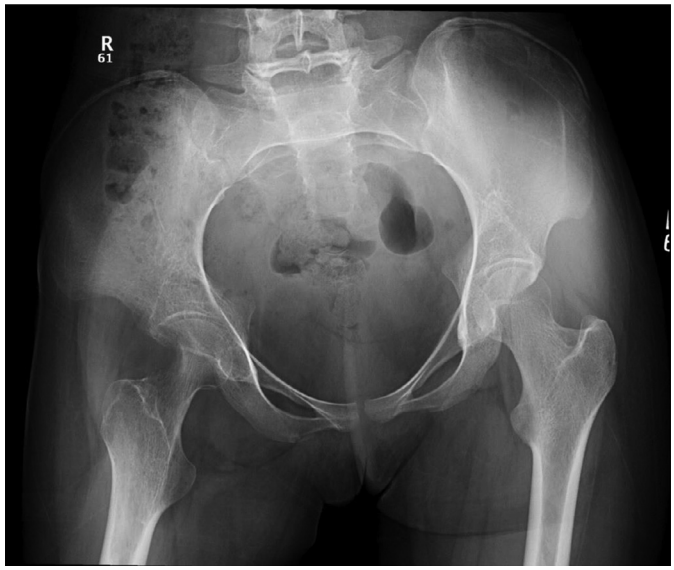

Fig. 1 – AP view of the pelvis showing permeation and erosion of the superior lateral aspect of the right femoral head and neck, associated with soft tissue swelling. Right ward tilting of the pelvis.

grade neoplasm with a high propensity for distant metastasis [1].

We present a rare case of intra-articular synovial sarcoma arising from the right hip joint that was confirmed by a pathology assessment in a 17-year-old girl.

Case Report

A 17-year-old female patient with no prior medical complaints presented with a progressive six-month history of right hip pain and limping. She did not have a history of trauma. The physical examination revealed a large, tender, firm mass in the right upper thigh causing pain to radiate throughout the right lower limb. The patient was able to walk, but she could not sit probably. No sensory deficits were observed, but there was a mild limitation in the hip flexion and extension range of motion in the right hip joint. No other lumps were identified elsewhere in the body.

The pelvic radiograph (Fig. 1) showed right hip regional osteopenia with permeation and erosion of the lateral aspect of the right femoral head and neck associated with significant soft tissue swelling. No identifiable soft tissue calcifications were seen. A right hip MRI scan showed an intra-articular, ill-defined, heterogeneous soft tissue mass in the right hip joint that extended inferomedially. The mass measured approximately 7 × 4 × 6 cm in the craniocaudal, transverse and anteroposterior dimensions, respectively. The intra-articular mass caused pressure erosions observed in the femoral head at the lateral aspect of the femoral head-neck junction. Extensive bone marrow edema was noted in the femoral head and neck and extending to the intertrochanteric region. Small joint effusion was noted. No pathological fractures were identified. The mass demonstrated heterogeneously high signal intensity on the T2-weighted images and isointense signal intensity on the T1-weighted images compared to those of the adjacent muscles. The images taken after gadolinium contrast administration showed heterogeneous and septal enhancement (Fig. 2).

Ultrasound-guided core needle biopsy was carried out for histopathology assessment. The anatomical pathology gross specimen revealed multiple soft, tiny fragments measuring 0.3 × 0.2 × 0.1 cm overall. The histopathological examination revealed a cellular monophasic spindle cell tumor with hyperchromatic nuclei and a small intervening stroma with staghorn-like blood vessels (Fig. 3). Fluorescence in situ hybridization showed gene rearrangement involving SYT gene, these findings are consistent with synovial sarcoma.

The initial whole-body positron emission tomography (PET) computed tomography (CT) scan and chest CT scan requested as part of the tumor work-up for staging showed no distant metastatic lesions. The case was discussed in the sarcoma board meeting, where neoadjuvant chemoradiation therapy was recommended before the surgery. The hip joint could not be salvaged, as the tumor had spread diffusely throughout the joint. Following radiation and chemotherapy, the patient underwent right hemipelvectomy. Fortunately, the right lower limb was saved successfully.

The chest CT taken at the 6-month follow-up showed 2 small, lower lobe lung nodules on the left side. The patient underwent left lower lobe metastactomy, and the biopsy confirmed the metastatic nature of these nodules. The patient presented with a left-sided pleural-based soft tissue mass 1 year later. The additional assessment with PET/CT demonstrated Fluorine-18-fluorodeoxyglucose (FDG)-avid uptake in the corresponding area. The patient was not fit for surgery, and she received 6 cycles of palliative care chemotherapy and showed a partial response in the recent chest CT scan. The patient is alive in a stable condition, with no local disease recur-

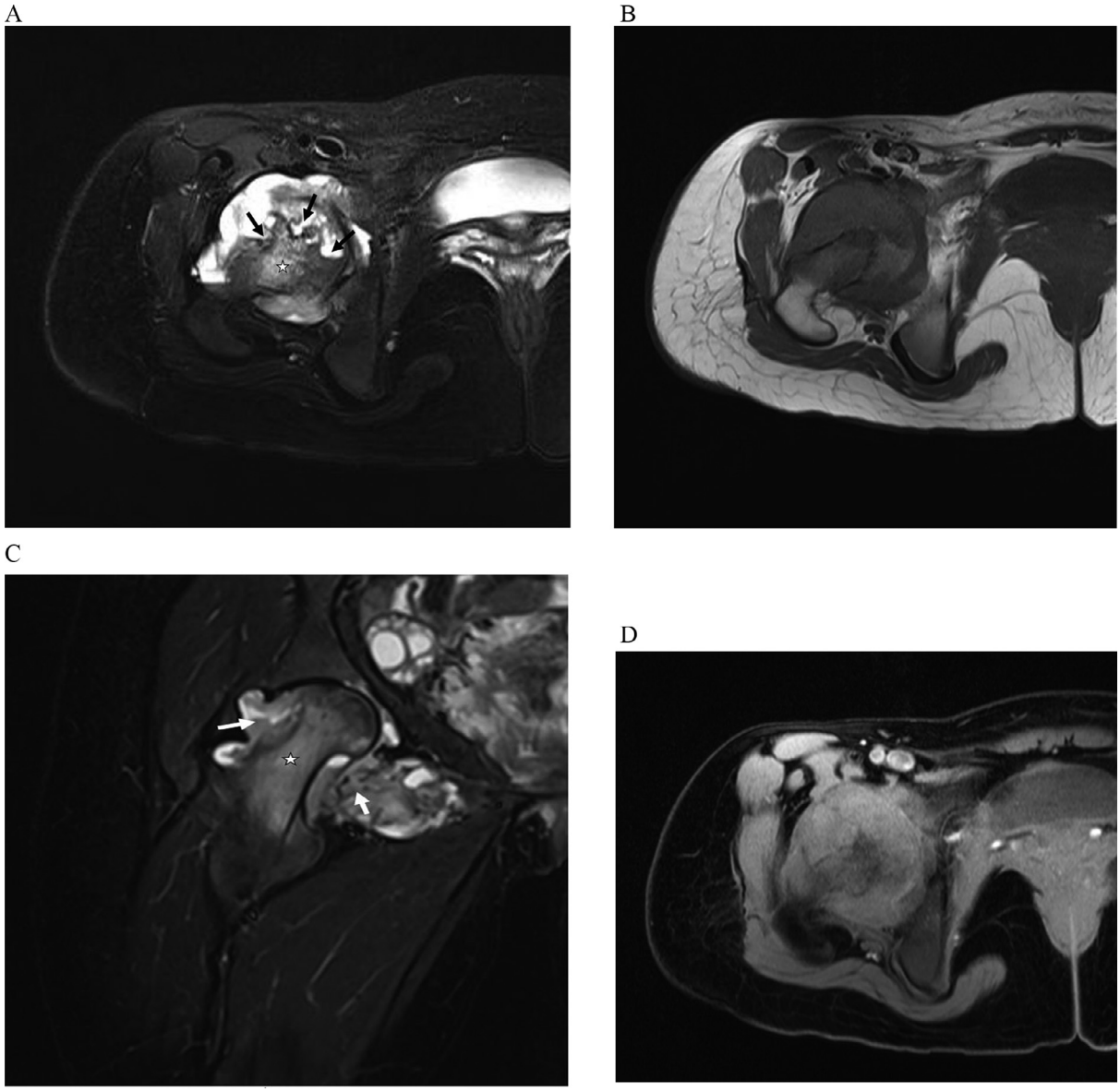

Fig. 2 – Multiple series of coronal T2 fat-saturated, axial T1, T2 fat-saturated and axial, sagittal and coronal T1 post-contrast images showing a heterogeneous soft tissue mass involving the right proximal femur and causing erosion of the femoral head and neck. The mass originated from the hip joint, extended inferomedially and displaced the obturator internus and adductor muscles (a), and it was measured to be approximately 7 x 4 x 6 cm. Reactive soft tissue effusion was observed. The mass demonstrates isointense signals on the T1 images (b) and moderately high signal intensity on the T2 sequence images (c), with heterogeneous avid enhancement in the post-gadolinium images (d, e and f). Multiple necrotic foci were noted.

rence at 3 years after hemipelvectomy. She is currently being followed up regularly with the oncology team.

Discussion

Synovial sarcoma is a type of primary mesenchymal soft tissue tumor that accounts for less than 10% of all primary soft tissue sarcomas [1,2,6]. Synovial sarcoma is found to develop around the joint in close relation to the tendons, tendon sheaths, and bursae [6]. Despite the nomenclature of synovial sarcoma, the tumor is usually extra-articular in location near the joints and usually aggressive in behavior [1,5]. Therefore, the term "synovial" is a misnomer, as the tumor does not originate from synovia. The term merely reflects its histopathological similarities with developing synovial tissue [7]. Nevertheless, primary intra-articular synovial sarcoma is extremely rare [2]. In the evaluation of 238 patients with synovial sarcoma, only 8 patients were found to have intra-

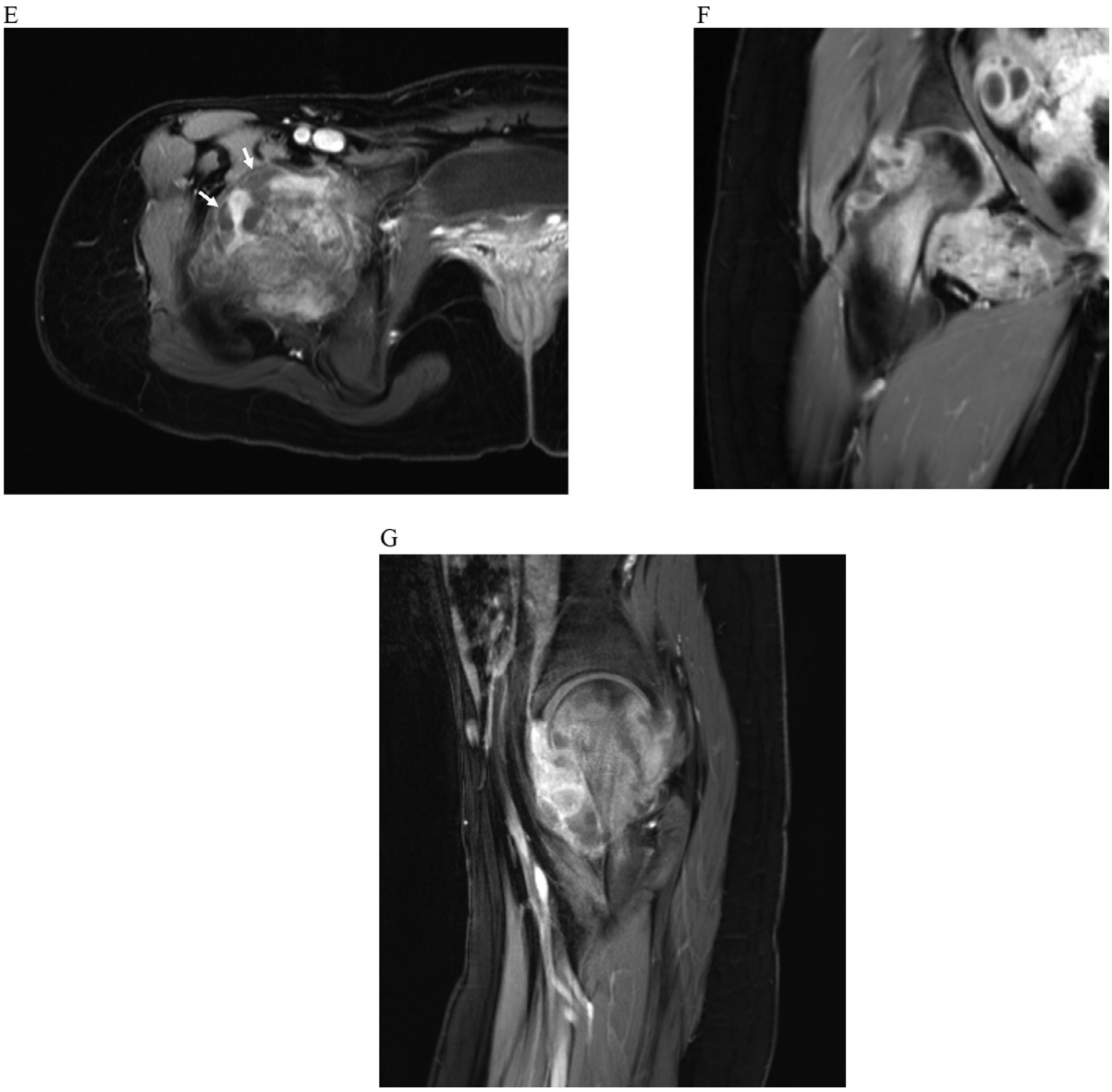

Fig. 2 – Continued

articular components, accounting for fewer than 5% of cases [5,7].

A review of published cases revealed that the most common location of intra-articular synovial sarcoma is the knee joint, followed by the elbow and ankle joints [1,3–5,8]. Only one case on intra-articular synovial sarcoma has been published [7]. In our case, the complete clinical course and radiological imaging findings were described.

Nonaggressive soft tissue masses known to involve the intra-articular joint space, such as pigmented villonodular synovitis and synovial osteochondromatosis, are more common and more frequently considered as differential diagnoses. Magnetic resonance imaging (MRI) is the imaging modality of choice to evaluate any soft tissue mass. It is used to evaluate the tumor extent and invasion in the surrounding tissue and is used for surgical planning [9].

From an imaging point of view, synovial sarcomas may have a nonspecific appearance, especially when they are smaller than 5 cm in size [5]. Different imaging features of synovial sarcoma in MRI studies have been described in the literature, such as a multilobulated appearance with intervening septa, as observed in our case. Other findings include the “bowl-of-fruit” appearance, fluid–fluid levels or the triple signal intensity sign [5]. In our case, benign intra-articular processes were considered in the differential diagnosis; however, the presence of extensive bone marrow edema from the erosive changes warranted further work up and imaging guided biopsy to exclude sinister lesions.

If the clinical presentation is not clear and the radiological investigation is nonspecific, tissue biopsy should be performed [8]. Once intra-articular synovial sarcoma is diag-

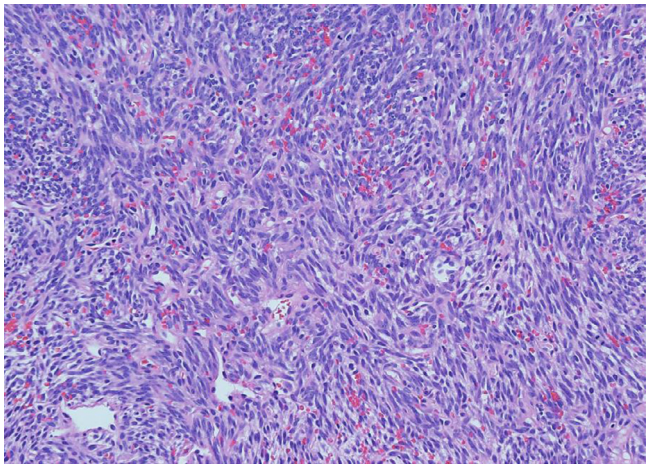

Fig. 3 – Monotonous atypical spindle cells in a fascicular pattern with focal staghorn-like blood vessels. (H&E: 200X).

nosed, the patient should be evaluated and followed up by a musculoskeletal oncologist [8].

The primary treatment of intra-articular synovial sarcoma is wide and free margins complete joint resection, retaining any part of a joint capsule will increase risk of tumor recurrence [6,10].

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