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General

Intra-Articular Nodular Fasciitis of the Knee, A Rare Clinical Presentation and Histological Diagnosis: A Case Report

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Introduction

Nodular fasciitis is a rare, benign soft tissue lesion that can prove to be difficult to diagnose, especially when intra-articular.

Case presentation

A 17-year-old female softball player presents with several year history of right knee pain and swelling initially attributed to patellar maltracking refractory to non-operative and operative management.

Management and Outcomes

Initial pathology suggested tenosynovial giant cell tumor; however, further tissue diagnosis revealed nodular fasciitis, which was eventually resected.

Conclusion

Intra-articular nodular fasciitis of the knee is rare and may easily be misdiagnosed due to its nonspecific clinical presentation. Careful histological examination can aid in diagnosis. Nodular fasciitis should be considered in the differential diagnoses for intra-articular lesions of the knee joint.

INTRODUCTION

Nodular fasciitis (NF) is a rare, benign soft tissue lesion of unknown etiology characterized by reactive myofibroblastic proliferation and an aggressive appearance on histology.[1, 2] NF typically presents as a rapidly growing, single subcutaneous mass occasionally associated with pain. The mass is most commonly found on the upper extremities, trunk, head, and neck.[3] NF affects both men and women equally and usually presents in patients between 10 to 50 years of age.[2] A previous history of trauma may also be present.

Although rare, NF may present in skeletal muscle and joints which can create a diagnostic challenge, as patients may have nonspecific symptoms of pain and reduced range of motion. While the intra-articular presentation of NF is rare, the knee appears to be the most common joint affected.[4] Previous reports of intra-articular NF have described differential diagnoses that include ganglion cysts, synovial chondromatosis, inflammatory arthritis, and tenosynovial giant cell tumor (TGCT), as well as malignancies like sarcoma.[1,4]

Diagnosis of NF requires a multidisciplinary approach, as imaging is often non-specific and immunohistochemistry is needed for confirmation.[3] Namely, *USP6* gene rearrangement typically confirms intra-articular NF in suspected cases.[5] With respect to treatment, surgical excision typically provides complete relief of symptoms, and furthermore, a review of 21 cases reported zero recurrences after excision.[4] Therefore, accurately diagnosing NF may prevent unnecessary diagnostic investigations and surgical interventions. Here, the authors present a rare case of intra-articular NF of the knee which had previously undergone surgical procedures for earlier presumed diagnoses. The patient was informed the data concerning the case would be submitted, and she provided consent.

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CASE PRESENTATION

ETHICAL CONSIDERATIONS

Consent to publish was obtained from the patient as well as the patient's parents. The consent form is uploaded for review.

A 17-year-old female softball player with a history of bilateral knee pain and instability status post bilateral tibial tubercle anteromedialization was seen at an outside hospital for several months of persistent right knee pain and swelling following her right tibial osteotomy. MRI at that time was suggestive of possible synovitis and diagnostic arthroscopy and partial synovectomy was performed. Postoperatively, the patient continued to have pain and swelling, refractory to non-steroidal anti-inflammatory drugs (NSAIDs), steroid injections, and aspirations, some of which were noted to be serosanguineous. Repeat MRI was unremarkable. The patient was also evaluated by rheumatology, who ruled out potential inflammatory etiologies of her persistent pain and swelling.

The patient subsequently presented to another sports medicine surgeon for a second opinion of persistent right patellar subluxation, pain, and swelling. She underwent right medial patellofemoral ligament (MPFL) reconstruction. Synovial tissue pathology suggested TGCT, and postoperatively she continued to have recurrent pain and swelling refractory to aspirations and injections.

MANAGEMENT AND OUTCOMES

The patient was then referred to our orthopedic oncology clinic. On exam, she was tender to palpation over the right tibial tubercle with mild effusion and tenderness to palpation of the medial joint line. Her right knee arc of motion ranged from 10° to 95° of flexion. New MRI revealed a 2.2 cm irregular signal intensity in the synovium with localized lobulated components in the infrapatellar fat pad, which was concerning for early TGCT **[Figure 1]**. Our institution's musculoskeletal pathologist reviewed the outside histology, which showed synovium with organizing fibrin, hemorrhage and hemosiderin deposition, granulation tissue and fibroblastic proliferation with giant cells, which is not diagnostic of TGCT. The patient underwent image guided biopsy and pathology showed myofibroblastic proliferation, synovial inflammation, and dense fibroconnective tissue **[Figure 2]**. Notably, the tissue stained positive for alpha-smooth muscle actin (SMA) and positive for *USP6* gene rearrangement by FISH, which is frequently identified in NF. The patient was discussed at our multidisciplinary sarcoma conference and was diagnosed with NF. The patient and her family agreed to proceed with open synovectomy.

For the surgical procedure, she was placed on the operating table and a medial parapatellar arthrotomy was performed to reveal a significant nodular mass in the anterior compartment of the knee. The mass was resected along with complete synovectomy without complications. Final pathology of the resected mass was consistent with NF.

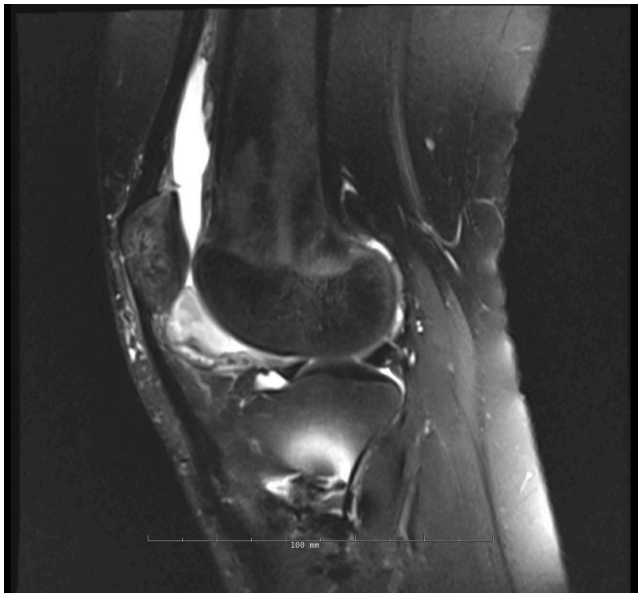

Figure 1. Irregular signal in synovium with T2 heterogeneously hyperintense lobulated component in the infrapatellar fat pad on sagittal magnetic resonance imaging of the knee.

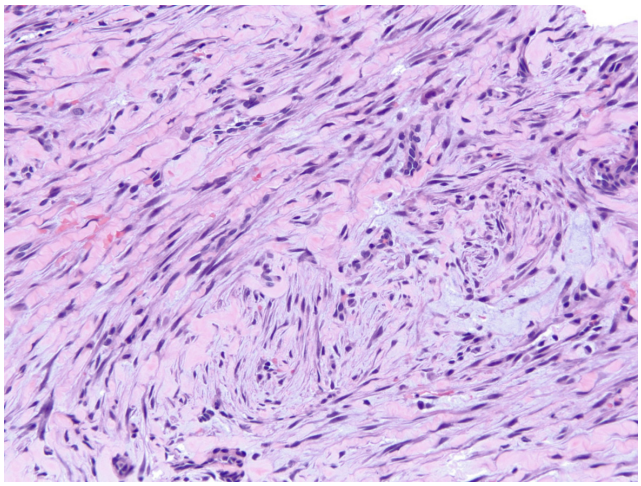

Figure 2. Myofibroblastic proliferation with storiform pattern and myxoid to collagenous stroma, suggestive of nodular fasciitis.

Post-operative MRI revealed resolution of the previously noted synovial signal; however, the patient continued to have intermittent discomfort and pain. Her right knee arc of motion was improved now ranging from 5° to 125° of flexion. She was seen by another sports surgeon at our institution who then referred her to Pain Management three months after surgery. She was diagnosed with neuropathic pain of her right knee and received several lumbar sympathetic nerve injections which provided significant relief of her right knee pain. Ultimately, she received a genicular nerve block to the right knee, which resolved her pain. The patient has continued to follow up with Pain Management now one year post-operatively and has improvement in her complaints of pain and instability.

DISCUSSION

Nodular fasciitis (NF) is a rare, pathologic soft tissue lesion first described by Konwaler et al. as a subcutaneous pseudosarcomatous fibromatosis.[6] Although rare, NF may occur in skeletal muscle and joints and cause nonspecific symptoms which creates a challenge in diagnosis. Our patient had a history of bilateral patellar instability and nonspecific pain for over two years despite prior non-operative and operative interventions. Of note, however, she did not have a palpable mass on physical examination, which is in contrast to eight previously reported cases of intra-articular NF in which a palpable mass was present on initial examination.

Diagnosis of NF requires a multidisciplinary approach as imaging is usually non-specific and confirmatory testing is done through immunohistochemical staining. MRI features of NF appear to be non-specific in the literature, with iso- to slight hyperintensity on T1 weighted imaging and hyperintensity on T2.[3] In this case, the localized mass appeared heterogeneously hyperintense on T2, isointense on T1, and avidly enhanced post-contrast administration.

Given the non-specific nature and wide differential diagnoses, biopsy or surgical excision is indicated for confirmatory diagnosis. This was especially evident in this patient's pre-operative course, as she required two different biopsies, with the second biopsy being evaluated by a fellowship-trained musculoskeletal pathologist. Additionally, two immunohistochemical stains, beta-catenin and alpha-smooth muscle actin (SMA), were used to aid in diagnosis. The cells showed negative nuclear staining for beta-catenin and positive staining for alpha-SMA. Previous case reports of intra-articular NF have all stained positive on immunohistochemistry for alpha-SMA.[4] *USP6* gene rearrangement has also been previously shown to confirm the histological impression of NF and was noted to be present in this case.[5] However, it must be noted that fibroma of tendon sheath, particularly the cellular variant, has overlapping histological features with NF and may also harbor USP6 gene rearrangement.[7]

According to the literature, the most common differential diagnoses of NF in the knee joint have been TGCT and synovial chondromatosis.[4,8-14] In the case presented, the patient was thought to have TGCT based on the initial pathology. A repeat biopsy and final pathology after surgical resection confirmed the diagnosis of NF.

Treatment of intra-articular NF focuses on surgical excision, which provides symptomatic relief with no risk of recurrence. Notably, the review by Wang et al. reported three cases of intra-articular NF, one shoulder and two knees, in which conservative treatment was sufficient for symptom management. Prior to surgical resection of the lesion, our patient had failed non-operative management with NSAIDs, steroid injections, and joint aspirations, none of which alleviated her pain. She had additionally undergone bilateral tibial tubercle anteromedialization several years prior to presentation at our institution for pain and instability, although her symptoms at that time may have preceded her development of intra-articular NF. The initial pathology suggestive of TGCT was collected during her MPFL reconstruction which did not provide significant clinical benefit to the patient. A history of trauma is often absent in reported cases of NF; however, Yamamoto et al. reported a soft tissue NF that developed at a surgical port site after robotic surgery for colorectal cancer.[15] Therefore, there may be some consideration of NF arising as a reactive inflammatory scarring process.

CONCLUSION

The case presented highlights the difficulty of diagnosing intra-articular NF, consistent with the existing literature. The final diagnosis was prefaced by a prolonged clinical course including failed non-operative and operative management. Several orthopedic subspecialists were involved in her care and the definitive diagnosis required histopathological evaluation by a fellowship-trained musculoskeletal pathologist. Although the patient continued to report intermittent pain after excision of the mass, her pain was thought to be neuropathic in nature when evaluated by Pain Management, and it eventually completely resolved after a genicular nerve block.

CLINICAL MESSAGE

Intra-articular NF is a rare pathology and remains difficult to diagnose. Multidisciplinary care can be beneficial in leading to a quicker diagnosis and preventing repeated surgical intervention.

CONSENT

The patient was informed the data concerning the case would be submitted, and she provided consent.

COMPETING INTERESTS

The authors declare that they have no competing interests.

DISCLOSURES

ATB: (BMJ Case Reports: Editorial or governing board; Clinical Orthopaedics and Related Research: Editorial or governing board; exparel/pacira: Stock or stock Options; Journal of Oncology Practice: Editorial or governing board; Journal of Surgical Oncology: ad hoc reviewer; Lancet - Oncology: Editorial or governing board; Musculoskeletal Tumor Society: Board or committee member; Onkos Surgical: Paid consultant; Pediatric Blood and Cancer: Editorial or governing board; Rare Tumors: Editorial or governing board; Rush Orthopedic Journal: Editorial or governing board; Swim Across America Cancer Research Grant: Research support; Orthopedic Reviews: Editorial or governing board)

AUTHORS' CONTRIBUTIONS

NPB, LL, and CG analyzed and interpreted the data, drafted, and edited the manuscript. ATB edited the manuscript for intellectually critical content and was responsible for the conception of the manuscript. All authors read and approved the manuscript for submission.

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PREVIOUS PRESENTATIONS

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