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CASE REPORT

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Simultaneous bifocal and asymptomatic intramuscular nodular fasciitis of the thigh: A case report

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Abstract

Nodular fasciitis (NF) is a rapid-growth benign that is misdiagnosed as sarcoma and leads to overtreatment. The spontaneous regression of NF is a possible phenomenon. "Wait and see" ideal is one of the treatment strategies of NF.

KEYWORDS

asymptomatic, bifocal, intramuscular, nodular fasciitis, spontaneous regression

1 | INTRODUCTION

Nodular fasciitis (NF) is a benign proliferation of fibroblasts and myofibroblasts that are misdiagnosed as sarcoma. The spontaneous regression of NF is a possible phenomenon. As a result, we believe that the occurrence of NF maybe higher than previously thought.

Nodular fasciitis (NF) is a pseudosarcomatous myofibroblastic benign of unknown causes. The reported incidence of NF is 11.3% of benign soft tissue tumors.[1] The majority of NF occurs in the subcutaneous tissue and underlying fascia, and about 5.9% of cases are intramuscular.[2] Clinical features present as a solitary nodule with rapid growth between 2 and 3 cm in size within a few weeks in addition to local tenderness. It mostly occurs in young to middle-aged adults between 20 and 40 years of age.[3,4] A magnetic resonance imaging (MRI) and histopathological features of NF are nonspecific and can be mistaken for soft tissue sarcoma.[5] To our knowledge, no previous study has reported NF with simultaneous bifocal and asymptomatic intramuscular lesions.

2 | CASE REPORT

A 21-year-old man was diagnosed with acute lymphoblastic leukemia (ALL) for19 years. The patient received a bone marrow transplantation after subsequent relapse. An MRI was performed to examine the osteonecrosis of the femoral head (ONFH), and two intramuscular soft tissue masses were incidentally found in different locations. The patient denied a history of trauma and family history of connective tissue tumors. He did not complaints of thigh pain, and we were unable to palpate the masses. The ovoid tumors were located at the adductor magnus and vastus intermedius of the thigh muscles, which exhibited homogeneous low T1-weighted and heterogeneous high T2-weighted signal intensity (Figures 1A and 2A). FDG PET/CT scans showed hypermetabolic soft tissue masses mimicking soft tissue sarcoma (Figure 3). The ALL relapse would account for a secondary malignant neoplasm, both masses (Figures 1A and 2A) underwent ultrasound-guided needle biopsy. The histological result showed rapidly growing lesions owing to their dense cellularity especially spindle-shaped myofibroblast and high mitotic activity.

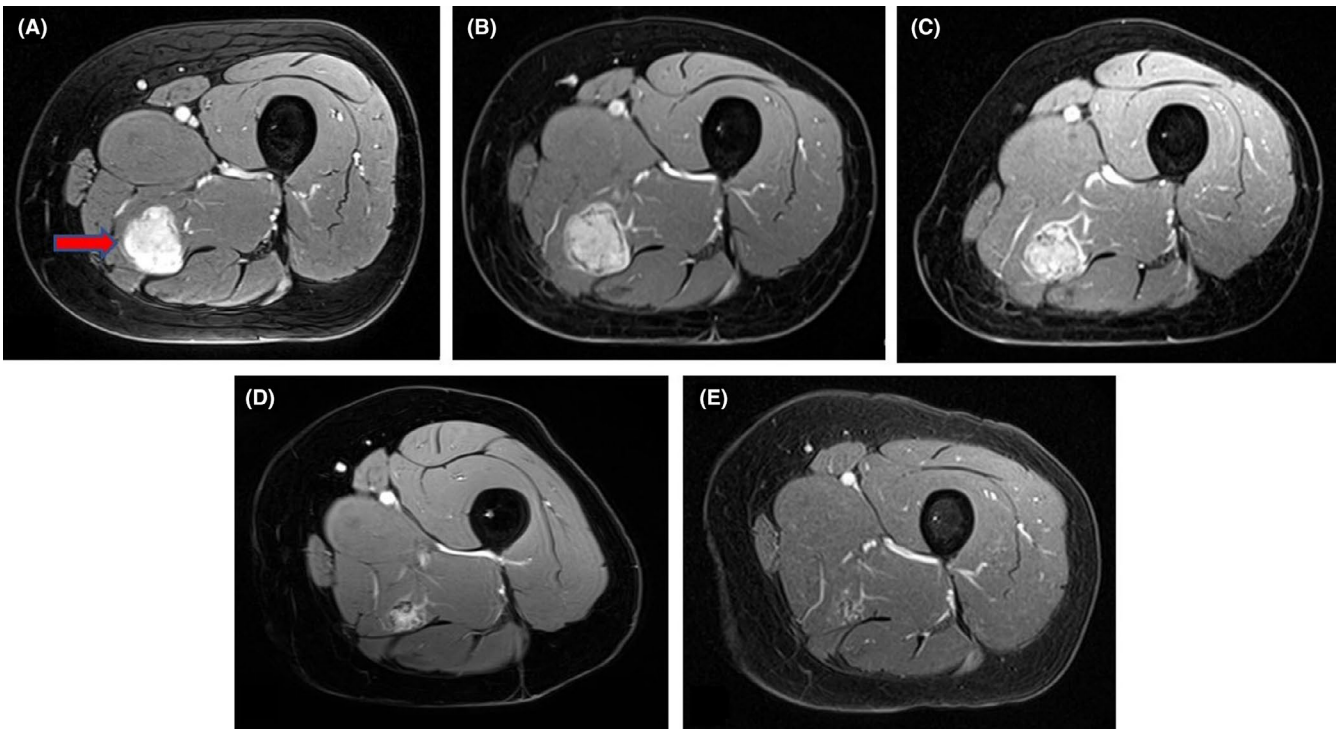

FIGURE 1 A, MRI showed an ovoid mass measuring 2.3 × 1.7 × 2.4 cm in size (red arrow) and heterogeneous high T2-weighted signal intensity at adductor magnus muscle at first visit. Images showed mass at B, 2 mo, C, 4 mo, D, 20 mo, and E, complete spontaneous regression at 30 mo

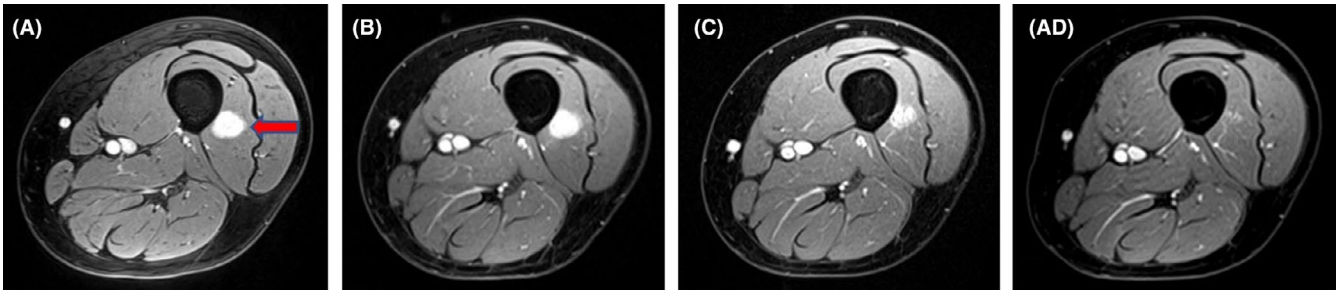

FIGURE 2 A, MRI showed an ovoid mass measuring 1.0 × 1.3 × 1.6 cm in size (red arrow) and heterogeneous high T2-weighted signal intensity at vastus intermedius muscle at first visit. Images showed mass at B, 2 mo, C, 4 mo, and D, complete spontaneous regression at 6 mo

The final pathological diagnosis was nodular fasciitis for both lesions (Figure 4). Immunohistochemically, the proliferating cells were positive for smooth muscle actin, and negative for desmin, HHF35, CD34, S100, and AE1/AE3.

Therefore, we followed up and observed the patient without any surgical intervention, and both tumors demonstrated spontaneous regression after six months and two and a half years, respectively (Figures 1A-E and 2A-D). "Wait and see" ideal is one of the treatment strategies for NF.

3 | DISCUSSION

Our case report reveals three combinations of uncommon features of nodular fasciitis: deep tumor site on an intramuscular region of the thigh, bifocal masses as shown by MRI, and asymptomatic lesion of more than 2 cm in diameter. Almost all nodular fasciitis demonstrate a self-limited myofibroblastic proliferation which presents with a single 2-3 cm mass that grows rapidly on superficial areas of the upper extremity, especially the forearm in individuals between 20 and 40 years of age.[3,4] Rarely, NF arises in uncommon sites such as the cutaneous, intermuscular, intramuscular, intra-articular, intraoral, intravascular, and intraneural regions. Asymptomatic lesions incidentally discovered by PET/CT scan have been reported in the literature, and most of these lesions were small hypermetabolic soft tissue nodules.[6,7] In the presented case, NF was found in two different sites. As these masses were deep-seated and uninvolved with neurovascular structures, they became asymptomatic.

MRI features of NF are nonspecific, usually present as an ovoid or spherical shape with broad fascial contact, and demonstrate homogenous low T1-weighted and heterogeneous intermediate to high T2-weighted signal intensity with surrounding edema and slightly inhomogeneous enhancement. It can be found with transcompartmental spread, osseous changes, or intra-articular extension which can be mistaken for a soft tissue sarcoma. None of the intramuscular lesions show transcompartmental spread due to being well-contained within the muscle belly.[5] In this case, an MRI showed two ovoid masses with homogeneous low T1-weighted and heterogeneous high T2-weighted signal intensity. The tumors were confined within the muscle with no transcompartmental spread. Chronologically, though a heterogeneous high T2-weighted signal intensity was observed at the initial visit and uniform MRI contrast enhancement was observed in the T1Gd image, the contrast effect gradually decreased, and the low-signal area expanded from the center.

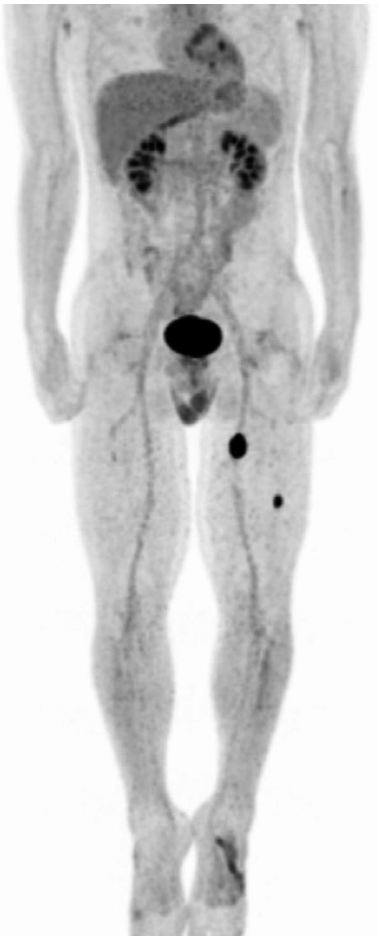

FIGURE 3 FDG PET/CT revealed two hypermetabolic soft tissue masses at the left thigh with a standardized uptake value (SUVmax) of 14

The following categories were created to classify our own cases that were diagnosed as NF using images and clinical findings: (a) localization: superficial or deep-seated, (b) symptoms: painful or indolent, (c) morphology of the mass: ovoid or spherical shape, and (d) period from onset to spontaneous regression: reduced size within a short period of time after initial diagnosis or reduced size after a fixed period of growth. According to these categories, each case is rich in variety, making their preoperative clinical diagnoses difficult. Our case was asymptomatic ovoid deep-seated masses with spontaneous regression after a fixed period of growth.

However, NF is not very difficult to diagnose if it develops with painful symptoms, occurs in the superficial fascia, and a flat mass can be palpated. On the other hand, clinical differentiation based on image findings is difficult compared with sarcomas when the lesion is painless, occurs in deep regions, and shows spherical tumor growth. The histopathological differential diagnosis between NF and sarcomas of low malignant may not be possible because of the limited availability of specimens by needle biopsy. Furthermore, the deep area of the anterior elbow might potentially contaminate or damage the vascular nerve with incision biopsy. In this case, the ALL patient exhibited simultaneously bifocal and asymptomatic masses with no specific sign on MRI and PET/CT scan. Diagnosis can easily be mistaken with acute lymphoblastic leukemia relapse, secondary malignant change, or primary soft tissue sarcoma. An incorrect diagnosis can result in improper treatment being administered.

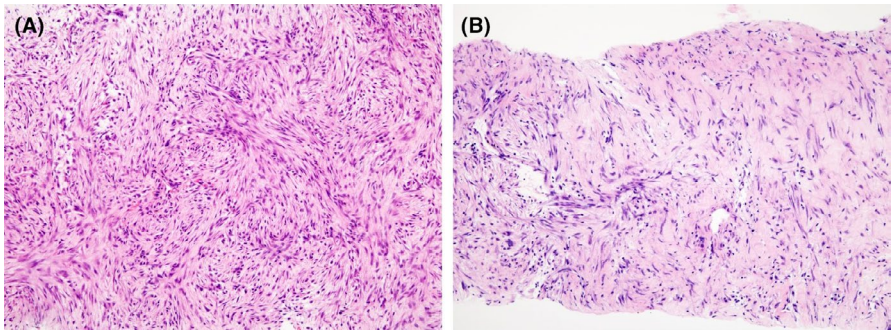

FIGURE 4 A histopathological examination showed fibroblastic and myofibroblastic proliferation with a loose fascicular to the storiform pattern in a mildly myxoid background. A, Adductor magnus muscle lesion; mild chronic inflammatory cell infiltration and small foci of extravasated erythrocytes. B, Vastus intermedius muscle lesion; the lesion was less cellular and more collagenous

Recently, NF can be confirmed by molecular diagnostic tests with a sensitivity of 93% and specificity of 100% using fluorescence in situ hybridization (FISH) for *ubiquitin specific proteases 6* (*USP6)*.[8] However, our case was diagnosed by histopathological needle biopsy since molecular-based tests are not available.

The standard treatment is complete excision in superficial or dermis NF. However, spontaneous regression of NF has been reported followed by biopsy or incomplete excision of the lesion. For example, the 20 cases of breast NF, an excisional biopsy was performed in 14 of 20 (70%) patients, while a "wait and see" approach was adopted in 4 of 20 (20%) individuals in which spontaneous resolution was observed.[9] An evidence based on NF, it is a self-limiting process and occasionally undergo spontaneous regression.[10,11] Few recurrences, disseminations, or deaths were reported. In cases of spontaneous resolution, the time lag between the diagnosis and the confirmed resolution ranges from 1 month to 2 years.[9] Some clinicians might regard complete excision as overtreatment and favor incisional or fine-needle biopsy, because these procedures may lead to tumor regression, less complication and morbidity. Nevertheless, a suspected nodular fasciitis lesion should be regarded as a potentially malignant tumor (sarcoma) until the final histologic diagnosis is made.[12] There is a report of cranial fasciitis, a rare variant of nodular fasciitis in children, in which rapid tumor growth occurred after fine-needle biopsy.[13] Above reason, we followed up the patient for two and a half years and the mass showed complete spontaneous regression on MRI.

4 | CONCLUSION

If NF develops as a painless and impalpable mass, the patient may not notice its presence. However, as in our present case, the tumor may not be diagnosed as NF and may lead to its disappearance. Thus, NF cases that are diagnosed by health facilities may only be a fraction of the total number of cases.

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Published with written consent of the patient

CONFLICT OF INTEREST

None declared

AUTHOR CONTRIBUTIONS

SS: involved in conducting the literature review and writing the manuscript. SM: involved in idea/concept, collected the case information and revised the final manuscript. KA, TT, KH, YF, YM, and MS: involved in the patient care team. KY: reviewed the pathology, provided histological slides and described for each slide. All authors approved the submitted content

CONSENT

Written informed consent was obtained from the patient for anonymized information to be published in this article.

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