Desmoplastic fibroma of the distal tibia: A case report of a minimally invasive histological diagnosis

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Abstract. Desmoplastic fibroma (DF) is a benign, rare fibroblastic intraosseous neoplasm histologically resembling a desmoid soft tissue tumor. Although classified as benign, DF frequently exhibits an aggressive behavior, has a moderate-to-high recurrence rate, and often causes pathological fractures and extensive bone destruction. This case report presents an incidentally detected DF of the tibia, which was diagnosed using a minimally invasive approach. A 36-year-old African female patient was referred to the Department of Diagnostic Imaging of Arcispedale Santa Maria Nuova-IRCCS (Reggio Emilia, Italy), to be examined by a computed tomography scan on an outpatient basis, after an x-ray examination of the tibia, which was performed after an injury to exclude the presence of a fracture, revealed a hyperlucency of unknown origin. The aim of this study was to discuss the clinical, histological, immunohistochemical and radiographic characteristics of this rare neoplasm, with a focus on image-guided bone biopsy.

Introduction

DF of the bone is an extremely rare, lytic, locally aggressive but non-metastatic primary bone tumor. Histologically, DF consists of wavy fibroblasts and abundant collagenous tissue, bears a close resemblance to a desmoid tumor of the soft tissues and exhibits a characteristic production of collagen fibers (1). DF may involve any bone, but is most often found in the mandible (22%) and less frequently in the femur (15%), pelvic bones (13%), radius (12%) and tibia (9%) (2,3).

The incidence of DF is estimated to be 0.06% among all bone tumors and 0.3% among benign bone tumors (1,3-5). DF of the bone most often occurs during the first three decades of life and exhibits no gender predilection (6). The most common symptoms include pain and swelling, and 12% of the patients present with pathological fractures. However, a number of patients may be asymptomatic; thus, the tumor is often incidentally identified (7).

On radiographs, the lesion is lytic, occasionally with poorly defined borders, while on computed tomography (CT) it is mildly hyperdense, with thin sclerotic margins, with or without destruction of the contiguous cortex, and exhibits low signal intensity on T2-weighted magnetic resonance imaging (MRI). The diagnosis of DF is predominantly based on pathological examination. The recurrence rates in patients treated with and without resection are 17 and 55-72%, respectively (7).

This disease was first identified by Jaffe in 1958 (8). To date, ~200 cases of desmoplastic fibroma of the bone have been reported in the literature; of these, 13 have been reported in the femur (9-20), with only 2 in the proximal femur (9,14) and the remaining in the distal femur (10-13,15-18,20,21).

Image-guided percutaneous biopsy is becoming an increasingly accepted modality for initial diagnosis in the majority of musculoskeletal tumors. However, despite its well-documented specificity and sensitivity (22,23), there has been continued reluctance on the part of orthopedic oncologists to accept this technique as the modality of choice for initial biopsy in primary bone tumors. Detractors of percutaneous needle biopsy suggest that sufficient material cannot be obtained to ensure an accurate diagnosis, particularly in patients with sclerotic bone lesions (24). Historically, all biopsies to date were performed via an open surgical biopsy, which remains the gold standard for a proportion of physicians. The reported accuracy of an open surgical biopsy, whether incisional or excisional, is 98% (23,25), whereas needle biopsy under CT guidance has a reported accuracy estimated between 78 and 98.4% (26).

The needle pathway is determined on the basis of the expected definitive treatment, in order to avoid intercompartmental contamination, minimize the amount of biopsy tract to be removed at the time of definitive surgery in case of malignancy, and avoid neurovascular structures. The choice of CT vs. fluoroscopy is generally determined on the basis of three factors, namely size, location and attenuation (27).

To the best of our knowledge, this study presents the only published case of minimally invasive diagnosis of DF of the bone. Written informed consent was obtained from the patient for the publication of the case details.

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Case report

In August 2015, a 36-year-old African female patient presented to the Emergency Room of Arcispedale Santa Maria Nuova-IRCCS (Reggio Emilia, Italy), which is a medium-sized general hospital, complaining of pain in the rib cage, knees and lower thighs following a mild trauma she had suffered a few days earlier after a fall, for which she had not been hospitalized. There were no evident abnormalities on physical examination, with the exception of pain on percussion. Radiography showed a well-defined, eccentric, low-density lesion in the distal metaphysis of the left tibia (Fig. 1), adherent to the external cortex. The contiguous cortex was apparently not eroded, but minimally thinned due to compression by the expanding tumor. The margins were intact and sclerotic. The initial diagnosis on admission, based on radiological evidence, was bone fibroma. The patient was advised on an outpatient basis to undergo a magnetic resonance imaging (MRI) scan and a subsequent orthopedic evaluation.

Due to the X-ray and bone MRI results, the orthopedic surgeon suggested an image-guided bone biopsy.

After a CT scan confirmed the location and imaging characteristics of the lesion (solid, well-circumscribed, no cortex erosions), local anesthesia with lidocaine was administered, a 17-gauge bone biopsy needle (Osteo-Site Ratchet; Cook Medical, Limerick, Ireland) was inserted, passing through the anterolateral face of the skin overlying the tibia, the tibialis anterior muscle and, finally, the cortex, into the core of the lesion (Fig. 2) and a single specimen was extracted. Since no peri-procedural complications occurred, the patient was discharged after a few hours in good health and without the need for oral pain medication, with the recommendation to refer to her general practitioner in case of any discomfort.

Histological examination of the bone specimen revealed focal peripheral proliferation of spindle-shaped cells resembling fibroblasts, showing strong reactivity for anti-vimentin antibody; the appearance was in accordance with a fibroblastic desmoid tumor of the bone.

Discussion

Percutaneous needle bone biopsy is a safe and accurate method (22,25,27-29) for obtaining a tissue diagnosis. In general, better results are obtained with lesions of the extremities or pelvis compared with those in the spine (30). Percutaneous needle biopsies have a very low complication rate (1.1%), whereas open biopsy has a complication rate of ≤16% (31).

A serious complication of percutaneous biopsy is the risk of seeding malignant cells along the needle track, particularly if the lesion is a sarcoma, which would necessitate resection of the needle track en bloc with the tumor in limb-sparing reconstructive surgery (32). Thus, selecting the appropriate needle path is critical for limb-salvaging procedures. Fine-needle aspiration (FNA) may differentiate between a metastasis and a benign lesion; however, core biopsy is superior to FNA in determining cell type and tumor grade, which is necessary for the diagnosis of primary bone tumor. In our experience, of the various options that are available for bone biopsy (33), the Osteo-Site Ratchet (Cook Medical) 13-gauge coaxial bone biopsy system is effective in obtaining tissue from sclerotic lesions where normal bone cortex needs to be traversed. In general, all bone biopsies are performed under local anesthesia; however, conscious sedation may be required in children or uncooperative patients. An adequate biopsy sample is considered to be that providing adequate abnormal tissue for the pathologist to reliably make a diagnosis. We have found that, despite the low incidence of DF, a single long core of abnormal tissue obtained with a 13-gauge needle is sufficient.

In conclusion, this study reported the case of a 36-year-old African female patient with a DF of the bone incidentally detected by an X-ray and diagnosed by means of percutaneous CT-guided bone biopsy. This case differs from other published studies due to the minimally invasive, preoperative, image-guided histological diagnosis. This type of diagnosis...
is commonly made based on a resection sample obtained through incisional biopsy under general anesthesia. The results of image-guided biopsy in terms of cost-to-benefit ratio are similar to those of open biopsy, with reduced comparable morbidity.

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