

Reconstruction of Tibial Nonossifying Fibroma Accompanying with Undetected Ewing's Sarcoma by Ilizarov Method

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The clinical behavior of nonossifying fibroma (NOF) appears to be a benign lesion without recurrence or canceration.^[1] Ewing's sarcoma (ES) is usually regarded as a differential diagnosis of NOF.^[2] There has not been any report on NOF and ES appearing in the same position simultaneously. Here, we first report a case that the simultaneous onset of two entities was mistaken for the development of NOF into ES.

A 17-year-old boy referred to hospital in October 2010 for a durative ache in his right knee after an injury during football training. Initial radiographs revealed a low-density area with sclerotic margin on the proximal part of the right tibia. Computed tomography (CT) scan showed a radiolucent, eccentric, and cortically based lesion in the posterolateral metaphysis of the right tibia. Then, the patient underwent magnetic resonance imaging (MRI), in which a lesion revealed long mixed signs on T1-weighted imaging (T1WI) and short signs on T2WI accompanied with bone contusion around it. These reports suggested that the eccentric area could be an osteoid osteoma or a NOF. A percutaneous biopsy of the right proximal tibia lesion was performed, and then NOF was pathologically confirmed. Hence, curettage and bone grafting were performed.

However, no more than 1 month later, wound infection and skin necrosis with progressive swelling were observed. An asymptomatic 4 cm magnitude soft tissue mass appeared on the same area. According to the report of MRI, it was considered to be abscess and osteomyelitis. Surgical incision and drainage were performed and pathologic result was proved to be ES. Chemotherapy was accepted after the surgery. However, tumor was growing back in November 2011, and radiofrequency ablation was performed. Then, the patient underwent about nine operations from 2010 to 2015 because of wound fester and osteomyelitis (multi-resistant

bacteria, multidrug-resistant organisms). In May 2015, ipsilateral dual-fibular reconstruction of the right knee was performed, which was unsuccessful. Then, the patient was admitted into our hospital and received limb-lengthening treatment by Ilizarov external fixator. During the final follow-up visit after 12 months, we did not find any signal of local recurrence or distant metastasis.

Images and history were studied. On the sagittal position of the right tibia radiography, an ill-defined decreased bone density lesion at the front of the lucent lesion appeared simultaneously, which was missed initially. On the initial MRI images, the corresponding ill-defined lesion showed a low signal on T1WI and an equal-high signal on T2WI with Codman's triangle reaction, which did not appear on the plain radiographs, thereby leading to a missed diagnosis of ES [Figure 1].

Metaphysical fibrous defects, also called fibrous cortical defects (FCD), are the asymptomatic, nonneoplastic and self-healing tumors of bone, which occur commonly in the metaphysis of long bone, especially in the lower extremities. NOF is known as bigger FCD that is classified as >30 mm.^[3] Radiologically, NOF appears as a solitary, eccentric, radiolucent, and cortically based lesion with a thin osseous border, which can be easily distinguished. According to statistics, <2% ES may represent a benign radiographic

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Figure 1: Anteroposterior view of knee reveals an increased bone density area, whereas two eccentric lesions are shown in the lateral (a). The lesion (arrow) is just the initially missed diagnosis. Nonossifying fibroma is clearly shown in computed tomography scan without trabecular abnormality (b). In the coronal T1-weighted imaging, Ewing's sarcoma appears as bone contusion with signal slightly lower than fat. The well-defined Ewing's sarcoma (arrow) in low signal is represented on the sagittal T1-weighted imaging at the front of nonossifying fibroma clearly. On proton density sequence, the mass (arrow) with Codman's triangle reaction is represented obviously in the sagittal and axial position (c).

appearance, which leads to a delay in diagnosis. Therefore, ES is usually regarded as a differential diagnosis of NOF.^[2,4] Image study is critical to the diagnosis of the two diseases. Radiographs are the first-line choice for NOF, whereas the plain film of ES is lack of characteristic features. MRI is suitable to identify the two diseases.^[5] CT scan is not contributory.

In our case, the patient's CT was not representing any abnormalities in trabecular bone, but a lucent NOF lesion with a well-defined sclerotic margin [Figure 1b]. It was more likely to make a benign lesion diagnosis according to CT scan. For the plain radiography, there were two eccentric lesions in a well-defined increased bone density area [Figure 1a]. The radiographer suggested that the patient should be followed up by MRI. Actually, the patient had undergone MRI twice in two different hospitals. The first one diagnosed that it was a benign tumor with bone contusion around it. The second one suggested that the "bone contusion" may

be a malignant bone tumor. From the sagittal position, there was a cortically based lesion with clear osseous margin at the posterior part of tibia with a mixed high signal on proton density (PD) sequence and a low signal on T1WI at the front of it [Figure 1c]. However, the depicted "bone contusion" with a clear margin at the front of NOF was ES. Here, we make a plausible inference that the surgeon only focused on the NOF lesion and mistook the well-defined ES for 'bone contusion'. And, because of the rigid sclerosis margin of NOF during the operation, ES had not been discovered postoperatively by pathology till the second time.

In conclusion, ES is rare but has a significant overlap with nonneoplastic entities. It is important to bear in mind that despite the rarity of this neoplasm, a meticulous attention should be kept to every detail. This is the first case presenting ES together with NOF, which showed a binary entity in one place. This may alert radiologist and clinicians to the diagnosis of such malignant tumor pattern.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the from, the patient and his guardians has given their consent for his images and other clinical information to be reported in the journal. The patient and his guardians understand that his name and initials will not be published and due efforts will be made to conceal her identity; however, anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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