

Case Report

Intraosseous lipoma of the tibia

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ABSTRACT

ABSTRAK

Introduction: Intraosseous lipoma is a rare benign bone tumor originating from proliferative mature lipocytes. The incidence of intraosseous lipoma is considered to be less than 0.1 % of all primary bone tumors. In recent years, an increasing number of cases of this disease have been reported, and the real incidence of the disease seems higher than the previously recognized. With the advancement of computed tomography (CT) and magnetic resonance imaging (MRI), the identification of intralesional fat is enabled, as well as dystrophic calcification and cyst formation.

Methods: We report one case of patient with intraosseous lipoma of tibia diagnosed by biopsy from curettage of the lesion. The patient underwent curettage, bone graft and internal fixation with plate and screws. Histological examination of the specimen revealed a chronic osteomyelitis with no marked anomalies. The patient was discussed in the Clinical Pathological Conference (CPC).

Results: Initially, from the CPC, the patient was diagnosed with chronic osteomyelitis of the left tibia. The histological examination was then re-reviewed by performing deep section of the preparation and the result was lipoma of the bone. One year after the surgery, the oncologic outcome was evaluated, there were no local recurrence or infection detected. The orthopaedic outcomes showed that the graft had been united, but the plate was still needed to be retained and the screw needed to be fixed because the patient decided to remove the implant when she had her ligament operated. The functional outcomes showed that the patient could walk and had full range of motion.

Conclusion: Intraosseous lipoma is a rare benign bone lesion that is difficult to diagnose with plain film imaging alone. However, CT and MRI can reveal intraosseous lipoma accurately. In spite of this, the diagnosis of intraosseous lipoma still need to be confirmed by the collaboration of orthopaedics, radiologist and pathologist in a clinical pathological conference.

Pendahuluan: Lipoma intraosseus adalah tumor jinak langka yang berasal dari proliferasi liposit matur. Kasus kejadian lipoma intraosseus diperkirakan kurang dari 0,1 % dari keseluruhan tumor tulang. Dalam beberapa tahun terakhir, peningkatan jumlah kasus telah dilaporkan, dan kejadian nyata dari penyakit ini diperkirakan meningkat dari apa yang telah diketahui. Perkembangan *computed tomography* (CT) dan *magnetic resonance imaging* (MRI) memungkinkan identifikasi lemak intralesi, demikian pula dalam identifikasi kalsifikasi distrofi dan pembentukan kista.

Metode: Kami melaporkan satu pasien dengan lipoma intraosseus pada tibia yang didiagnosis dari biopsi kuretase pada lesi. Pasien menjalani kuretase, *bone graft*, dan fiksasi internal dengan *plate and screws*. Pemeriksaan histologi dari spesimen bedah menunjukkan osteomielitis kronik tanpa anomali. Pasien kemudian didiskusikan pada *Clinical Pathological Conference* (CPC).

Hasil: Pada mulanya, dari hasil CPC, osteomielitis kronik ditemukan pada tibia kiri. Lalu, pemeriksaan histologis diulang menggunakan sayatan dalam dan hasil menunjukkan adanya lipoma tulang. Satu tahun setelah operasi, luaran onkologis dievaluasi dan tidak ditemukan adanya rekurens atau infeksi. Luaran ortopedi menunjukkan graft telah menyatu namun masih perlu dipertahankan dengan *plate and screw* dikarenakan pasien menjalani pengangkatan implan saat operasi ligament. Luaran fungsional menunjukkan pasien dapat berjalan dan memiliki *full range of motion*.

Kesimpulan: Lipoma intraosseus adalah tumor jinak langka yang sulit didiagnosis menggunakan pemeriksaan foto polos saja. Penentuan lipoma intraosseus melalui CT atau MRI dapat dilakukan secara akurat. Namun demikian, penegakan diagnosis lipoma intraosseus masih membutuhkan kolaborasi antara dokter bedah ortopedi, radiologi, dan patologi dalam clinical pathological conference.

Keywords: intraosseous lipoma, rare, radiological diagnostic, need clinical pathological conference.

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INTRODUCTION

Intraosseous lipoma is a rare benign bone tumor originating from proliferative mature lipocytes^{1,2}. Although bone marrow contains abundant adipose tissue, intraosseous lipomas are rarely found³. The incidence of intraosseous lipoma was thought to be less than 0.1 % of all primary bone tumor³. In recent years, an increasing number of cases of this disease have been reported, and the real incidence of the disease seems higher than the number previously recognized⁴⁻⁶. The most frequent site involved are calcaneus, femur and tibia⁶. The age of the patient ranges around the fourth to fifth decade.⁶ There is no substantial difference in sex distribution.⁶ With the advancement of computed tomography (CT) and magnetic resonance imaging (MRI), the identification of intralesional fat, as well as dystrophic calcification and cyst formation is enabled.^{7,8} By using Milgram's histopathologic and radiologic classification, we divided intraosseous lipomas into three categories on the basis of their imaging characteristics⁸. We report one case of patient with intraosseous lipoma of tibia who was diagnosed by biopsy from curettage of the lesion.

CASE ILLUSTRATION

A 24 year-old female presented with history of increasing pain in her left knee for the last 6 months before admission. The pain was dull and was not referred to other locations, aggravated with physical activities and relieved with rest, and no nocturnal exacerbations. Patient also felt instability in her knee accompanied by 'locking knee' sensation. No swelling on the knee and no complains in other areas of the body. The patient then came to the orthopedic outpatient clinic in RSCM and suspected with knee ligament injury. She received oral analgesics and referred to radiology department for an MRI. After further examination, she was diagnosed with bone tumor in the left lower extremity and meniscal tear. The patient is handled by the orthopedics oncology division, consequently.

On physical findings there was tenderness, no mass, normal skin color, normal range of motion, and Mc Murray test was positive on the left knee (Fig. 1 a-b).

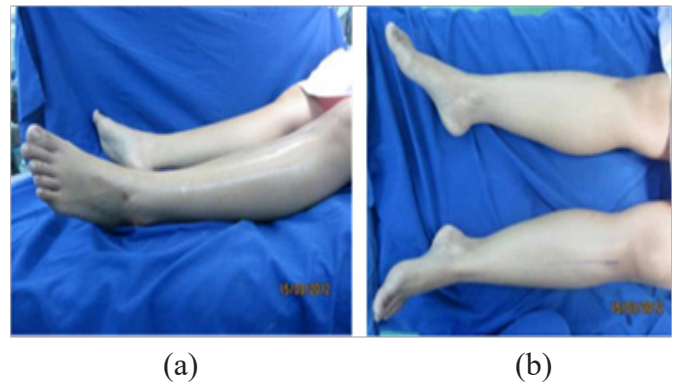


Figure 1 a-b. Clinical picture showing no abnormality of left leg

Laboratory findings showed the level of hemoglobin and white blood cells of 12.7 g/dl and 10.540/ μ l, respectively, normal erythrocyte sedimentation rate (20 mm/h) with normal serum alkaline phosphatase 154 μ /l (0 – 448) and increased lactic dehydrogenase 262 μ /m (100 – 190).

Anteroposterior and lateral radiograph of the left cruris showed geographic lytic lesion at middiaphysis of the left tibia with scalloping of the anterior cortex. There was no periosteal reaction and soft tissue mass (Fig. 2 a-b).



Figure 2 a-b. Anteroposterior and lateral radiograph of the left cruris showed geographic lytic lesion at middiaphysis of the left tibia with scalloping of anterior cortex, there is no periosteal reaction and soft tissue mass.

Magnetic resonance imaging demonstrated partially heterogenous cystic lesion (diaphyseal) in the left tibia suspected to be the primary bone tumor (dd/ fibrous dysplasia – ABC) and occult fracture in the left tibia diaphysis. There are also left medial and lateral meniscal tears (Fig.3 a-d).

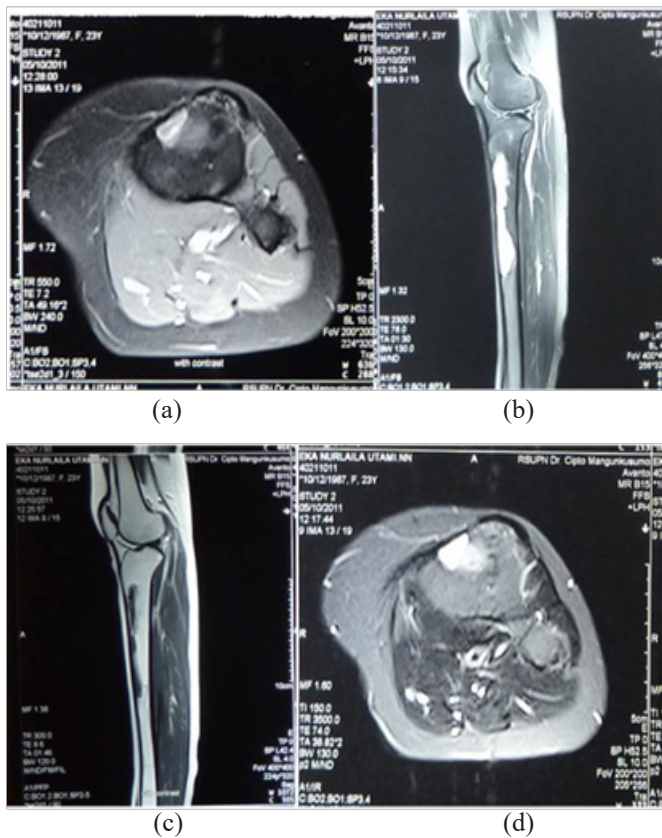


Figure.3 a-d. Partially heterogenous cystic lesion (diaphyseal) in the left tibia suspected to be the primary bone tumor (dd/fibrous dysplasia – ABC). Occult fracture in left tibia diaphysis. There are also left medial and lateral meniscal tears.

Thoracic radiograph showed no abnormalities (Fig.4).

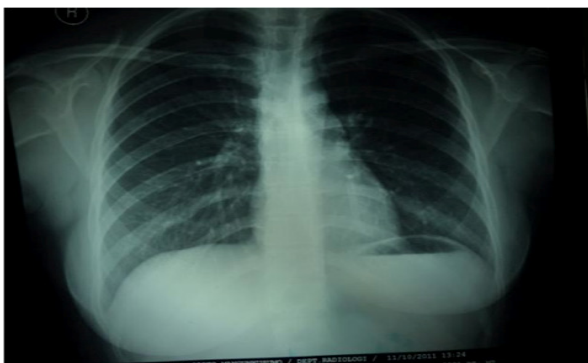


Figure.4. Anteroposterior radiograph of the chest showed no abnormalities.

The patient was admitted and underwent curettage, bone graft, and internal fixation with plate and screws. The anterolateral approach of the tibia was performed. The skin, subcutaneous and fascia incision were drawn along the proximal and shaft of the left tibia. The drilling of the tibial cortex was done and then serohemorrhagic fluid

came out. The curettage of the lesion was done in the intramedullary of diaphyseal tibia and then the tissue was sent to the Pathological Anatomy Department. In order to strengthen the shaft of the tibia after curettage, the insertion of a 12-hole T plate with 4 screws was performed to the shaft of the left tibia. (Fig. 5 a-e).

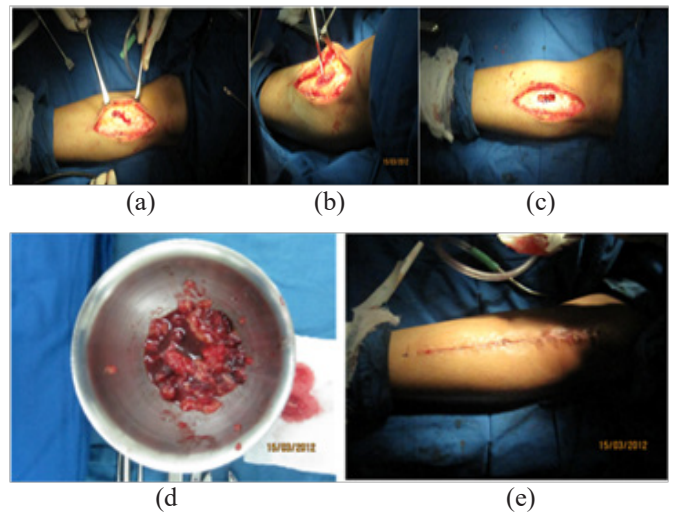


Figure. 5a: anterolateral approach of the tibia was done.

Figure. 5b: the cortex of the tibia had been drilled.

Figure. 5c: the T-plate had been inserted.

Figure. 5d: the surgical specimen from curettage

Figure. 5e: showing the sutured incision.

Postoperative anteroposterior and lateral radiograph of the left tibia showed the lesion fixed with a T plate and screw (Fig.6).



Figure.6. Postoperative anteroposterior and lateral radiograph of the left tibia

Histological examination of the surgical specimen revealed a chronic osteomyelitis, with no marked anomalies (fig.7). The patient was discussed at the Clinical Pathological Conference (CPC). Initially, from the result of CPC, the patient was diagnosed with

chronic osteomyelitis of the left tibia. The histological examination was then re-reviewed by conducting a deep section of the preparation and the result was lipoma of the bone (Fig. 8 a-b).

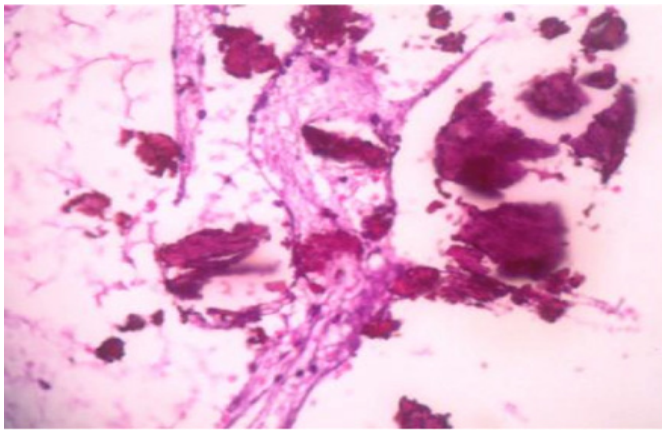


Figure.7. First histology examination showing inflammation cell with necrotic bone.

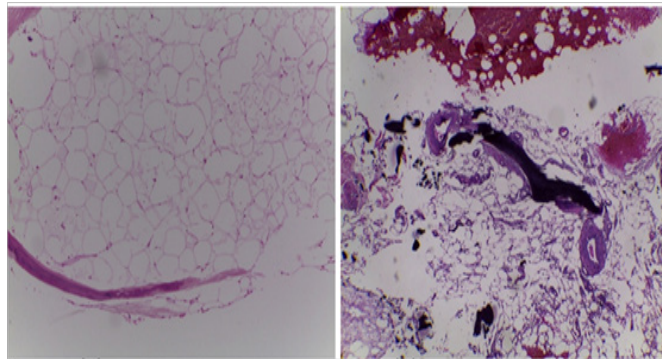


Figure.8 a-b. Second histology examination from deep section of the preparation showed large mature lipocytes with scattered necrotic bony trabeculae.

DISCUSSION

Intraosseous lipoma is regarded as a rare benign lesion of bone, accounting for less than 0.1% of primary bone tumors³. In recent years, an increasing number of cases of this disease have been reported, and the real incidence of the disease seems higher than the number previously recognized.⁴⁻⁶ However, Chow and Lee reported a higher incidence in their institution (2.5%).⁵ The sex predilection in intraosseous lipoma is controversial. Some authors reported that there was no sex predilection, while others reported bias to male.^{4,7} In this case, the patient was a female. The reported age distribution is fairly even, from the young to the elderly, with predilection in the fourth, fifth, and sixth decades of life^{1,4,7}. The age of our patient, however, was not in a good agreement with this distribution. Some authors reported up to 70%

patients with intraosseous lipomas presented with pain, while other authors reported most of the patients were asymptomatic.^{1,4,6,7,9} Our patient came to the orthopaedic outpatient clinic with pain in her knee, in which this pain could be due to the instability of the knee related to the medial and the lateral meniscal tears. Our patient did not have any pain in the region of the left tibia, therefore the lesion was considered as asymptomatic. The calcaneus and metaphysis of the long tubular bones, such as femur, tibia, fibula, and humerus, are the common anatomical locations, and multifocal locations are rarely found^{4,6,7,9}. We found the lesion of our patient in the tibia and it was solitary.^{1,3,6,7,9,11,12}

In plain radiograph, the diagnosis of intraosseous lipoma is so difficult due to the wide range of age groups with nonspecific clinical and radiologic findings¹. Radiographic differential diagnosis includes bone infarct, fibrous dysplasia, enchondromatosis, chondromyxoid fibroma, aneurysmal bone cysts, and other benign bone tumors.^{1,4,9-11} Milgram divided this lesion into three radiographic groups: stage 1, pure radiolucent areas that expand the cortex; stage 2, well defined radiolucent areas and central calcifications due to fat necrosis; and stage 3, bone resorption, new calcific areas at peripheral sides of the lesions, ossifications in the surroundings, and cyst formation.^{1,4,6,7,9,11,12} In our case, the expert of the left cruris plain radiograph from the radiologist only said that there was minimal bulging in the middiaphysis of the left tibia with intact cortex. But actually, we could see that there was radiolucent lesion in diaphysis of tibia with thinner cortex (Fig. 2a-b).

Although the diagnosis of intraosseous lipoma may be difficult based on plain radiographs alone, either CT or MRI is useful in the detection of fat within the lesion, allowing for a more accurate diagnosis.¹⁰ In this case, we did the MRI with the following results: partially heterogenous cystic lesion (diaphyseal) in the left tibia suspected to be a primary bone tumor (dd/fibrous dysplasia – ABC), occult fracture in left tibia diaphysis, and there were also found left medial and lateral meniscal tears. Cystic degeneration may be the primary feature shown in CT or MRI in stage 3 intraosseous lipoma.¹¹ Despite the heterogenous appearance of an involuted intraosseous lipoma in CT and MRI, identifying fat in the lesion leads to definitive diagnosis of intraosseous lipoma.^{8,11,13} The fat in the lesion shows high-signal intensity on both T1- and T2-weighted images and is similar to the subcutaneous fat in fat suppressed images^{4,11}. In this patient, we could

see fat appearance with high-signal intensity around the cystic lesion on T1-weighted and this was confirmed with the fat suppressed images, the area of fat around the cyst that was previously present was suppressed and becoming hypointense (Fig. 9 a-b).

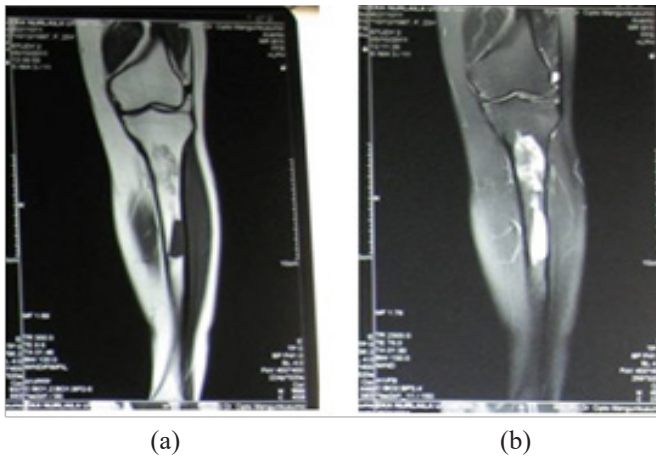


Figure 9. a T1-weighted image shows a low-signal intensity area in diaphysis of the left tibia suggesting cystic change of the lesion. The margin of the lesion shows high signal intensity, representing residual adipose tissue, which was verified histologically. b. T2-weighted with fat suppressed image shows higher signal intensity than the subcutaneous adipose tissue in the cystic lesion, but the margin of the lesion has the same signal intensity with the subcutaneous fat.

The incidence of bone lipoma is very rare in our Center. So, pre-operatively, we diagnosed this patient with aneurysmal bone cyst. The patient was admitted and underwent curettage, bone graft and internal fixation with plate and screws. From the first histological examination of the specimen, it was revealed to be a chronic osteomyelitis with no marked anomalies (Fig. 7). However, the clinical appearance did not support this diagnosis. The patient was discussed in the Clinical Pathological Conference (CPC). The CPC then suggested to repeat the histological examination. The result was bone lipoma (Fig. 8 a-b). The different results between the first and the second histological examination may be caused by several conditions, such as lack of samples and lack of experience. Lipocytes can be found in normal bone because they are normal structure of the bone marrow. Inflammation cells are frequently seen in histological examination of bone. Lacking experience with intraosseous lipoma, we assumed that the lipocytes were normal and the inflammation cells were the main problem. The inflammation cells found in our patient could be caused by the inflammation process near the necrotic bone.

From literature, it is stated that CT or MRI alone can almost establish the diagnosis, because the tumor consist of adipose tissue.⁴ Adipose tissue can be easily distinguished from fibrous tissue or usual tumor tissues that are rich in tumor cells by the Hounsfield value in CT⁴. In intraosseous lipomas, CT has a uniform soft tissue density, with the same attenuation as adipose tissue of about -100 Hounsfield units.⁴ MRI represents an additional method that is capable of demonstrating fatty tissue exquisitely. Intraosseous lipomas show homogenous, high signal intensity, identical to that of normal adipose tissue in both T1, and T2-weighted images.⁴ MRI with fat suppression signal (STIR) can be helpful in diagnosing intraosseous lipoma, because both T1-weighted and T2-weighted MRI of hematoma may show high signal intensity, mimicking the adipose tissue⁴.

The need for surgical treatment is controversial.^{9,11} Most intraosseous lipoma, however, can be managed conservatively.^{4,6,7,9} Curettage and bone grafting are the treatments of choice when surgical intervention is needed.¹¹ The following are the indications for surgery: (1) painful tumor, (2) occurrence of pathological fracture, (3) necessity for histological diagnosis, and (4) required to decrease the risk of malignant transformation.^{4,6,7,14} In our case, we did curettage, bone grafting, and internal fixation with T plate and screw. The indication of curettage in this case was to obtain the tumor mass for histological diagnosis. We still could not diagnose the patient from the radiological examination only. The incidence of this case was very rare in our Center and we did not have sufficient experience in managing this case. We did bone grafting to fulfill the empty space that was left after curettage. The indication for insertion of internal fixation was impending fracture due to post-curettage lesion in the shaft of tibia. Additionally, the MRI showed occult fracture in the shaft of the left tibia. Mirels proposed a scoring system for impending fracture based on four characteristics: (1) site of lesions; (2) nature of lesion; (3) site of lesion; (4) pain.¹⁵ By following the Mirels scoring system for diagnosing impending pathologic fractures, our patient got a score of 9. According to Mirels' recommendation, prophylactic fixation is highly indicated for a lesion with an overall score of 9 or greater.¹⁵

One year after the surgery, we evaluated the oncologic outcome, there was no local recurrence or infection detected. The orthopaedic outcome showed that the graft was already united, but the plate and screw fixation still

needed to be retained because the patient decided to remove the implant when she had her ligament operated. The functional outcome showed that the patient could walk and had full range of motion (Fig. 10 a-c)

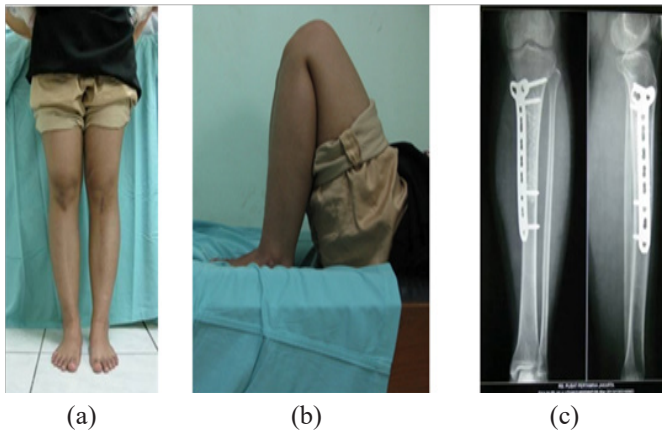


Figure. 10. A year after surgery. (a-b) Clinical pictures show the patient can stand and has good range of motion. (c) Anteroposterior and lateral cruris radiographs show that the graft is already united and no sign of local recurrence

CONCLUSION

Intraosseous lipoma is a rare benign bone lesion that is difficult to diagnose based on plain film imaging alone. However, CT and MRI can reveal intraosseous lipoma accurately. In spite of this, the diagnosis of intraosseous lipoma still need to be confirmed by the collaboration of orthopaedics, radiologist and pathologist in a clinical pathological conference.

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