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Delayed union fracture post wide excision anterior compartment of femur because of liposarcoma: a case report from radiologic perspective

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Abstract. The prevalence of delayed union fracture is estimated by 5-10% in all long bone fracture. In this case report, we will present a complete report of delayed union in a patient post thigh liposarcoma wide excision. A 52 years old woman was assigned to emergency department after slipped and fell down on the floor. Her last operation was 10 years ago with debulking and wide excision on anterior compartment of thigh region. Clinical examination shows deformity on her thigh. Her sensory nerves and distal vascularization was found to be normal. X-Ray showed comminutive fracture on one third of medial femur. Callus formation seen on eight month. Delayed union fracture is a multifactorial case so as clinicians we must pay attention to various patient conditions in handling a fracture case. Radiology is an important additional examination, however radiology cannot stand alone.

Keywords: delayed union fracture, liposarcoma, wide excision, communutive fracture

Abstrak : Prevalensi fraktur union tertunda diperkirakan 5-10% pada semua fraktur tulang panjang. Dalam laporan kasus ini, kami akan menyajikan laporan lengkap tentang penundaan penyatuan pada pasien pasca eksisi luas liposarkoma paha. Seorang wanita berusia 52 tahun dibawa ke unit gawat darurat setelah terpeleset dan jatuh ke lantai. Operasi terakhirnya adalah 10 tahun yang lalu dengan debulking dan eksisi luas pada kompartemen anterior daerah paha. Pemeriksaan klinis menunjukkan deformitas pada pahanya. Saraf sensorik dan vaskularisasi distalnya ditemukan normal. X-ray menunjukkan fraktur kominutif pada sepertiga femur medial. Pembentukan kalus terlihat pada bulan ke delapan. Fraktur union tertunda merupakan kasus multifaktorial sehingga sebagai klinisi kita harus memperhatikan berbagai kondisi pasien dalam menangani suatu kasus fraktur. Radiologi merupakan pemeriksaan tambahan yang penting, namun radiologi tidak dapat berdiri sendiri.

Kata kunci: delay union fracture, liposarcoma, eksisi luas, fraktur komutif

Introduction

Delayed Union Fracture is the absence of radiographic progression of healing instability of a fracture upon clinical examination between 4 and 6 months after injury. The prevalence of delayed union fracture is estimated by 5-10% in all long bone fracture. Delayed union fracture caused by many factors including the severity of the fracture, location, bone loss, immobilization, blood supply, soft tissue damage, tumor, radiation therapy, and the presence of infection. Comorbidities such as diabetes, vascular insufficiency, osteoporosis, nutritional deficiency, anemia, hormonal deficiency, smoking, advanced age, or certain medication may predispose to delayed or impaired healing.[1]
The primary management of liposarcoma is surgery but multi modality for high grade liposarcoma use wide and deep surgical excision, radiation adjuvant, and/or chemotherapy. However, the response of liposarcoma to chemotherapy is not well documented. Radiation adjuvant may have valuable benefit especially in myxoid subtype. For esophageal liposarcoma surgery can be submucosa resection minimal invasive endoscopy or partial esophagectomy or total aggressive.[2]
Wide excision and radiotheraphy usage in liposarcoma can be contributed to delayed union fracture. In this case report, we will present a complete report of delayed union in a patient post thigh liposarcoma wide excision

Case Description

A 52 years old woman was assigned to emergency department with right thigh pain. She has height 155 cm and weigh 60 kg. She reported that she slipped and fell down on the floor. Her medical history included third time operation on her thigh because of soft tissue tumor. The last operation was 10 years ago with debulking and wide excision on anterior compartment of thigh region. She denied history of smoking, diabetic, and hypertension. Clinical examination shows deformity on her thigh. Her sensory nerves and distal vascularization was found to be normal. The X-Ray showed comminutive fracture on one third medial femur and showed osteolytic representation that raised the assumption of

malignancy (figure 1). Because of that we conclude pathological anatomy (PA) examination on this patient. The PA showed muscle tissue fragment and fibrous tissue fragment, unseen malignancy, unseen tumor cell. Patient timeline in described in table 1.

Patient installed plates on one third medial femur (figure 2). Preoperative patient administered cefazolin (1 gram) for 24 hours. After three days of admission patient was discharged. Patient advised to non-weighbearing for two weeks. Patient can walking with four-legged walker on three weeks then patient have to do rehabilitation for eight times to exercise the muscle, knee range of motion (ROM), and balance the gait.

Callus formation on patient can be found at eight month after surgery. On follow up showed that the plate installed properly, with no angulation and deformity. After two years patient can walk without walker again.

Figure 1. Closed comminutive fracture on 1/3 medial femur with three fragment fracture (butterfly fracture). Red circle showed osteolytic representation that make an assumption of malignancy.

Figure 2. Xray AP ORIF plating on 1/3 medial femur

Table 1. Patient Timeline

No	Date	Event
1	December 2007	Patient reported a mass in 1/3 proximal femur with the size of hazelnut seeds. On USG 3 dimension showed that patient have neurofibroma with vascularization around it. Patological Anatomy (PA) resulted Mixed Neurofibroma with size 1.5 cm x 1 cm x 1 cm
2	2008	Patient have surgery in Al Irsyad Hospital. During surgery doctor suspects that there is malignancy, because he discovered fat tissue on tumor lesion.
3	May 2009	Mass appeared again “recurrent”.
4	June 2009	Patient have surgery in Onkologi Surabaya Hospital . PA resulted liposarcomawith size 2,5cm x 2 cm x2 cm. Then the doctor decide to remove the abductors muscle following with chemotherapy for 6 times and radiotheraphy for 35 times.
5	May 2013	Mass appeared again. Patient have surgery again with debulking and wide excision at Graha Amerta Surabaya Hospital.
6	December 2020	Pasien slipped and fell down on the floor and have closed comminutive fracture on 1/3 femur.

Discussion

Liposarcoma is the most common soft tissue sarcoma, accounting for approximately 20% of all malignant neoplasms occurring in mesenchymal tissue. The World Health Organization (WHO) for the Classification of Soft Tissue Tumors in 2020 categorizes soft tissue liposarcoma into five types including well differentiated liposarcoma, dedifferentiated liposarcoma, myxoid liposarcoma, pleomorphic liposarcoma, and myxoid-pleomorphic liposarcoma.[2]

Liposarcoma cases are most often found in the retroperitoneum, lower extremities where the most common sites are the femur region, upper extremities, and in the head and neck region. Liposarcomas are usually found in deep soft tissue, originate from primitive mesenchymal cells, differentiate into mature adipose tissue. Patients usually present with large masses without pain.[3]

On radiographic examination, it may appear as a soft tissue mass, depending on the size and location of the lesion. Fatty tissue is often seen in large masses which can affect the aesthetic function of the extremities. On ultrasonography (USG), well-differentiated liposarcoma appears as a heterogeneous, multilobular mass, usually well circumscribed. On CT scan and MRI, well-differentiated liposarcoma usually has a relatively characteristic appearance as an adipose mass that predominantly contains non-lipomatous components. Generally, more than 75% of well-differentiated liposarcomas consist of fat. The non-lipomatous component is most commonly seen as prominent thick septa (>2 mm) which may indicate nodularity. Focal nodular or globular non-adipose areas may also be seen, but are usually less than 2 cm in size.[4]

Dedifferentiation liposarcoma usually coexists with well-differentiated liposarcoma. Dedifferentiation liposarcoma has the same radiological features as well differentiated liposarcoma. The large component of differentiated liposarcoma, usually predominant by size, appears identical to the previously described features of well-differentiated liposarcoma seen on radiographs and ultrasound, CT, and MR images.[5]

Plain radiographs of myxoid liposarcoma may appear normal or more often reveal a nonspecific soft tissue mass. Radiolucent fat features are rare compared to well-differentiated liposarcomas, due to the much smaller adipose tissue volume (often >10%). Ultrasound can provide a complex, well-defined, hypoechoic but dense, noncystic mass with posterior acoustic enhancement. On CT and MR imaging, the myxoid liposarcoma is usually large, well-defined, and multilobular intermuscular lesions. The high water content of the lesions was reflected as a predominant low attenuation on CT images, low signal intensity on T1 MR images, and marked high signal intensity on T2 MR images. The pathognomonic feature is adipose tissue seen in the mass.[6,7]

The use of MRI is superior to CT in imaging myxoid liposarcoma, because the contrast resolution is increased. Fat is also usually only a small volume of the overall mass (10% of lesions) and is often seen in septa (lacy or linear pattern) or as small fine nodules in the lesion. Pathognomonic appearance of fatty septa or small adipose nodules in myxoid masses has been reported in 42%-78% of cases. Identification of fine or small fat should be assisted by a comparison of weighted T1 and T2 in the same plane (the axial plane is optimal). [6,7]

The most common treatment for liposarcoma is surgery. The main problem with liposarcoma therapy is the frequent recurrence of the tumor. In a retrospective study involving 97 subjects with well differentiated liposarcoma. A total of 76.3% with radical excision and 23.7% with excisional excision. From these studies found as many as 13% of patients experienced tumor recurrence with a life expectancy of 60-85%. Patients with tumors of the extremities have a significantly better prognosis than those with retroperitoneal or scrotal tumors. Prognosis will be worse in patients with well differentiated changes or co-occurs with dedifferentiated liposarcoma.[2,3,8]

Bone healing is influenced by several factors. In this case, the patient had a wide excision that took up almost the entire anterior femur compartment, leaving only the blood vessels. This will reduce the formation of stem cells as one of the precursors of bone healing. External trauma that causes extensive soft tissue and bone injury will affect the origin of stem cells which decreases the effect on healing, the degree of differentiation of cartilage cells into bone cells. and progression of fracture healing. The periosteum and muscles each have an important role in bone healing. Periosteal cells specifically respond to bone morphogenetic protein 2 (BMP-2) to the induction of chondrogenesis and osteogenesis, whereas cells in the marrow space will only form bone in response to BMP-2.[9] Studies using transgenic mice with periosteum harvesting with additional muscle fenestration, showed that nearly half of the cells in the callus were derived from the surrounding muscle.[10]

Very extensive injury, too much damage to the muscle and periosteum can create problems in bone healing process. Also important is to consider the extent to which the vascular tissues in the surrounding muscle sheath are compromised; failure of angiogenesis after fracture or osteotomy can lead to nonunion.[11]

Callus formation in the healing process of bone can usually be evaluated radiologically. Plain radiographs are usually used to evaluate callus formation because of their relatively low cost, availability and low radiation. Therefore, a scoring using plain photographs was developed, namely Radiographic Union Scale for Tibia (RUST), Radius Union Scoring System (RUSS), and Radiographic Union Scale for Hip (RUSH). The scoring system can be used to predict the presence of non-union.[12] In 2015 the presence of mRUSH was found to evaluate fractures of the femur and tibia, but the mRUSH score has not been applied to predict nonunion.[13]

The use of ultrasound for fracture cases is very rarely done. Ultrasonography can be used for the diagnosis of pediatric fractures, identification of long bone fractures in the prehospital setting and difficult-to-detect stress fractures. The ability to detect callus early holds great promise for monitoring fracture healing. Callus formation at the corticotomy site after distraction osteogenesis of limb length has been discovered earlier by ultrasound than by radiography. In addition, ultrasound evaluation of soft cartilaginous callus in long bone fractures with nonunion revealed a marked lack of calcification and density.[12]

The use of computed tomography (CT) scanning to evaluate fracture healing is becoming increasingly popular as it provides high-resolution multi-planar imaging to determine evidence of bridging callus. It has the ability to detect callus prior to radiography and appears to increase the sensitivity and agreement of fracture union between observers. However, there is evidence to suggest CT may 'overdiagnose' nonunion with poor specificity for fractures of long bones such as the tibia. The optimal timing of using CT scans to determine formation has not been well evaluated.[12,14]

MRI has also been used to investigate callus development in the early phase of fracture healing in animal models, but, similar to CT scans, it has not been shown to be useful in the clinical setting as an early predictor of nonunion in the first few weeks after injury. Dynamic contrast-enhanced MRI (DCE-MRI) can be used to evaluate the vascularity of the nonunion site. [15]

Conclusion

Delayed union fracture is a multifactorial case so as clinicians we must pay attention to various patient conditions in handling a fracture case. The diagnosis of delayed union fracture was made when no callus was formed for more than 6 months.

Radiology is an important additional examination in the field of medicine. However, radiology cannot stand alone, it must be confirmed using the patient's history and physical examination. In the case of tumors such as liposarcoma, histopathological examination is very important to determine the grade and type of liposarcoma. Wide excision performed for tumor management can cause problems with bone healing in the future.

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