Outcomes of prophylactic intramedullary fixation for benign bone lesions

İyi huylu kemik lezyonlarında profilaktik intramedüller fiksasyon sonuçları

Çağrı Neyişci, Yusuf Erdem, Ahmet Burak Bilekli

Gülhane Training and Research Hospital, Department of Orthopedics and Traumatology, Ankara, Turkey

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ÖZET

Amaç: Bu çalışmada benign kemik lezyonu sebebiyle profilaktik intramedüller tespit ameliyatı yaptığımız hastaların sonuçlarını sunmayı amaçladık.

Gereç ve Yöntem: Bu çalışmaya 2008-2017 yılları arasında benign kemik lezyonu nedeni ile küretaj, greftleme ve profilaktik intramedüller tespit ameliyatı yaptığımız 22 hasta dâhil edilmiştir. Lezyonlar preoperatif dönemde Mirels sınıflamasına göre incelenerek patolojik kırık riski belirlenmiştir. Tüm hastaların tedavisinde küretaj, greftleme ve intramedüller tespit yöntemi kullanılmıştır. Kontrol muayenelerinde hastalar; eklem hareket açıklıklarına, ağrı durumuna, lezyonun ve implantın radyolojik görüntüsüne göre değerlendirilmiştir.

Bulgular: Hastaların yaş ortalaması 24,8 (aralık, 7-38) yıldı. Hastalar ortalama 35,8 (aralık, 13-80) ay takip edildi. Çalışmaya dâhil edilen 21 hastanın ilk başvuru nedeni ağrı olup 2 hastada ağrı fonksiyon kaybına neden olmaktaydı. Patolojik humerus kırığı olan bir hasta akut ağrı ve fonksiyon kaybı ile başvurdu, iki ay konservatif olarak takip edildi ve ardından profilaktik cerrahi yapıldı. Hastaların ortalama Mirels skoru 9,3 (aralık, 9-10)'tü. Takiplerde tüm hastaların ekstremite fonksiyonları tamdı. Ameliyat sonrası ortalama VAS 8,09'dan 2,54'e gerilemiştir.

Sonuç:9 veya daha fazla Mirels skoruna sahip olan iyi huylu kemik lezyonları için profilaktik fiksasyonun, olası patolojik kırık riskini azalttığı, VAS skorlarını azalttığı, ayrıca fonksiyon kaybını önlediği ve daha erken normal aktivitesine geri dönüşe olanak sağladığı sonucuna vardık.

Anahtar Kelimeler: Benign kemik lezyonu, intramedüller tespit, profilaksi.

ABSTRACT

Background: In this study, we aimed to present the results of patients who underwent prophylactic intramedullary fixation for benign bone lesions.

Materials and Methods: Twenty-two patients who underwent curettage, grafting and prophylactic intramedullary fixation for benign bone lesions between 2008 and 2017 were included in this study. The lesions were examined according to the Mirels' classification in the preoperative period and the pathological fracture risk was determined. Curettage, allografting and intramedullary fixation were used in the treatment of all patients. In the follow-up examination, patients were evaluated according to the range of motion, pain, radiological appearance of the lesion and the implant.

Results: The mean age of the patients was 24.8 (7-38) years. The mean follow-up period was 35.8 (13-80) months. The initial complaint of twenty-one patients was pain which caused loss of function in two patients. One patient with pathological humerus fracture was admitted with acute pain and loss of function. He was followed conservatively for two months, then prophylactic surgery was performed. The mean Mirels' score of the patients was 9.3 (9-10). In the follow-up examination, the range of motion was full in all patients. The mean VAS score decreased from 8.09 to 2.54 postoperatively.

Conclusions: We conclude that prophylactic fixation for benign bone lesions which has 9 or more Mirels' score reduces the risk of impending pathological fractures, reduces VAS scores, and also prevents loss of function and enables to return normal activity earlier.

Key Words: Benign bone lesion, intramedullary fixation, prophylaxis.

INTRODUCTION

Benign bone lesions have a heterogeneous nature as "true bone tumors" and "tumor-like lesions" (1,2). Most of the benign bone lesions are asymptomatic and their incidence is unknown. Their location, the tissue which they originate from, clinical presentation and their progression differ significantly. They are usually encountered in children and young

Adress for correspondence: Uzm. Dr. Çağrı Neyişci, Gülhane Eğitim ve Araştırma Hastanesi, Ortopedi ve Travmatoloji Anabilim Dalı, Emrah Mahallesi, Gen. Dr. Tevfik Sağlam Cd., Ankara, 06010, Turkey
e-mail:cagri_neyis@yahoo.com
Available at www.actaoncologicaturcica.com

adults which may be asymptomatic or present with clinical signs such as pain, swelling, compression of surrounding tissues and pathological fracture. Diagnosis is made by using radiological methods carefully blended with the clinical presentation. The definitive diagnosis is made by biopsy. Most benign bone lesions are diagnosed incidentally. Treatments are controversial and the risk of recurrence is high (3,4,5).

The treatment includes surgical methods such as resection, curettage only, curettage and grafting. The aim of treatment is to achieve high recovery rate, low probability of recurrence and early return to normal activity (6).

Pathological fracture is a common cause for clinical admission in benign bone lesions and also an important cause of morbidity. The most common benign bone lesions which cause pathological fractures are simple bone cyst, non-ossifying fibroma, aneurysmal bone cyst, and fibrous dysplasia (7). Furthermore, the most common initial presentation of simple bone cyst in children and adolescents has been reported as pathological fracture (8).

Various surgical methods are used in the treatment of pathological fractures and preventive procedures. Prophylactic surgery will eliminate the loss of function after impending fracture. In this study, we aimed to present the clinical and radiological results of twenty-two patients who underwent allografting and prophylactic intramedullary fixation for benign bone lesions between 2008 and 2017.

MATERIALS and METHODS

Twenty-two patients (15 male, 7 female) who underwent curettage, allografting and prophylactic intramedullary fixation for benign bone lesion between 2008 and 2017 were included in this study. Patients with benign bone lesions which did not require prophylactic surgery according to the Mirels classification were excluded (7). Demographic characteristics of the patients, their initial symptoms, localization and radiological appearance of the lesions, histological findings and postoperative morbidity data were evaluated. The lesions were scored preoperatively according to Mirels classification (Table 1) (7) and pathological fracture risk was determined. According to this classification, the risk of pathological fracture is between 33% and 100% for the patients with a total score of 9 or more and thus prophylactic fixation is recommended (7).

After curettage of the lesion, bone cavity was filled with allografts and intramedullary fixation was utilized. Postoperatively, physiotherapist-controlled passive range of motion (ROM) and isometric exercises were initiated at days 1-3. At days 15-30, active ROM exercises were introduced, and at days 30-45 active strengthening exercises were initiated. Postoperative follow up examinations were performed for three months interval for the first year, biannual for second year and then annually. Patients evaluated clinically via range of motion and VAS scores. For radiologic evaluation, anteroposterior and lateral radiographs were obtained to examine the appearance of the lesion, presence of any recurrence or pathological fracture, the position or any mechanical complications of the implants.

The study protocol was approved by the Institutional Review Board. The study was conducted in accordance with the principles of the Declaration of Helsinki. A written informed consent was obtained from each patient.

RESULTS

The mean age of the patients was 24.8 (7-38) years. The mean follow-up period was 35.8 (13-80) months. The initial symptom of twenty-one patients was pain and it caused function loss in two patients. One patient was admitted to emergency department with acute pain and loss of function and diagnosed with pathological humerus fracture. He was followed conservatively and prophylactic surgery was performed two months later. In twelve patients benign bone lesions were localized in the humerus and in the femur in remaining ten. Of the twenty-two patients, fourteen had simple bone cyst (8 humerus, 6 femur) (Figure 1), four had enchondroma at the humerus (Figure 2), and four had fibrous dysplasia at the femur (Figure 3) (Table 2). The mean Mirels' score of the patients was 9.3 (9-10). At the postoperative period, one patient who had titanium elastic nailing of the

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e-mail:cagri_neyis@yahoo.com
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humerus sustained a non-displaced pathological fracture at the second month and was followed conservatively. There were no additional pathological fractures or complications in the other patients. For twelve humeral lesions, titanium elastic nailing (TEN) was performed for eight patients and intramedullary nailing (IMN) for four patients. For ten femoral lesions, IMN was performed for six patients and proximal femoral nail (PFN) was performed for four patients (Table 2). The mean VAS decreased from 8.09 to 2.54 postoperatively. Postoperatively, there were no loss of ROM and extremity functions compared to the preoperative period.

DISCUSSION

Benign bone lesions have a wide spectrum according to their histological structure. They are classified according to the tissue they are derived from. Usually they do not present any symptoms and the diagnosis is made incidental. The main diagnostic method used in bone tumors and tumor-like lesions is plain radiographs and most of them can be diagnosed only by plain radiographs. Cross-sectional imaging (CT, MRI) is used to evaluate some lesions that cannot be diagnosed by direct radiography. Radiological appearance of benign bone lesions shows cortical thinning, enlargement of the medulla and loss of normal trabecular pattern. Ultimately, the lesion changes the bone geometry (1-4).

Simple bone cyst was first described by Virchow in 1876 and its etiology has not been revealed in every aspect (9). The most popular theory trying to explain its etiology is the obstruction of venous return and increased interosseous pressure within the bone (10). A simple bone cyst can involve any bone and can be seen in any age group. It is most commonly seen in the first decade (11). In our study, the mean age of fourteen patients diagnosed with simple bone cyst was 19.8 (range, 7-38) years. The most common symptom is pathological fracture (12). It presents with 75% pathological fracture in the pediatric age group (13). In our study, the only patient who had preoperative pathological fracture was diagnosed with simple bone cyst. Literature reveals that the most common affected bones are the humerus (55-65%) and the femur (25-30%), respectively. Occasionally tibia, fibula, radius, ulna and rarely pelvis are affected (14). In our study, eight of the fourteen simple bone cysts were located at the humerus and six of them were located at the femur, which is consistent with the literature. Previous studies determined the cyst activity by measuring its size (15). Recent studies have shown that the size of the cyst alone is not sufficient (16,17,18). Therefore, not only the cyst size, but also the other parameters of the Mirels' classification (Table 1) (7) were evaluated in our study. For the treatment of the simple bone cysts, follow-up, intralesional injections, curettage, curettage and grafting, internal fixation or combination of these methods are used. Steroids, bone marrow aspirate, demineralized bone matrix (DBM), calcium sulfate are the choices for intralesional injections (16,17,18). Intralesional steroids cause degeneration of the cyst wall, decrease the fluid in the cyst and increase the osteoblastic activity (19). The long-term results of percutaneousmethylprednisolone administrationare not satisfactory (20). In the adolescent group, curettage and grafting yields 55-65% improvement (21). But the recurrence in 35-45% of patients necessitates additional surgical interventions (22). Therefore, these treatment modalities alone may not be sufficient (23) and the lesions should be evaluated thoroughly for pathological fracture risk.

Enchondroma is a benign tumor originating from hyaline cartilage. It constitutes 12-24% of all benign bone tumors. It is most commonly located in the metaphysis of the bones. It is seen equally among men and women. The most common locations are the proximal humerus (13%), distal femur (7%) and the proximal tibia (7%) (24). In our study, all of the four enchondromas were located at the humeral metaphysis, which is consistent with the literature. It is generally asymptomatic, and when the patient is symptomatic, the most common complaint is pain, and it should be kept in mind that the patient presenting with pain is an important marker for the pathological fracture risk (24-27). In our study, all our patients with enchondroma presented with pain. Direct radiography, MRI and bone scans are generally utilized for diagnosis. Follow-up, curettage and graft or cement application and internal fixation are occasionally used for the treatment. However, most authors recommend

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e-mail:cagri_neyis@yahoo.com
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curettage only. The rationale behind this technique is that mesenchymal cells in the blood filled into the bone cavity after curettage of the lesion will differentiate into osteoblasts and fill the cavity (24, 25). Enchondromas may recur after treatment and may even differentiate into sarcomatosis form (26). The recommended treatment is conservative and patient is followed at 3 or 6-month intervals. (27). In our study, a patient who presented with pain had a preliminary diagnosis of enchondroma and was followed conservatively at 3-month intervals. Mirels' score was calculated as 6 at the time of admission. Nine months after the onset of symptoms, the Mirels' score increased to 9 and curettage, allografting and prophylactic TEN fixation was applied. Other patients with enchondroma had a Mirels' score of 9 at the time of their admission and curettage, allografting and prophylactic fixation was the first choice of treatment.

Fibrous dysplasia is a benign intramedullary fibro osseous lesion. It constitutes 5-7% of all benign bone tumors (30). The bone marrow is replaced with fibrous tissue and abnormal bone development occurs. Although the etiology is uncertain, genetic factors are thought to be responsible. Mutations in 20q13.2-13.3 genes on chromosome 20 have been reported (28). Fibrous dysplasia can occur in a single bone or in multiple bones. In addition to long bones, it can also settle in the head of bones. Proximal femur, maxilla and the tibia are the most commonly involved bones. It's followed by humerus, ribs, radius, and iliac bones (29). In our study, one of the four cases was located at the proximal femur; the other three patients had lesions at the femur and tibia. Involvement of multiple bones could be a manifestation of systemic diseases. McCune-Albrigt syndrome is an endocrinopathy which affects skin, endocrine tissues and the bones (polyostotic fibrous dysplasia). Another syndrome associated with fibrous dysplasia is Mazabraud syndrome. It is characterized by single or multiple intramuscular myxomas seen along with fibrous dysplasia (31, 32). None of our cases had concomitant endocrinopathy. Pain is the most common cause of admission and can be aggravated during pregnancy and menstrual period. It is thought to be related with the estrogen receptors detected in fibrous dysplasia (33). The initial complaint of four fibrous dysplasia patients in our study was pain and it caused function loss in one patient. All of the patients' Mirels score was 10. Ground- glass appearance on X-rays is typical for fibrous dysplasia (34). Malignant transformation is very rare and below 1% (35). The most common transformation is osteosarcoma, followed by fibrosarcoma and chondrosarcoma (36). Fibrous dysplasia can localize at the load bearing regions of long bones and cause deformities during bone development. The most common deformity is the "shepherd's crook deformity" which is encountered at the fibrous dysplasia lesions located proximal to the femur (30,35). Although the lesions were located in the proximal femur in all of our cases, no deformity was observed.

Pain is the most common symptom in benign bone lesions and micro fractures should be suspected in the patients presenting with pain. CT and/or MRI can be used for evaluation. Pain is a parameter used to estimate the risk of pathological fracture (37). Pathological fracture depends on the fragility and load bearing limits of the bone (38). Benign bone lesions may cause skeletal deformities. This is thought to be caused by recurrent micro fractures. The greatest difficulty of preventing pathological fractures is late diagnosis. Most cases present with pathological fracture at initial presentation (13). In our study, the mean VAS score decreased from 8.09 to 2.54 postoperatively. This significant decrease in VAS scores suggests that micro-fractures may be present at our patients at the time of their admission.

The need of surgery is controversial and has not been fully clarified yet in which lesions should be operated or followed (37-40). The "undesirable result" is that the patient presents with a pathological fracture. Traditionally, the size of the lesion has been emphasized to determine the risk of pathological fracture (15). At the present, lesion size alone is not sufficient to reveal this risk. The localization and the radiological appearance of the benign bone lesion, degree of pain and the patient's age are the other factors that predispose to pathological fracture. In 1989, Mirels defined a classification of 4 parameters to reveal the risk of pathological fracture (7). This classification has been a guide in terms of surgical decision.

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Although many methods have been described for the treatment of benign tumors and tumor-like lesions of the bone, fixation may be required after decompression/curettage (1-4). Guille et al. reported recurrence and micro fractures in 66% of patients who underwent curettage and grafting for fibrous dysplasia (39). Aggressive debridement to be performed in benign bone lesions has many complications and may cause infection, intraoperative fractures, increased intraoperative blood loss, physeal damage and epiphyseal premature closure, limb length discrepancy and long-term immobilization (40,41). One patient who had titanium elastic nailing of the humerus sustained a non-displaced pathological fracture at the second month and was followed conservatively. There were no additional pathological fractures or complications in the other patients. No loss of function developed in any of our cases.

Pathological fractures in the axial and appendicular skeleton cause pain and loss of function, so the first goal should be to prevent fracture. Simple bone cyst and non-ossified fibroma could spontaneously heal and the pathological fracture risk should be evaluated thoroughly (1-4,37,41). With the help of a reliable method, the risk should be determined. If it is considered high, it should be decreased with an appropriate treatment method.

Currently, there is no proven clinical or radiological guide to accurately assess the risk of pathological fracture. Previous studies have attempted to assess the pathological fracture risk based on the patient age; anatomical location, activation, the size and the cortical deformation of the lesion (6-8,13,21,22,27,30,34,35,37,38,42,43). Based on these parameters, the progression of the lesion should be evaluated and the approximate pathological fracture risk and the need for prophylactic fixation should be performed. Catier et al. applied flexible intramedullary nails after curettage of simple bone cysts located at the proximal femur of two cases (42). Most authors consider the use of intramedullary nails is an easy, minimally invasive and complementary method for healing in simple bone cysts. In addition, early stabilization after decompression allows early mobilization and low risk of complications (43).

With this surgical technique, we achieved rapid return to normal activity and prevented the loss of function after pathologic fracture in the treatment of benign tumors and tumor-like lesions of the bone.

Conflict of interest: The authors declare that there is no conflict of interest.

Table 1: Mirels' Classification.

Variable	**Score**		
	1	**2**	**3**
Site	Upper limb	Lower limb	Intertrochanteric
Lesion	Blastic	Mix	Litic
Size[1]	<1/3	1/3-2/3	>2/3
Pain	Mild	Moderate	Functional [2]
Total Score	**Fracture Risk (%)**	**Recommendations**	
≥9	33-100	Prophylactic fixation recommended	
=8	15	Clinical status should be evaluated	
≤7	<4	Observation and radiotherapy	

[1]Size of lesion relative to bone diameter [2]Pain limiting limb function

Table2: Histopathologic diagnosis of the patients, affected extremity and the applied surgical fixation methods in our study.

	Humerus		**Femur**	
	TEN	IMN	PFN	IMN
Simple bone cyst	6	2	4	2
Enchondroma	2	2	-	-
Fibrous dysplasia	-	-	-	4

(TEN: Titanium elastic nail, IMN: Intramedullary nail, PFN: Proximal femoral nail)

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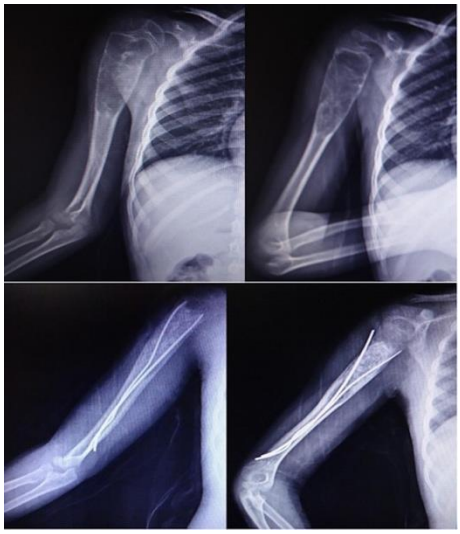

Figure 1: Preoperative and postoperative anteroposterior and lateral radiographs of a patient with a simple bone cyst located proximal right humerus.

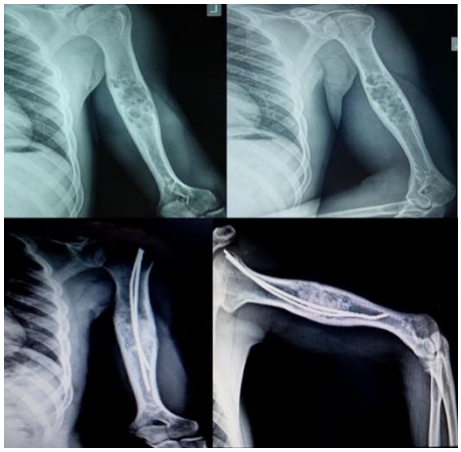

Figure2: Preoperative and postoperative anteroposterior and lateral radiographs of a patient with an enchondroma located in the left humerus diaphysis.

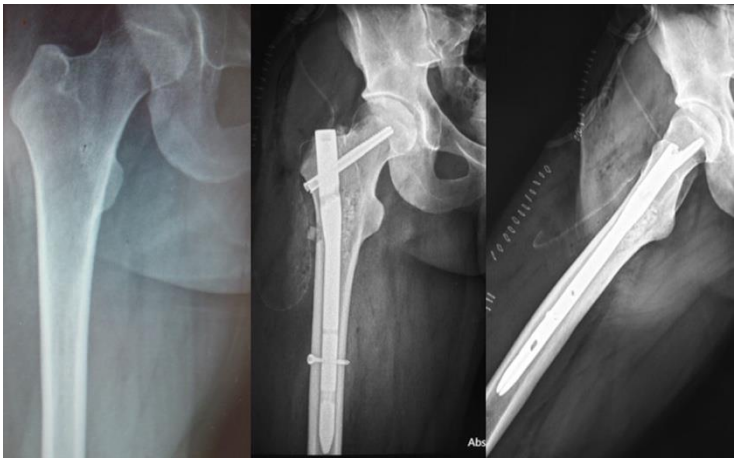

Figure 3: Preoperative and postoperative radiographs of a patient with fibrous dysplasia at the right proximal femur.

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e-mail:cagri_neyis@yahoo.com
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Adress for correspondence: Uzm. Dr. Çağrı Neyişci, Gülhane Eğitim ve Araştırma Hastanesi, Ortopedi ve Travmatoloji Anabilim Dalı, Emrah Mahallesi, Gen. Dr. Tevfik Sağlam Cd., Ankara, 06010, Turkey
e-mail:cagri_neyis@yahoo.com
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