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Chondromyxoid fibroma: an evaluation of 11 patients

Kondromiksoid fibroma: 11 olgunun değerlendirilmesi

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Amaç: Kondromiksoid fibroma nedeniyle cerrahi tedavi uygulanan hastalar değerlendirildi.

Çalışma planı: Çalışmaya kondromiksoid fibroma tanısıyla cerrahi tedavi uygulanan 11 hasta (6 kadın, 5 erkek; ort. yaş 31; dağılım 8-53) alındı. En yaygın kemik tutulumu tibiada (3 hasta) görüldü. Yedi hastada histopatolojik tanı ameliyat öncesinde yapılan tru-cut biyopsi ile kondu. Tüm hastalarda kondromiksoid fibroma tanısı cerrahi sonrasında histopatolojik olarak doğrulandı. Radyografik incelemeye ek olarak, 10 hastada bilgisayarlı tomografi, altı hastada manyetik rezonans görüntülemeye başvuruldu. Cerrahi tedavi yöntemleri olarak, geniş rezeksiyon, marjinal eksizyon ve otojen greft, intralezyoner küretajı takiben otojen greft veya kemik çimentosu uygulandı. Hastalar ortalama 62.8 ay (dağılım 2-162 ay) takip edildi.

Sonuçlar: Hastaların tamamı ağrı yakınmasıyla başvurdu. Toraks duvarı ve tibiada lezyon olan birer hastada ek olarak kitle yakınması vardı. İki hastada görüntüleme yöntemleriyle yumuşak doku tutulumu saptandı. Üç hastada nüks gelişti (%27.3). Bu olgulara ilk ameliyatta ikisine sadece küretaj, diğerine küretaj ve iliyak greft yapılmıştı. El falanksında tümörü olan bir hasta, ikinci ameliyat yapıldıktan 46 ay sonra ağrı yakınmasıyla tekrar başvurdu. Tru-cut biyopsi ile sekonder kondrosarkom tanısı konan hastaya ray amputasyonu yapıldı. Cerrahiye bağlı olarak yara yeri enfeksiyonu görülmedi, hastalarda fonksiyon kaybı meydana gelmedi.

Çıkarımlar: İyi huylu bir kemik tömörü olan kondromiksoid fibroma vücudun değişik kemiklerinde ve değişik yaşlarda ortaya çıkabilir. Tedavide küretaj ve otogreft etkili bir yöntemdir.

Anahtar sözcükler: Kemik tümörü/tanı/cerrahi; kondroblastom/cerrahi; tibia.

Objectives: We evaluated surgical treatment of patients with chondromyxoid fibroma.

Methods: The study included 11 patients (6 females, 5 males; mean age 31 years; range 8 to 53 years) who underwent surgical treatment for chondromyxoid fibroma. The most common site of involvement was the tibia in three patients. Diagnosis was made preoperatively by tru-cut biopsies in seven patients and all the diagnoses were confirmed postoperatively by histopathologic examination. In addition to plain radiographs, computed tomography was used in 10 patients, and magnetic resonance was used in six patients. Surgery included wide resection, marginal excision or intralesional curettage followed by autologous bone graft or bone cement. The mean follow-up was 62.8 months (range 2 to 162 months).

Results: The main presenting symptom was pain in all the patients. Two patients with thoracic wall and tibia involvement, respectively, complained of a mass. Radiologic imaging showed soft tissue involvement in two patients. Recurrence occurred in three patients (27.3%), in whom initial surgical procedures were curettage alone (n=2) or with iliac graft (n=1). One patient with involvement in the phalanx of the thumb presented with pain 46 months after the second operation. A tru-cut biopsy yielded a diagnosis of secondary chondrosarcoma for which ray amputation was performed. No wound site infections or functional loss developed after surgical treatment.

Conclusions: Chondromyxoid fibroma may develop in various bones of the body and occur at a wide age range. Curettage with autologous bone graft is an effective surgical method.

Key words: Bone neoplasms/diagnosis/surgery; chondroblastoma/surgery; tibia.

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Chondromyxoid fibroma is a benign, very rarely seen bone tumor.[1-3] In its etiology, chromosome anomaly and immunologic factors have been proposed.[4] While diagnosis is generally made in patients under the age of 30, the disease may occur in a wide age distribution.[1-3,5] Its development is more aggressive in children.[6] It is generally seen in the metaphysis of long bones.[7] Flat bones, facial bones, and bones of the hand and foot are other sites affected. The most frequently involved bone is tibia.[2,8,9] The tumor is thought to originate from the physis cartilage. It is a bit more frequent in men than in women.[2-4,6] In the diagnosis, chondrosarcoma, chondroblastoma, fibrous dysplasia, non-ossifying fibroma, giant cell tumor, aneurysmal bone cyst and simple bone cyst should be kept in mind.[2,10-12] Especially when it is seen in old patients or when it develops in unexpected locations, an erroneous diagnosis of chondrosarcoma may be assigned.[13] It may rarely show malignant transformation.[6]

In long bones, en bloc excision of the tumor from the marginal border and grafting is the preferred method of treatment. When en bloc excision cannot be made, successful results can be obtained by employing curettage and bone grafting.[7] Due to the risk of malignancy, radiotherapy is not a preferred treatment.[3]

In this study, patients treated surgically due to chondromyxoid fibroma have been evaluated.

Patients and methods

During the period March 1986 - April 2006, 14 patients with the diagnosis of chondromyxoid fibroma were treated surgically. Three of the patients were lost to follow-up. In this study the treatment results of 11 patients (6 women, 5 men; mean age 31 years; range 8-53 years) were evaluated. In all patients at the time of diagnosis, age, physical findings, symptoms, lesion location and radiologic findings were recorded. In seven patients, histopathologic diagnosis was made by means of pre-operative tru-cut biopsy. In all patients, the diagnosis of chondromyxoid fibroma was post-operatively confirmed through histopathological examination.

The distribution of the lesions by location and the surgical treatment applied are shown in Table 1.

In addition to plain radiographs, computed tomography (CT) was used in 10 patients, and magnetic resonance imaging (MRI) in 6 patients. With the imaging methods, the presence or absence of cortical erosion and soft tissue spread was evaluated.

Surgical treatment

Surgical treatment methods included wide resection, marginal excision with autologous graft, and intralesional curettage followed by autologous graft or bone cement (Table 1).[14] To provide for a more effective curettage procedure, a high speed burr was used and the cavity walls were cauterized in most cases. Radiographic examinations were made during the operation.

The mean follow-up of eleven patients was 62.8 months (range 2 to 162 months). During follow-up the patients were examined on the basis of their complaints. Radiographic examinations were repeated after six weeks, three months and six months. Then, for the next two years x-ray examinations were made every six months, and thereafter annual examinations were made. If despite the operation the complaints of the patient continued and tumoral tis-

Table 1. Distribution of lesions by site and surgical treatment applied

Tumor Site	Number	Surgical Treatment
Tibia	3	Curettage + iliac graft
Acetabulum	1	Curettage + iliac graft
Iliac Bone	1	Curettage + bone cement
Calcaneus	1	Curettage + iliac graft; curettage + iliac graft after recurrence
1st metatarsal	1	Curettage + iliac graft
Phalanx (hand, 1st finger, proximal)	1	Curettage; en bloc resection + iliac graft after recurrence, ray amputation after secondary chondrosarcoma
Ulna	1	Curettage + iliac graft + costal graft
Thoracic wall (12th rib)	1	Partial rib resection
Fibula	1	Curettage; resection after recurrence

sue was detected with imaging methods, then recurrence was considered and histopathologically confirmed.

Results

All patients presented with a complaint of pain. As additional complaints, one patient had a mass on the thoracic wall and another had a mass on the tibia (Figure 1). In two patients, imaging methods showed soft tissue involvement. In three patients (27.3%) recurrence developed. In two of these patients, the first operation included only curettage, and the other curettage + iliac graft (Table 1). The patient with the tumor in the phalanx of the hand presented again with a complaint of pain at 46 months after the second operation. For the patient diagnosed with secondary chondrosarcoma confirmed by tru-cut biopsy, ray amputation was made. Surgical wound infection or loss of function due to surgery did not occur.

Discussion

Chondromyxoid fibroma is a benign bone tumor originating from the cartilage.[13] Though it is generally seen in patients under the age of 30, it can be encountered in a wide age distribution, as in our patients. It accounts for less than 1% of benign bone tumors.[1-3,5] It has been reported that it is seen somewhat more in men than in women.[2-4,6] In our patients, the number of women was higher.

The patients generally present with a complaint of chronic pain.[8] In some patients there may be problems related to movement of the joints.[6,15] In superficial tumors, swelling may also be seen.[3] All of our patients had a complaint of pain, and in two

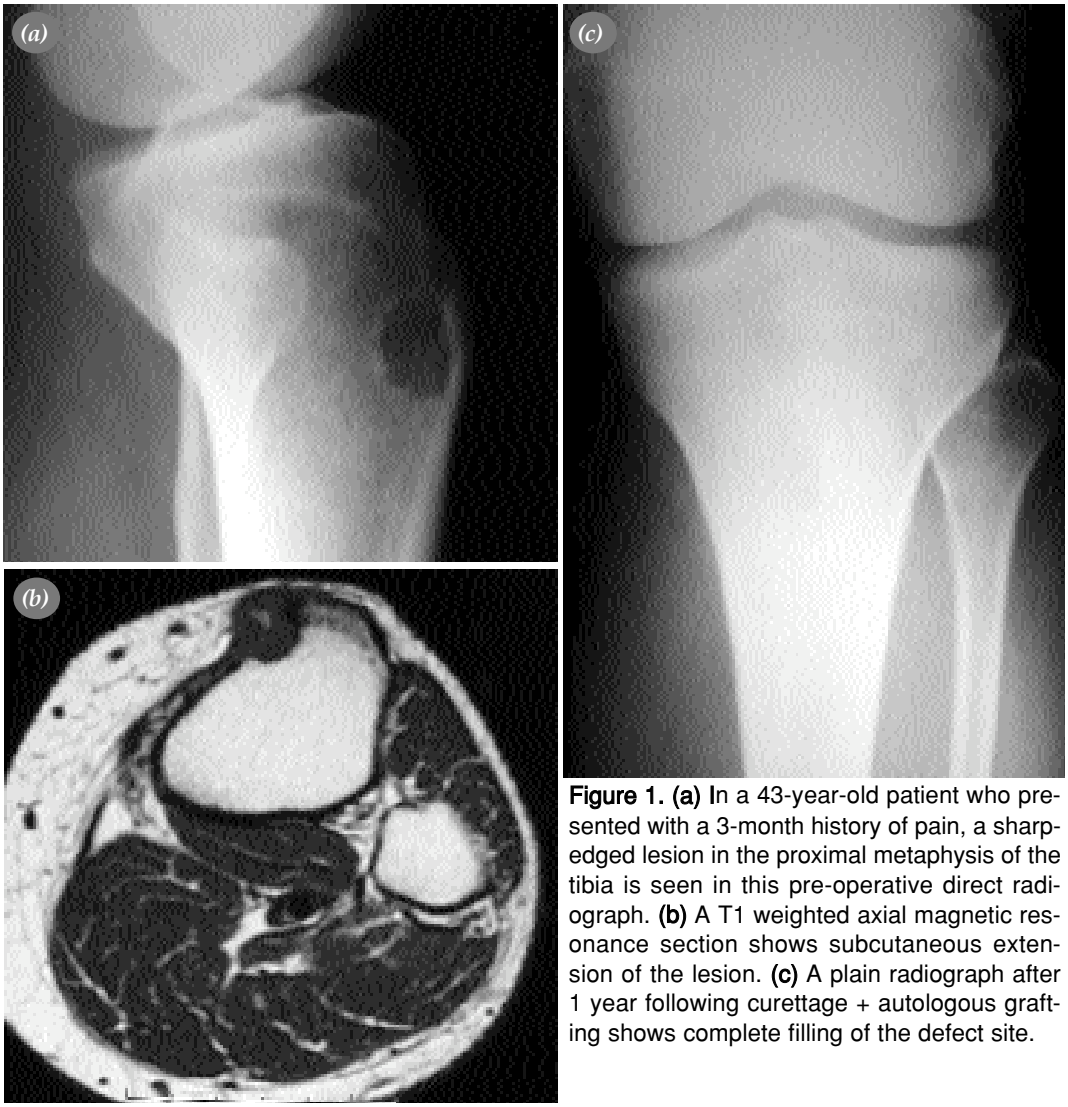

Figure 1. (a) In a 43-year-old patient who presented with a 3-month history of pain, a sharp-edged lesion in the proximal metaphysis of the tibia is seen in this pre-operative direct radiograph. **(b)** A T1 weighted axial magnetic resonance section shows subcutaneous extension of the lesion. **(c)** A plain radiograph after 1 year following curettage + autologous grafting shows complete filling of the defect site.

there was also a complaint of mass. Examination of a patient with acetabulum involvement revealed approximately 20 degrees of limitation in active hip flexion.

The lesion generally occurs in the metaphysis of long bones. It may affect different bones of the body and the most commonly involved bone is the tibia.[2,3,7,9] In our study too the lesions were found in a wide range and the most frequently affected site was the tibia.

On plain radiographs, cortical thinning and a lesion with sharp borders that causes expansion are generally seen. Magnetic resonance imaging and CT may demonstrate spread to soft tissue in some cases. Rarely it may locate on epiphysis and may be confused with chondroblastoma or giant cell tumor.[10] In very rare cases calcification can bee seen.[6] Though it generally develops eccentrically in the metaphysis of long bones [2] it shows a centric location in phalanx, metatarsals and metacarpals. Rarely, intracortical location has also been reported.[16] In chondromyxoid fibroma, periosteal reaction is not expected. In cases of periosteal reaction, fracture must be considered.[10] Radiographic examination of our patients revealed that borders of the lesions were distinct, that there was no periosteal reaction and that the cortex was generally expanded, that the tibial lesions were eccentrically located in metaphysis and that the lesions in hand and foot were centrally located. With the imaging methods, calcification was not detected. Computed tomography and MRI revealed soft tissue involvement in two patients.

In the literature, very few cases have shown transformation to malignancy.[2,17] In our study, secondary chondrosarcoma was found and ray amputation was performed in the follow-up of a patient who had been surgically treated previously for chondromyxoid fibroma in the proximal phalanx of the first finger of the right hand. Histopathologically, chondromyxoid fibroma may be confused with chondroblastoma or chondrosarcoma.[2,4,10,18]

In sacral involvement, generally there is progressive pain in the lower back and gluteal region and the symptoms may last for years. Since their clinical features are similar in this region, it is difficult to distinguish chondromyxoid fibroma from giant cell tumor or chondrosarcoma.[7] In one of our patients whose tumor was in the iliac wing, the complaints were initially thought to be of lumbar origin and different treatments were used in other centers. When the diagnosis was made two years later, the tumor had considerably enlarged. The patient was treated with intralesional curettage and bone cement, and during two years of follow-up recurrence has not been seen.

Wu et al.[2] in their study of 278 patients reported a recurrence rate of 25%. In other studies, recurrence has ranged from 7% to 80% depending on the treatment.[4,6,19] Gherlinzoni et al.[6] reported an 80% recurrence rate in patients treated with curettage alone, and a rate of 7% in patients treated with curettage + graft. Lersundi et al.[4] found a 50% recurrence rate in patients treated with curettage alone, and a rate of 10% in patients treated with curettage + graft or bone cement. In our study, the rate of recurrence was consistent with the literature and was 27.3%. Our preferred method of treatment was curettage + graft or en bloc resection and in the two patients who were treated with curettage alone (one at another center) recurrence was seen. Although our number of patients is not large enough for statistical comparison, we too believe that treatment with only curettage increases the risk of recurrence. Gherlinzoni et al.[6] explain the possible mechanism creating this difference as being the more effective curettage that is extended to the living spongious bone so that the graft can adhere.

As a conclusion, chondromyxoid fibroma can be seen in different bones of the body and in patients of different ages. Clinically and radiologically, it may be confused with other benign bone tumors, and for this reason the histopathological diagnosis is important. Curettage + autologous graft or en bloc resection is an effective method in the treatment of chondromyxoid fibroma.

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