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Clinical Outcomes after En Bloc Resection of Periosteal Chondroma: A Retrospective Clinical Study

Periosteal Kondromanın en blok Rezeksiyonu Sonrası Klinik Sonuçları: Geriye Dönük Klinik Çalışma

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ABSTRACT

Aim: Periosteal chondroma is a rare chondroma that is difficult to differentiate. Its localization is similar to other surface periosteal lesions. These lesions have a wide distribution of age. Curettage, marginal excision, or en bloc resection are applied in the surgical treatment. En bloc resection is preferred to reduce recurrence. In this study, we aimed to share the experience of two orthopedic oncology centers in the differential diagnosis and treatment of periosteal chondroma.

Material and Methods: Data from two clinics were analyzed retrospectively. Data were collected on demographic data (age, gender), clinical findings (pain, swelling, pressure-related symptom, duration of follow-up), radiological findings (size, bony invasion), pathology results (biopsy, excision), and postoperative complications (recurrence).

Results: Fourteen patients were included in the study. En bloc resection was performed in all cases. The mean age of the patients was 31.5±16.5 (range, 8-58) years. 10 (71.4%) patients were male. The mean duration of symptoms was 6.6±4.8 (range, 0-18) months, and the mean follow-up was 46.7±39.6 (range, 6-132) months. Nine (64.3%) patients had pain. Six (42.9%) patients had swelling. One patient (7.1%) had a palpable mass. There was no complaint in 1 (7.1%) patient. One (7.1%) patient underwent biopsy. During the follow-up, no recurrence or complication was observed after en bloc resection.

Conclusion: Imaging and histopathological findings of benign and malignant periosteal chondroid tumors may overlap, and accurate differential diagnosis is crucial in the treatment of these lesions. En bloc resection prevents recurrence during follow-up.

Keywords: En bloc resection; periosteal chondroma; chondrosarcoma; recurrence.

ÖZ

Amaç: Periosteal kondroma oldukça nadir gözlenen ve ayırıcı tanısı zor bir kondroid tümördür. Yerleşimi diğer yüzeyel periosteal lezyonlar ile benzerlik gösterir. Bu lezyonlar farklı yaş gruplarında görülmektedir. Cerrahi tedavisinde küretaj, marjinal eksizyon veya en blok rezeksiyon uygulanmaktadır. Nüksü azaltmak amacıyla en blok rezeksiyon tercih edilir. Bu çalışmada, periosteal kondromanın ayırıcı tanı ve tedavisinde iki ortopedik onkoloji merkezinin tecrübesinin aktarılması amaçlanmıştır.

Gereç ve Yöntemler: İki kliniğe ait veriler geriye dönük olarak incelendi. Demografik veriler (yaş, cinsiyet), klinik bulgular (ağrı, şişlik, basıya bağlı semptom, takip süresi), radyolojik bulgular (kitle büyüklüğü, kemik invazyonu), patoloji sonuçları (biyopsi, eksizyon) ve ameliyat sonrası komplikasyonlar (nüks) hakkında veri toplandı.

Bulgular: Çalışmaya 14 hasta dahil edildi. Tüm vakalarda en blok rezeksiyon uygulandı. Hastaların ortalama yaşı 31,5±16,5 (aralık, 8-58) yıl idi. 10 (%71,4) hasta erkek cinsiyetti. Ortalama şikayet süresi 6,6±4,8 (aralık, 0-18) ay, ortalama takip süresi ise 46,7±39,6 (aralık, 6-132) ay idi. Dokuz (%64,3) hastada ağrı şikayeti mevcuttu. Altı (%42,9) hastada şişlik şikayeti mevcuttu. Bir (%7,1) hastada palpe edilebilen bir kitle mevcuttu. Bir (%7,1) hastada şikayet bulunmuyordu. Bir (%7,1) hastaya biyopsi yapıldı. Takip süresince nüks veya en blok rezeksiyon sonrasında herhangi bir komplikasyon görülmedi.

Sonuç: Benign ve malign periosteal kondroid tümörlerin görüntüleme ve histopatolojik bulguları çakışabilir ve bu lezyonların tedavisinde, ayırıcı tanının doğru yapılması oldukça önem arz eder. En blok rezeksiyon takip sırasında nüksü önlemektedir.

Anahtar kelimeler: En blok rezeksiyon; periosteal kondroma; kondrosarkom; nüks.

INTRODUCTION

Periosteal chondroma is a rare benign cartilaginous lesion. It is very rare and is not usually involved in the differential diagnosis of chondromatous lesions (1). It was first defined by Liechtenstein (2) and then by Jaffe (3).

These tumors originate from the periosteal surface. The differential diagnosis is challenging as many other benign and malignant chondroid lesions can be mistaken, located at the periosteal surface. These entities comprise periosteal chondrosarcoma, periosteal osteosarcoma, and surface high-grade osteosarcoma, osteosarcoma, cortical desmoid, non -epiphyseal chondroblastoma. The most common frequent sites are metaphyses, metadiaphyses of long bones (femur, tibia, and humerus). Atypical lesions can also be encountered at the spine and the rib (4).

These lesions are generally 3 cm in size (5). In lesions bigger than 7 cm, malignancy should be suspected. Periosteal chondroma and malignant chondroid lesions can spread to extracompartmantal areas (6). Palpable painful mass is the most common symptom. However, patients can present with asymptomatic lesions. Radiographic images usually have well-defined borders with marginated erosions and endosteal scalloping. The imaging feature may resemble both benign and malignant lesions. So, biopsy does not help in making a definite diagnosis. Pathologic analysis revealed nuclear pleomorphism and binucleation. These findings may sometimes lead to misdiagnosis as chondrosarcomas. Hyaline cartilage demarcated by the periosteum is a typical finding. There is a narrow transition zone between the soft tissue and the periosteal chondroma. There is no invasion of the underlying bone with no atypical mitotic figures. En bloc resection is safe in terms of prevention of recurrence and improvement in symptoms.

In this study, we aimed to share the clinical experience of two centers in the surgical treatment of periosteal chondromas.

MATERIAL AND METHODS

The study was approved by the Ethics Committee of İstanbul Medeniyet University (2021/285). A retrospective review was performed in two clinics between 2015 and 2021. A total of 14 cases were identified. A prior diagnosis was made based on clinical and radiological findings. Final histopathologic evaluation by two pathology experts in musculoskeletal oncology verified the diagnosis. Inclusion criteria included patients with complete data at least one year of follow-up, lesions of appendicular skeleton involving upper and lower extremities. Exclusion criteria included incomplete patient data, lesions of axial skeleton, final pathological diagnosis of malignancy. In all lesions, surgical excision was performed using direct approach over the lesion under general anesthesia.

Statistical Analysis

Descriptive statistics were given as mean, standard deviation and range values. Categorical variables were summarized as numbers and percentages.

RESULTS

The demographic and clinical findings of patients were presented in Table 1. 14 patients were included in this study. All cases underwent en bloc resection. The mean age of the patients was 31.5±16.5 (range, 8-58) years, and 10 (71.4%) patients were male. The mean duration of symptoms was 6.6±4.8 (range, 0-18) months. The mean follow-up was 46.7±39.6 (range, 6-132) months. Symptoms included pain in 9 (64.3%) patients and swelling in 6 (42.9%) patients. One (%7.1) patient had a palpable mass. One (%7.1) patient was asymptomatic. Lesions were located at distal femur in 4 (28.6%) patients, hand in 4 (28.6%) patients, metatarsal in 3 (21.4%) patients, tibia in 1 (%7.1) patient, calcaneus in 1 (%7.1) patient, and humerus in 1 (%7.1) patient. The size of the tumor was more than 3 cm in all lesions. Preoperative biopsy was made in only 1 (%7.1) patient with a lesion more than 7 cm in size. There was no recurrence and no other complication after en bloc resection. Radiologic MRI features were well-defined lesions with a sharp sclerotic margin, scalloping of the cortex, and multilobular mass without bone marrow invasion. The lesions predominantly show high signal intensity on T2-weighted images and low signal intensity on T1-weighted images (Figure 1, 2). Pathologic specimens demonstrated double-nucleated cells with moderate myxoid changes in the matrix consistent with periosteal chondroma (Figure 3).

Table 1. Demographic and clinical findings of all cases

Case	Age	Gender	Localization	Symptom	Duration of symptom	Follow-up
1	58	Male	left third metatars	(-)	(-)	36
2	28	Male	left second finger	swelling	6	36
3	55	Male	left fourth finger	firm immobile sensitive mass	7	48
4	26	Female	right second metatars	pain	3	24
5	22	Male	left posterior knee	pain and sensitivity	12	12
6	27	Male	right proximal humerus	night pain	1	24
7	13	Female	left distal tibia	pain	18	24
8	21	Male	left fifth finger	swelling	5	120
9	20	Male	right fourth finger	swelling	3	132
10	48	Female	left proximal humerus	pain	7	36
11	55	Male	left calcaneus	pain and sensitivity	12	96
12	21	Male	distal femur	pain and swelling	5	37
13	8	Male	distal femur	pain and swelling	8	24
14	39	Female	left distal femur	pain and swelling	6	6

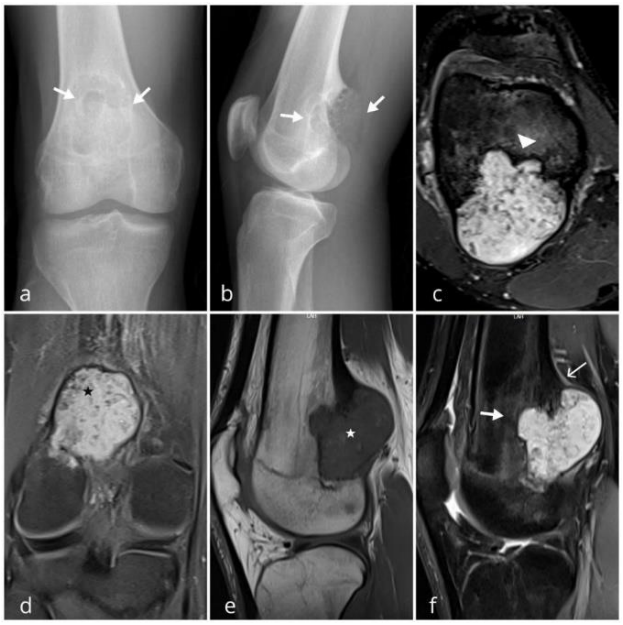

Figure 1. Radiologic features of periosteal chondroma located at distal femur (Case 12). **a, b)** Anteroposterior and lateral radiograph of right femur shows periosteal-based lesion with extrinsic scalloping of posterolateral cortex of distal femoral metaphysis (arrows) There is internal chondroid calcification. **c, d, f)** Axial, coronal and sagittal fat-supressed T2-weighted MR images show multilobular, predominantly high signal intensity mass (asteriks and arrow) along posterior cortex of distal femur without bone marrow invasion (arrow head). No adjacent soft tissue edema is seen. **e)** Sagittal T1-weighted MR image shows low signal intensity soft tissue mass causing cortical scalloping (arrow)

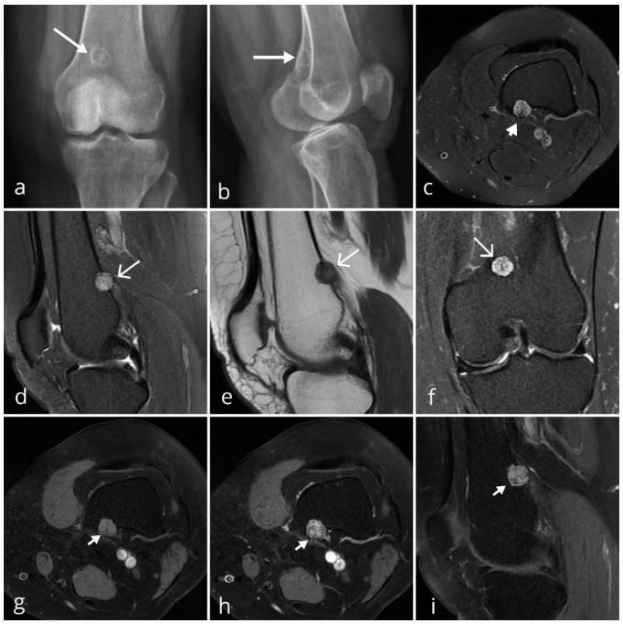

Figure 2. Radiologic features of periosteal chondroma located at distal femur (Case 14). **a, b)** Anteroposterior and lateral radiograph of left femur shows jukstracortical well-defined lesion with sharp sclerotic margin and extrinsic scalloping of posterolateral cortex of distal femoral metaphysis(arrows). **c, d, f)** Axial, coronal and sagittal fat-supressed T2-weighted MR images show high signal intensity mass (arrows) along posterior cortex of distal femur without bone marrow invasion. No adjacent soft tissue edema is seen. **e)** Sagittal T1-weighted MR image shows low signal intensity soft tissue mass causing cortical scalloping (arrow) **g)** Precontrast axial fat-supressed T1-weighted image show mildly intens signal in lesion. Contrast enhanced fat-suppressed T1-weighted image demonstrates internal patchy enhancement (arrows)

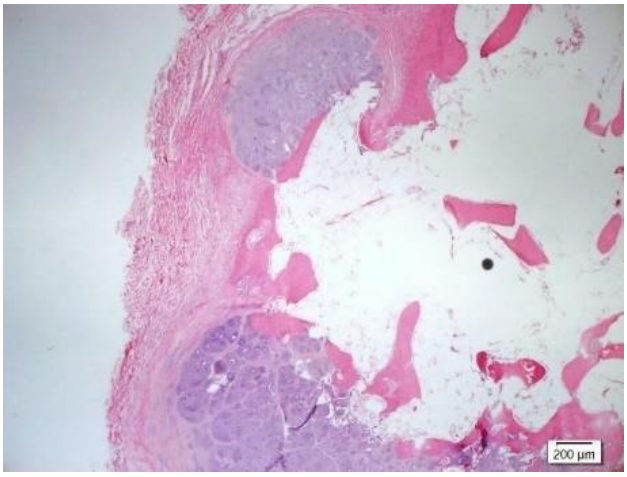

Figure 3. Radiologic features of periosteal chondroma located at distal femur (Case 13). Lobes of hyaline cartilage surrounded by fibrous connective tissue or cortical bone form the periosteal chondroma. This tumor is well-circumscribed. It does not penetrate into the bone and permeate the surrounding soft tissues. The cartilage is normocellular and the nuclei are plump and hyperchromatic. Double-nucleated cells are common, and moderate myxoid changes may be seen in the matrix (H&E, x40)

DISCUSSION

The diagnosis of periosteal chondroma is usually problematic and need clinical, radiological, and pathological correlation. In this study, we performed en bloc resection in 14 cases. At a mean of 4 years of follow-up, we were unable to observe any recurrence.

The surgical treatment in periosteal chondroma is usually marginal excision or en bloc resection with wide margins. However, intralesional curettage has the risk of local recurrence. Studies regarding outcomes after surgical treatment are usually limited to case reports and case series as it is a rare entity.

Previous studies demonstrated satisfactory clinical outcomes after excision. Boriani et al. (7) performed marginal or wide resection in 20 cases. Most lesions were located at the proximal metaphysis of long bones. Five lesions were located in the hand, which was also common in our study. Wide excision was done in three cases. He found that marginal excision is efficient in most cases. In atypical lesions except for the metaphysis of long bones, excision is a preferred treatment. Motififard et al. (8) reported a pelvic periosteal chondroma in a 39 year-old male with paresthesia, gluteal muscle atrophy, and claudication. He performed marginal excision. At six months after surgery, the patient reported clinical improvement. Samaddar et al. (9) performed second rib wide resection in a 12-year-old female child due to periosteal chondroma. The follow-up was not given, but the patient was reported to be good. Kang et al. (10) performed wide excision and left T5/6 hemilaminectomy in a 41-year-old male due to periosteal chondroma in the thoracic spinal canal. At 18 months of follow-up, there was no recurrence. Pandey et al. (11) observed no recurrence in radial diaphyseal lesion at two years after marginal excision of periosteal chondroma. Nishio et al. (12) performed excision in a 25-year-old female with a distal tibial lesion. At four months of follow-up, the patient was satisfied with no signs of recurrence. Debbarma et al. (13)

performed a subtotal scapulectomy in a 24-year-old male due to periosteal chondroma at the right scapula. At 1-year follow-up, the patient had an excellent outcome.

Rolvien et al. (14) performed en bloc resection in a periosteal chondroma of cuboid. At 9 months after surgery, the patient had no symptoms with good functional outcomes. Zheng et al. (15) made en bloc resection of distal femur periosteal chondroma in a 14-year-old female. At 6 months after resection, the patient was well and the fibular graft was well incorporated with complete healing of the defect. These studies demonstrated that en bloc resection provides satisfactory outcomes without recurrence.

Intralesional curettage is another option in the management of periosteal chondromas. Imura et al. (5) performed intralesional curettage, bone grafting, and plate fixation in the distal femoral lesion in a 17-year-old boy. There was no recurrence at 12 months after resection. However, intralesional curettage can present with recurrence in one year after initial surgery. In their case series involving 24 hand periosteal chondromas, Rabarin et al. (16) reported recurrence in a 10-year-old child, 10 months after curettage.

After resection or curettage of the lesion, cement or bone graft has been used to fill the defect in previous studies. Imura et al. (5) used bone graft after curettage of chondroma of the distal femur. At 6 months after surgery, the graft was consolidated. Rolvien et al. (14) used cement after en bloc resection of cuboidal periosteal chondroma. In our study, we did not use any bone void filler without concern about biomechanical stability, and the defect was healed uneventfully.

In terms of clinical complaints, localized swelling and pain are common findings in periosteal chondroma, and the clinical course facilitates the diagnosis of periosteal chondroma. Swelling followed by moderate pain is characteristic (7). In our series, 12 (%85.7) patients had a history of pain and swelling for 3-6 months which was comparable to previous studies (7,8). However, there are also cases who had symptoms for two years as reported by Pandey et al. (11)

Age at diagnosis is variable in previous studies. In our study, the mean age at diagnosis was 31.5 years. In series of Boriani et al. (7), the mean age was 22.15 years with a mean duration of symptoms of 15 months. In other studies, the age at diagnosis ranged between 12-41 years (5,8-12). Similarly, our study also confirmed that periosteal chondromas can be seen in patients with a wide range of age.

Imaging can help in the identification of periosteal chondroma and differentiation from other chondroid lesions. Lesion size with more than 3 cm increases the likelihood of chondrosarcoma. Although the size of lesions was more than 3 cm in some of our cases, a radiological appearance with sharp sclerotic margin, chondroid calcification, and extrinsic scalloping of cortex without adjacent soft tissue edema supports the diagnosis of periosteal chondroma. One recent study (17) indicated that PET-CT could aid in distinguishing chondromas from chondrosarcomas with a cut-off SUVmax value of 2.0. This is yet to be evaluated in other studies. Pathological clues depend on whether the lesion's microscopy demonstrated osteoid (osteosarcoma) or chondroid matrix (periosteal chondroma). In our cases, there was no peripheral ossification present which is possible in periosteal osteosarcomas.

Observation can be preferred in a painless periosteal chondroma. We did not encounter any morbidity at the affected extremity. However, if the size of the lesion exceeds more than 3 cm, it is reasonable to consider that periosteal chondroma should be approached like a malign tumor and en bloc resection without cementation or grafting can be preferred.

CONCLUSION

This study confirms the importance of differential diagnosis and wide excision in periosteal chondromas. These lesions have a wide distribution of age. Its localization is similar to other surface periosteal lesions, which is why the identification from malignant chondroid lesions is challenging.

Ethics Committee Approval: The study was approved by the Ethics Committee of İstanbul Medeniyet University Göztepe Training and Research Hospital (26.05.2021, 285).

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