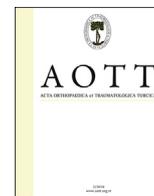


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Parosteal lipoma as a rare cause of peripheral neuropathy and local irritation: A report of 12 cases

Kerem Başarir^a, Ercan Şahin^{b,*}, Mahmut Kalem^a, Mustafa Onur Karaca^c, Yusuf Yildiz^a, Yener Sağlık^a^a Ankara University, Faculty of Medicine, Department of Orthopedics & Traumatology, Ankara, Turkey^b Bülent Ecevit University, Faculty of Medicine, Department of Orthopedics & Traumatology, Zonguldak, Turkey^c Simav State Hospital, Department of Orthopedics & Traumatology, Kütahya, Turkey

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ABSTRACT

Objective: The aim of this study was to evaluate the clinical features and functional results of patients with parosteal lipomas.**Methods:** A total of 12 patients (8 females and 4 males; mean age: 45 (10–62) years) with parosteal lipomas who were treated between April 1986 and April 2014, were included into the study. The medical records of the patients were reviewed to analyze the clinical features and functional results of the patients.**Results:** Of the 12 lipomas, 5 were localized in the proximal arm, 4 in the forearm, 1 in the distal arm, 1 in the distal thigh and 1 in the distal tibia. All patients presented with a progressive, slow-growing mass that was associated with thumb extension weakness in 1 case, and brachialgia-like symptoms in 1 case. Plain radiographs showed a juxtacortical mass in all cases and irregular ossification in 3 cases. In all cases, marginal excision was performed and no clinical recurrence was observed after a mean follow-up of 16 months.**Conclusion:** Parosteal lipomas are uncommon tumors that can be diagnosed with their characteristic radiological features. Parosteal lipomas occurring in the proximal radius may easily cause paralysis of the posterior interosseous nerve or muscle weakness.**Level of Evidence:** Level IV, Therapeutic study© 2017 Turkish Association of Orthopaedics and Traumatology. Publishing services by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Parosteal lipoma is rare lesion, accounting for less than 0.3% of all lipomas. Although histopathological characteristics are essentially the same as more common subcutaneous lipomas, distinctive feature is intimate relationship with the surface of the bone.¹ The most frequently affected sites are the diaphyseal and metaphyseal regions of the long bones and this type of tumor tends to be indolent due to deep localization.² However, it may cause nerve compression in certain anatomical regions where neurovascular structures remain close to the bone, including the proximal radius.^{3,4} From an oncological standpoint, as subcutaneous lipoma,

it is benign lesion. Clinical significance is that this rare lesion should be considered in differential diagnosis of atraumatic nerve compression and radiolucent soft tissue mass with osseous changes adjacent to the lesion.^{1,5} Taking parosteal lipoma into consideration can result in use of most appropriate imaging modality, and prompt decompression of the nerve can be applied while avoiding aggressive surgery.⁵ The clinical and imaging characteristics of 12 rare cases of parosteal lipoma are presented in this paper.

Patients and methods

A retrospective review of medical records and images of 12 patients with confirmed parosteal lipoma treated surgically between April 1986 and April 2014 was conducted. Diagnoses were confirmed with histological examination of excised lesion in all patients. Data obtained for each patient included age, gender,

* Corresponding author. Bülent Ecevit University, Faculty of Medicine, Department of Orthopedics & Traumatology, Esenköy-Kozlu, P.B.: 67100, Zonguldak, Turkey.

E-mail address: dr_erc_sah@yahoo.com.tr (E. Şahin).

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Table 1
Clinicopathological characteristics of parosteal lipoma in 12 patients.

Case no	Age (years)	Sex	Site	Symptoms	Size (cm)	Duration of symptoms (months)	Diagnostic work-up	Follow-up (months)
1	46	M	R-prox humerus	Pain, swelling	13 × 8 × 8	60	MRI	12
2	10	F	R-distal tibia	Swelling	6 × 6 × 6	12	MRI	12
3	40	M	L-prox forearm	Swelling	7 × 5 × 5	36	CT/MRI	12
4	50	F	L-radius prox exostosis	Swelling	9 × 6 × 4	84	MRI	48
5	39	F	R-forearm middle interosseous membrane both sides	Pain, swelling, PIN muscle weakness	10 × 3 × 3 6 × 3 × 3	12	MRI	16
6	46	F	R-humerus distal	Swelling	10 × 6 × 3	18	MRI	12
7	62	F	R-humerus prox	Pain, swelling	6 × 4 × 1	240	CT/MRI	12
8	33	M	R-forearm prox interosseous membrane both sides	Pain, swelling	7 × 5 × 3 6 × 3 × 2	60	MRI	12
9	46	F	L-humerus prox	Pain, swelling	10 × 8 × 4	24	MRI	12
10	57	F	R-thigh distal	Pain, swelling	10 × 8 × 6	96	MRI	12
11	56	F	L-humerus prox	Pain, swelling	5 × 5 × 7	3	MRI	12
12	60	M	R-humerus prox	Numbness in shoulder	10 × 7 × 5	4	MRI	24

CT: Computed tomography; F: Female; L: Left; M: Male; MRI: Magnetic resonance imaging; Prox: Proximal; R: Right; PIN: Posterior interosseous nerve neuropathy.

symptoms at presentation, skeletal location, radiological features, diagnosis, and treatment. Radiological studies performed included conventional radiography ($n = 13$), computed tomography (CT) ($n = 2$), and magnetic resonance imaging (MRI) ($n = 3$). CT or MRI determined anatomic location and relationship to surrounding structures before surgery better than X-ray. CT or MRI was assessed for lesion homogeneity, size and border definition, relationship to the neurovascular bundle, and relationship to the bone. CT images were assessed for density, and MRI for signal intensity on T1-weighted, T2-weighted, short-tau inversion recovery (STIR) T2, and post-contrast T1 STIR images. Systematic biopsy was not performed when lipoma had specific imaging features on CT or MRI (well-circumscribed homogeneous fatty mass; no or rare, thin septation; no post-contrast enhancement). All patients underwent surgery with marginal excision (shelling out) and osteotomy-resection (when needed) of the bone because these well-circumscribed lipomas are well encapsulated and are separated easily from the surrounding tissues, in contrast to infiltrative lipoma and lipoma-like well-differentiated liposarcoma. Post-operative histopathological examination confirmed diagnosis of lipoma and healthy surgical margin of the tumor in all cases. Patients were followed-up for minimum of 12 months.

Results

The data are summarized in Table 1. The patient group comprised 8 females and 4 males in age range of 10–62 years (<18 years = 1 patient), and mean age at time of treatment was 45 years. Most common site was the proximal arm (5 lipomas),

followed by the forearm (4: 3 proximal, 1 middle). Mean interval from onset of symptoms to surgical treatment was 54 months (range: 3–240 months). In 5 patients, complaint was of swelling only. One patient had swelling, pain, and generalized numbness (brachialgia/thoracic outlet-like symptoms) in the shoulder (Case 12), 1 had pain and swelling with muscle weakness (Case 5), and 6 patients had pain with swelling. Radiographs showed juxtacortical mass with normal bone appearance in 5 patients (Fig. 1), juxtacortical mass with hyperostosis in 3, juxtacortical mass and irregular osseous protuberance with radiolucent cap (exostosis) in 1 (Case 3) (Fig. 2) and irregular ossification in 1 (Case 4) (Fig. 1A). One patient was aged 10 years and had mass adjacent to the distal tibia (Fig. 3). Two of the patients had lipomas on both sides of the intramembranous membrane in the forearm, and 1 of these had complaints of muscle weakness with the thumb interphalangeal joint in extension (Case 5) (Fig. 4). One of the patients was surgically treated because of local recurrence in the distal arm (first surgery 2 years previously at another institution) (Case 6). T1-weighted MRI indicated high-intensity signal, identical to that of normal fat tissue. All lipomas were extirpated surgically by marginal resection (Fig. 5) and osteotomy or resection of bone cortex. Five of the lipomas were adjacent to the neurovascular bundle (1 with the brachial plexus, 4 with the radial nerve) and needed careful dissection to avoid injuring nearby neurovascular structures. Lipomas had mean size of 10 cm, ranging from 6 cm to 13 cm. The patient with radial nerve motor weakness recovered completely at 8 weeks postoperatively. At mean 16 months (range: 12–48 months) after resection, no complication or recurrence was recorded.



Fig. 1. Radiographs demonstrating juxtacortical mass adjacent to the bone. (A) Lateral radiograph of forearm illustrating juxtacortical radiolucent mass surrounding the hyperostosis and irregular ossification (Case 4). (B) Forearm (Case 5). (C) Humerus (Case 9).

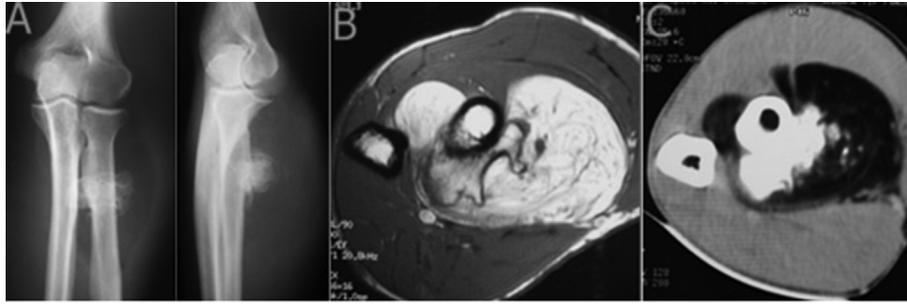


Fig. 2. Case 3. (A) Radiographs depicting juxtacortical mass and irregular osseous protuberance (exostosis). (B) T1-weighted axial (TR/TE: 600/11) magnetic resonance imaging revealed high-intensity lesion corresponding to fat adjacent to the forearm with osseous protuberance with radiolucent cap. (C) Computed tomography scan of the forearm shows well-defined hypodense mass with thin septa. Attenuation values within the lesion were equal to those of adipose tissue. Osseous part of the lesion around the radius is seen.

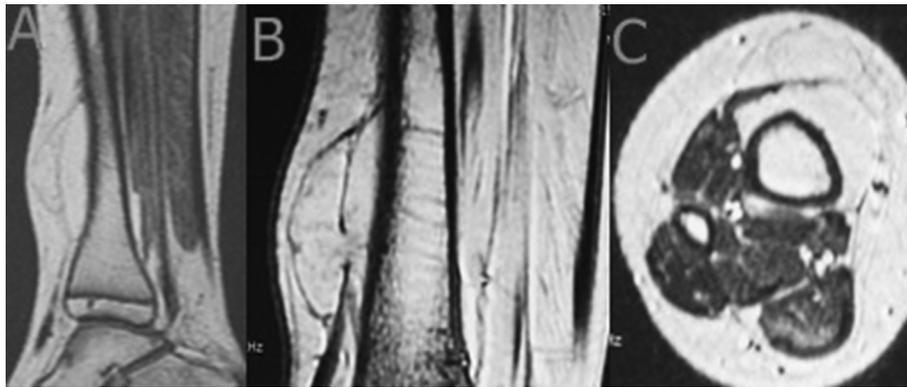


Fig. 3. (A) T1-weighted sagittal (TR/TE: 500/15) magnetic resonance image (MRI) demonstrating high-intensity lesion corresponding to fat adjacent to the tibia in a 10-year-old patient (Case 2). (B) T1-weighted coronal (TR/TE: 740/20) MRI (Case 2). (C) T2-weighted axial (TR/TE: 6000/92) MRI (Case 2).

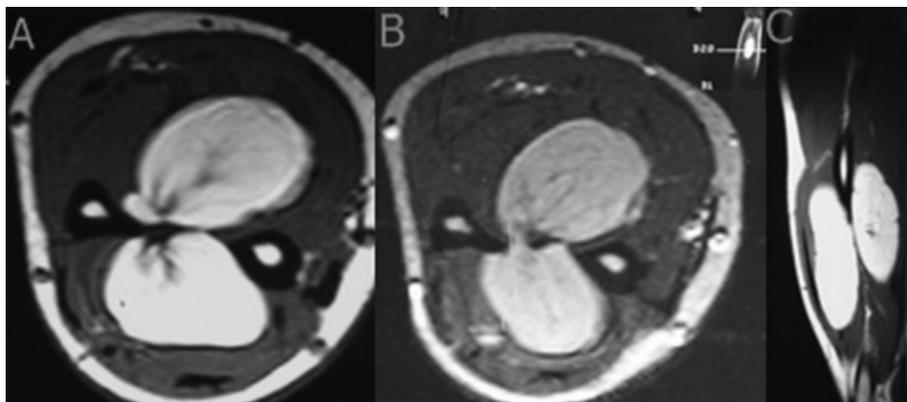


Fig. 4. (A) T2-weighted axial (TR/TE: 2700/60) magnetic resonance image (MRI) demonstrating high-intensity lesion corresponding to fat adjacent to the radius and intermuscular septum (Case 5). (B) T1-weighted axial (TR/TE: 660/20) MRI indicating relationship through the intermuscular septum (Case 5). (C) T1-weighted sagittal (TR/TE: 660/20) MRI showing bilobulated mass in the forearm (Case 5).

Discussion

The first description of this neoplasia was published in German medical literature by Seerig in 1836 (as reported by Fleming et al).¹ The subject was approached in English medical literature in 1886 by Smith.⁶ However, the term parosteal lipoma was introduced by Power in 1888 and is still used.⁷ Parosteal lipoma is unusual neoplasm that appears to emerge from multidirectional mesenchymal “modulation” within the periosteum. These tumors have been described as “periosteal lipomas,” “chondrolipomas of soft tissue” and “lipomas of nerves,” but they are most commonly

believed to originate from the periosteum. Sub-classification is divided into 4 variants: (I) no ossification, (II) pedunculated exostosis, (III) sessile exostosis, and (IV) patchy chondro-osseous modulation.²

Imaging features are usually distinctive. The bone may be normal even when near the periosteum.^{1,8} Juxtacortical mass is usually seen on radiographs. Occasionally, these lesions are associated with reactive changes in the adjacent bone,^{1,8,9} such as cortical erosion, deformity and hyperostosis.^{1,8,10,11} Most parosteal lipomas with bone reactions are associated with hyperostosis.¹ Prominent hyperostosis promotes a typical appearance that, when

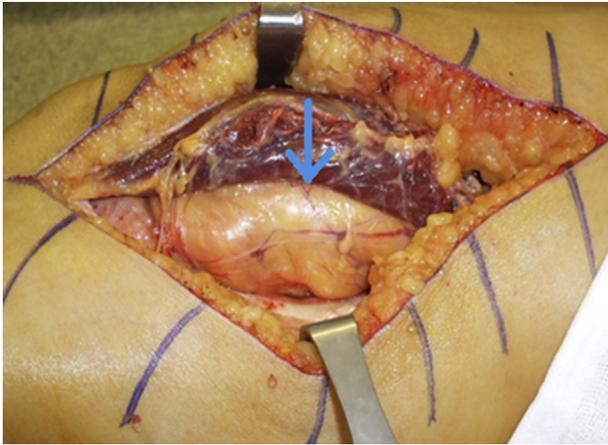


Fig. 5. Intraoperative photograph illustrating encapsulated lipoma (blue arrow) adjacent to the femur.

combined the radiolucency of the soft parts of the tumor, has been described as pathognomonic for parosteal lipoma.^{1,12} Rarely, parosteal lipomas arise from an exostosis, as in Case 3 of this series.¹⁴

Parosteal lipoma is characterized by non-tender, immobile, slow-growing mass over bone that is not fixed to skin. Other symptoms depend on volume and location of the tumor and effects of local pressure on adjacent nerves.¹³ As seen in Case 5 of this series, nerve compression is more common with proximal forearm lipoma adjacent to the neck of the radius, causing radial nerve compression with pain, paralysis of finger extension, or paresthesia.^{14,15,16,17} In the shoulder, lipoma can cause pain, such as brachialgia simulating thoracic outlet syndrome, as in Case 12.^{4,18}

On CT and MRI, parosteal lipoma is characterized by homogeneous lobulated appearance and is adhered to surface of the adjacent bone.^{5,19} MRI is considered superior to CT for evaluation of parosteal lipoma. Tumor is identified on MRI as juxtacortical mass with signal intensity identical to that of subcutaneous fat, regardless of pulse sequence. Heterogeneity in these lesions is invariably present and corresponds to pathological components in the lesion. Areas with intermediate signal intensity on T1-weighted images that are high signal intensity on T2-weighted images represent cartilaginous components in parosteal lipoma. Fibrovascular septa may cause lobulated appearance of fat component, with low-signal-intensity strands on T1-weighted images that become higher in signal intensity on long TR images (particularly with fat suppression). Larger areas of bone production surrounded by lipomatous components are also well demonstrated with MRI. Adjacent muscle atrophy, poorly demonstrated by CT, is identified on MRI as increased striations of fat in the affected muscle and is caused by associated nerve entrapment. This finding is best appreciated on T2-weighted images because of decreased signal intensity of normal muscle relative to fat. Finally, MRI best demonstrates the relationship of the tumor to the underlying native bone and muscle, and this information is important for surgical planning as parosteal lipoma is usually firmly adhered to the underlying cortex at the site of surface bone production.¹⁹

Treatment of parosteal lipoma is surgical resection. When there is nerve entrapment, the tumor must be removed and separated from the lipoma before irreversible muscle atrophy develops in order to be able to maintain function. Parosteal lipoma is characteristically encapsulated and strongly fixed to the underlying periosteum. Site where the tumor is strongly attached to the bone is the

area of most prominent osseous proliferation. Either subperiosteal dissection or use of an osteotomy to separate the lesion from underlying bone, or segmental resection of bone is required as result this characteristic.^{17,19,20} Local recurrence is unusual, but has been reported (Case 6).¹ Malignant transformation has not been reported.

Conclusion

In this paper, 12 cases of histopathologically confirmed parosteal lipoma were reported. The characteristic features allow diagnosis on radiographs, and MRI also provides excellent delineation of extent of the tumor, thereby aiding surgical planning. It was unusual to determine this entity in a 10-year old patient (Case 2) and local recurrence is also very rare. It was seen that parosteal lipoma in the proximal radius may easily cause paralysis of the posterior interosseous nerve.

Conflicts of interest

Each author certifies that he or she has no commercial associations (e.g., consultancies, stock ownership, equity interest, patent/licensing arrangements, etc.) that might pose a conflict of interest in connection with the submitted article.

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