Parosteal lipoma

Report of 15 new cases and a review of the literature

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Parosteal lipoma. Report of 15 new cases and a review of the literature

The lipoma is the most common tumor of the body and can be found in virtually every tissue or organ. However, a parosteal lipoma is a rare benign fatty neoplasm having an intimate relationship to the periosteum. The incidence of this tumor is 0.3% of all lipomas. Over 150 cases of parosteal lipoma have previously been described in the literature. Owing to the rarity of this condition and the difficulties encountered in its diagnosis and treatment, we wish to report fifteen new cases of parosteal lipoma.
We reviewed the clinical records of 15 patients who underwent surgery to remove a parosteal lipoma between November 2003 and July 2009. The diagnosis of a parosteal lipoma was made by the histological findings, the confirmation of fat content at Magnetic Resonance or Computed Tomography.
Surgery in all the cases entailed resecting the tumor with parosteal excrescence. In three cases with hyperostosis, a further exeresis of the bone was performed.
Parosteal lipomas are rare entities associated with periosteal involvement depending on their location. Current management should include computed tomographic scanning and magnetic resonance. Surgery, which is mandatory treatment, requires particular attention to ensure that any periosteal involvement is removed completely.

KEY WORDS: Computed Tomography, Diagnosis, Magnetic Resonance, Parosteal lipoma, Surgical treatment

Introduction

Parosteal lipoma is an extremely rare benign neoplasm consisting mainly of mature adipose tissue contiguous to the underlying periosteal bone [1]. Described in 1836 by Seering [2], the lesion was initially referred to as "periosteal lipoma". The term "parosteal lipoma" was suggested by Power in 1888 [3] to indicate that the lesion does not arise in the periosteum because the periosteum does not contain any fat cells. Parosteal lipoma is considered to be one of the rarest primary bone tumours. It is composed of mature fat cells and varying, though small, quantities of fibrous and vascular tissue. At the gross pathological examination, parosteal lipomas appear as greasy yellowish masses that adhere to the underlying periosteum. They are, like soft-tissue lipomas, composed of mature adult fat [1,4,5]. This tumor accounts for 0.3% of all lipomas [6]. These lesions are closely related to the periosteum; indeed, approximately 50% are associated with an underlying osseous reaction [7]. The most frequent complaints are a tumoral convexity, presenting as a visible or palpable mass, or mild pain [8].

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The most common sites of origin of this neoplasia are in the thigh adjacent to the femur or in the upper extremity near the proximal radius [4,7,9]. The lesion has also been reported in the tibia, humerus scapula, clavicle, ribs, pelvis, metacarpals, metatarsals, mandible and skull. Parosteal lipoma patients, whose age ranges from 40 to 60 years, usually present with a history of a slowly growing, large, painless, non-tender immobile mass that is not fixed to the skin [4,5,9]; the presence of parosteal lipomas ranges from as little as 3 weeks to as long as 41 years, the mean being 8±10 years [7].
Motor or sensory deficits due to nerve compression caused by parosteal lipomas are common if compared with other lipomatous lesions [10]. This clinical manifestation is most frequently associated with forearm lesions. In one study by Moon and Marmor [10], 11 out of 20 cases (55%) exhibited nerve palsy, which most often affected the posterior interosseous nerve. However, involvement of the radial, sciatic, ulnar and median nerves has also been reported in the literature [11,12].
The etiology of parosteal lipomas is not known. They are, on account of their intimate relationship with the bone, considered lipomas of the bone [13].
To our knowledge, about 150 cases of parosteal lipoma have previously been described in the literature [14,15].
Owing to the rarity of this condition and the difficulties encountered in its diagnosis and treatment, we wish to report fifteen cases of parosteal lipoma observed in three departments of plastic surgery.

Materials and Methods

We reviewed the clinical records of 15 patients who underwent surgery to remove a parosteal lipoma between November 2003 and March 2011. None of these cases has previously been described. The diagnosis of a parosteal lipoma was made by the histological findings, the confirmation of fat content at Magnetic Resonance (MR) or Computed Tomography (CT).
All the patients were admitted to the Plastic Surgery Departments of "La Sapienza" University of Rome, of the University of Catanzaro or of the University of Cagliari.
The radiographs were reviewed and assessed for lesion location, size, calcification, marginal sclerosis, trabeculation, bone expansion and modelling deformity. The MR and CT images were also evaluated for the same features.
The data regarding the patients' characteristics, lesion site, radiological appearance of the lesions, operating technique, histological findings, complications and need for further operations were collected and analyzed.

Results

A total of 15 cases are included in this analysis. Sex distribution was M8:F7 in our series.

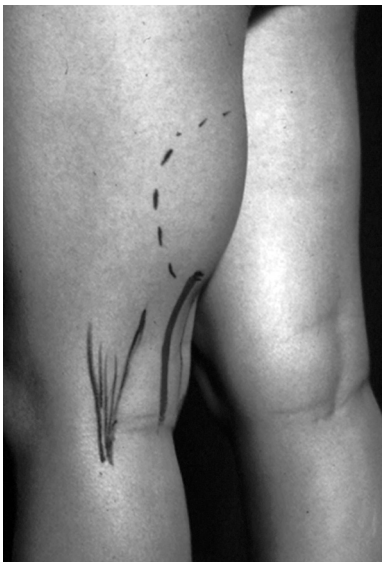

Fig. 1: Lesion of the thigh.

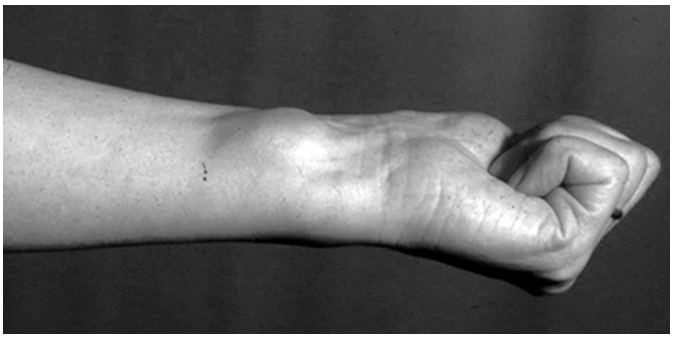

Fig. 2: Lesion of the of the proximal forearm.

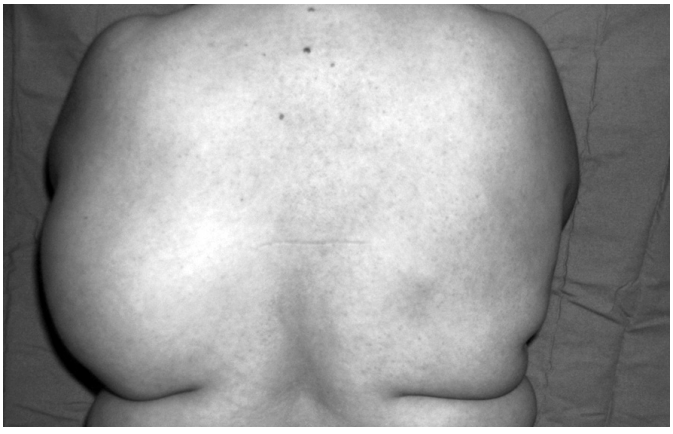

Fig. 3: Lesion of the the trunk along the midaxillary line.

All the patients was Caucasian.
The patients' age ranged from 26 to 66 years, with a median age of 44.5 years.
The site of the lesion was the thigh in four cases (26.7%) (Fig. 1), proximal forearm in four cases (26.7%) (Fig. 2), the shoulder in two cases (13.3%) , the sacrococ-

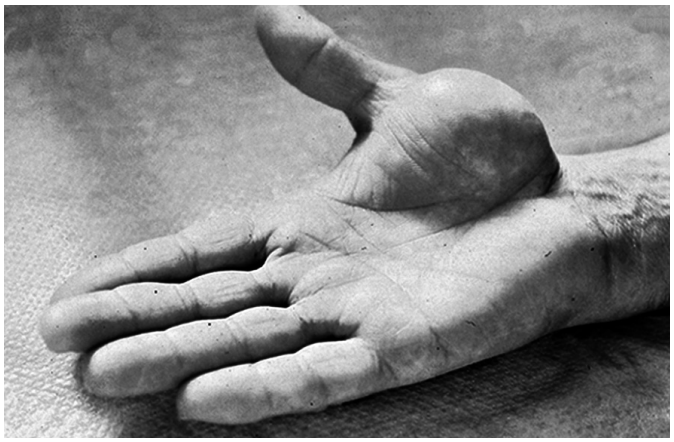

Fig. 4: Lesion of the hand.

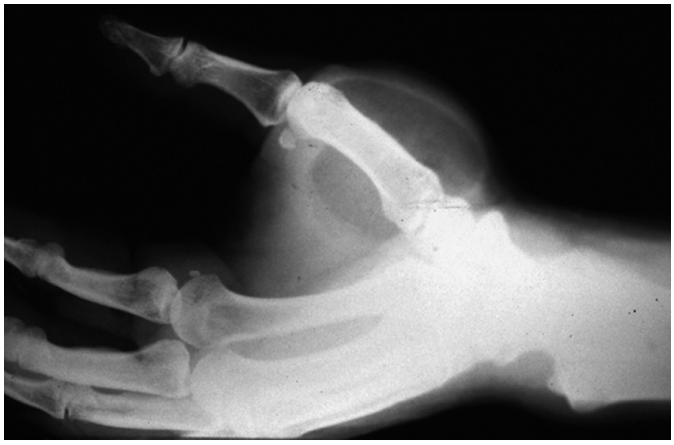

Fig. 5: Plain radiograph of a hand's lesion shows a radiolucent soft tissue mass and mild cortical thickening.

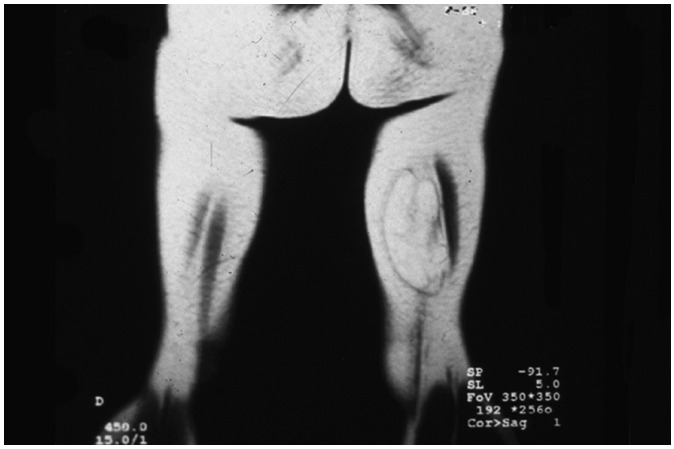

Fig. 6: TC images of a thigh's lesion scan reveals a septated soft tissue mass with fat attenuation adjacent to the femur.

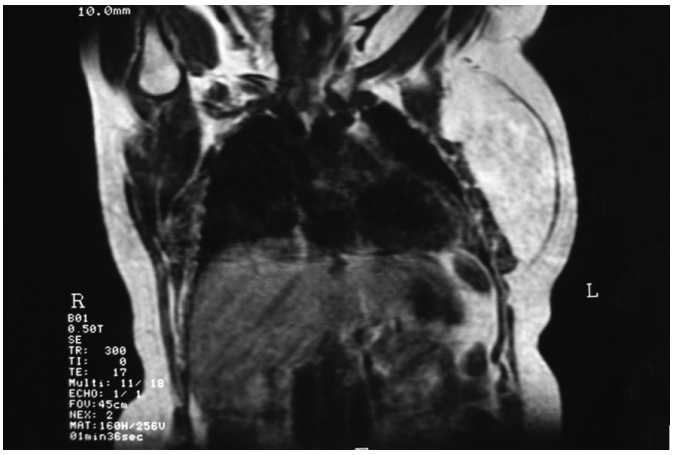

Fig. 7: TC images of a trunk's lesion shows lipomatous mass adhering to periosteum of the ribs.

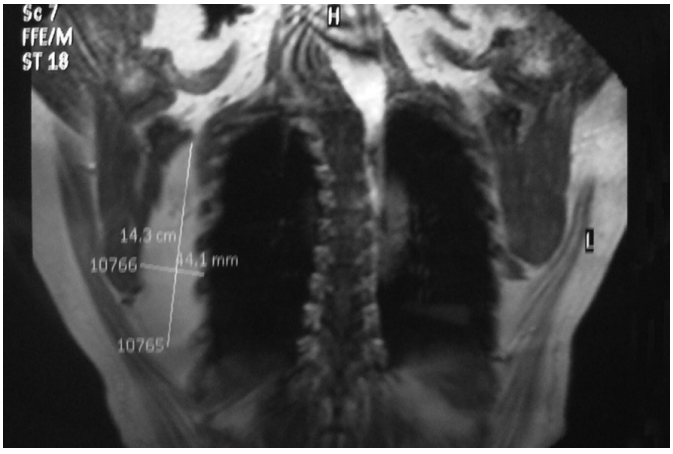

Fig. 8: MR images of a trunk's lesion reveals the hyperintense lesionto be attached to the ribs.

cygeal region in two cases (13.3%), the trunk, along the midaxillary line, with multiple rib adhesions in two cases (13.3%) (Fig. 3) and the hand in one case (6.7%) (Fig. 4). Nine patients (60%) reported pain in the area of the lesion, while the remaining six (40%) did not refer any symptoms. One of the patients (6.7%) with the lesion in the forearm displayed signs of superficial radial nerve compression; radiographs in this patient revealed hyperostosis.

The presence of the lesion ranged from as little as 11 months to as much as 16 years (mean 8 years and 3 months).

Plain radiographs showed a radiolucent lesion in the soft tissue adhering to the subjacent bone with some osseous changes in 13 cases (86.6%), while no osseous changes were evident in the remaining 2 cases (Fig. 5).

CT scans determined the extent of the mass as well as its relationship with the surrounding structures (Figg. 6, 7), while MR ruled out any malignancies (e.g. liposarcomas) (Fig. 8). All the patients underwent surgical excision under anesthesia and removal of the affected periosteum.

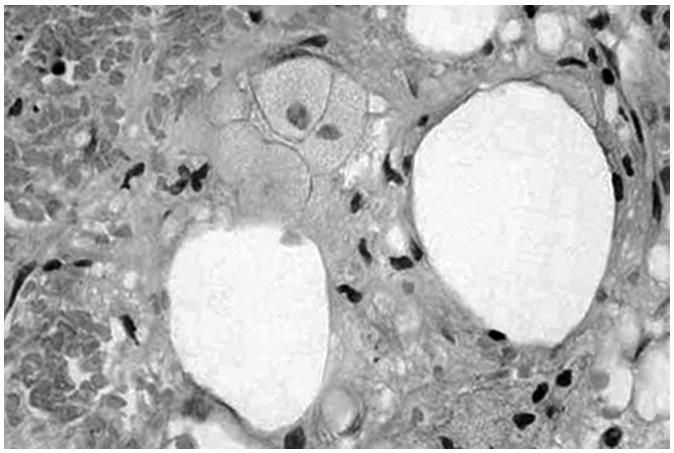

Fig. 9: Histological images of a lipoma, showing the typical appearance of mature adipocyte tissue (haematoxylin and eosin stain, original magnification x200).

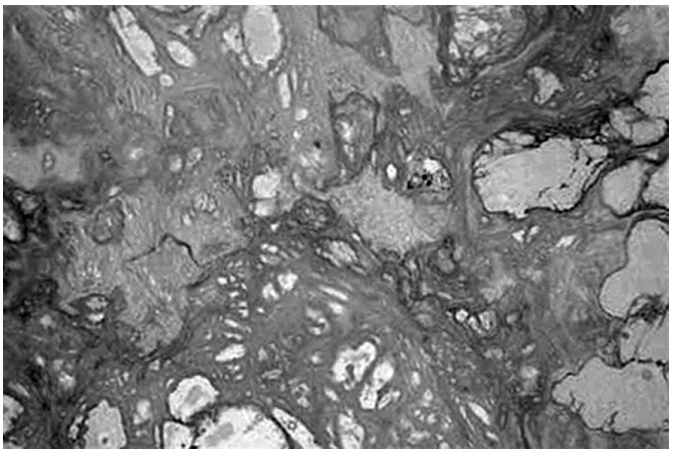

Fig. 12: Area of dense dystrophic calcification. There is the presence of ghost outlines of necrotic adipocytes (haematoxylin and eosin stain, original magnification x200).

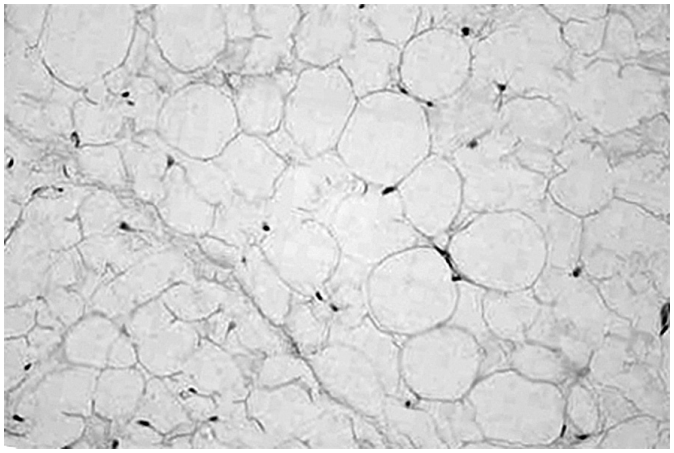

Fig. 10: A lipoma demonstrating areas of fat necrosis in the presence of reactive foamy histiocytes (haematoxylin and eosin stain, original magnification x400).

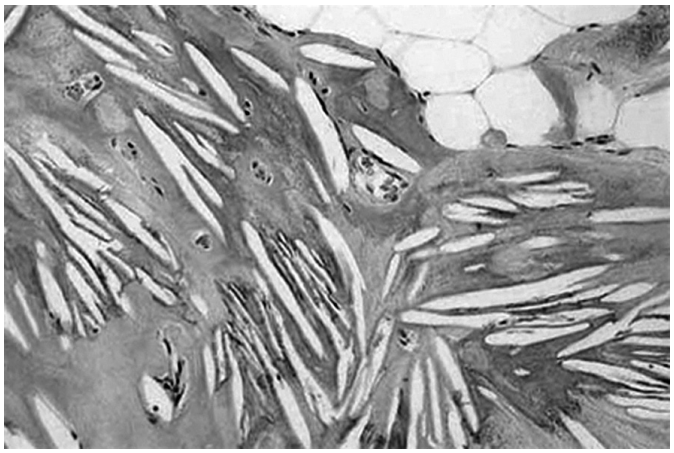

Fig. 11: Area of dystrophic calcification and cholesterol clefts within a lipoma (haematoxylin and eosin stain, original magnification x200).

The size of the lesions ranged from 6x5x5 cm to 25x15x10 cm.

Surgery in all the cases entailed resecting the tumor with parosteal excrescence. In three cases with hyperostosis, a further exeresis of the bone was performed.

The histological investigation revealed areas of mature fat in all the cases. No cellular atypia or mitoses were encountered (Fig. 9). In five cases, foci of fat necrosis were observed (Fig. 10). Areas of fat-containing cholesterol clefts, dystrophic calcification and focal reactive bone formation were observed in three cases (Fig. 11). In one case in which the dystrophic calcification was extensive, scattered ghost outlines of adipocytes were observed (Fig. 12). No cystic changes were detected at histology, nor was encapsulation observed in any case.

In one case located in the thigh, the postoperative course was complicated by superficial femoral vein thrombosis, which was successfully treated medically. Patient with nerve involvement recovered fully from these symptoms after surgery.

No signs of recurrence have been observed in any of the patients.

Discussion

Periosteal lipoma is a rare benign neoplasm that is intimately related to the periosteum, although no fat cells are present in the cambium layer of the periosteum [16]. The precise site of origin of parosteal lipomas is uncertain [16].

A large number of parosteal lipomas are asymptomatic and are discovered accidentally. Although there was a slight predominance of male patients in our series, the sex distribution in the meta-analysis group is more even and concurs with other reports. This finding is in marked contrast to the sex distribution of soft tissue lipomas,

which are more prevalent in females and are considered to be true benign tumours [17,18].

The age of patients upon presentation varies considerably. As many intraosseous lipomas are found accidentally, the lesions are likely to have been present for numerous years prior to discovery. Lipomas are usually solitary [17,18].

The clinical symptoms associated with parosteal lipomas are due to the compression of adjacent neurovascular and muscular structures; they cause motor and sensory function deficits whose severity depends on the size and location of the lesions, as displayed by one patient in our series [12].

The gross and histological findings of a parosteal lipoma are the same as those of a subcutaneous lipoma, the only difference being that the former has a broad-based attachment to the periosteum and bone. A parosteal lipoma often appears as a lobulated yellowish mass that is composed of mature adipocytes and is well encapsulated if we exclude the site in which it adheres to the bone. However, metaplastic bone formation in the lesion and periosteum, as well as various patterns of osseous change in the subjacent bone, have been reported [7,12]. Adipocytes were observed in the periosteum in all our cases; these had either infiltrated the periosteum or originated in the periosteum.

Pathologically, a parosteal lipoma is circumscribed by a thin, fibrous capsule. The multilobulated mass has a broad-based attachment to the underlying bone [19].

Cartilage, osteoid metaplasia and areas of osseous excrescences or cortical thickening extending from and attaching the lesion to the bone surface are common [21-23]. It is this relationship with the underlying bone that distinguishes such lesions from soft-tissue lipomas. As with other lipomas, parosteal lipomas display a lobular form of growth, often with intervening thin septations.

Recent cytogenetic analysis has shown a 3;12 translocation in parosteal lipomas, which is similar to that found in soft-tissue lipomas and consequently points to a common pathogenesis of these two types of lipoma [13].

A radiological evaluation, including CT [4] and MR [1,19,24], is essential to evaluate the location and nature of the lesion The imaging features of parosteal lipoma are usually distinctive. Indeed, nearly 70% of the patients in one study had abnormal underlying bone while 50% had an osseous reaction [7]. The classical findings are reactive changes, including bowing of the bone or cortical erosion due to the adjacent lipomatous mass [7]. The main radiographic features of parosteal lipoma include a juxtacortical radiolucent lipomatous mass with varying degrees of septation associated with surface bone changes ranging from very subtle to obvious cortical thickening and osseous protuberances or excrescences of varying size. These areas of cortical abnormality, which are best evaluated by means of radiography, do not display medullary or cortical continuity with the underlying bone, as would instead be expected in an osteochondroma [1].

At CT and MR imaging, parosteal lipomas have a homogeneous, lobulated appearance and adhere to the surface of the adjacent bone. CT also accurately distinguishes the lipomatous component of the mass, the varying degree of septation and the relationship of the mass to the underlying cortex [1]. The osseous protuberances may be quite prominent and exhibit both cortical and marrow components, though no continuity with the underlying bone is seen in this case either. Contrast material administration (at either CT or MR imaging) may reveal mild enhancement in the fibrous tissue rim of the parosteal lipoma, though this feature is uncommon [1].

MR imaging is considered to be a more means of evaluating parosteal lipoma than CT. The tumor is identified on MR images as a juxtacortical mass with signal intensity identical to that of subcutaneous fat, regardless of the pulse sequence [1]. Heterogeneity in these lesions is invariably present and corresponds to the pathological components in the lesion. Areas with intermediate signal intensity on T1-weighted images and high signal intensity on T2-weighted images represent the cartilaginous components in parosteal lipoma [1].

Adjacent muscle atrophy which is caused by associated nerve entrapment and is not well detected by CT, can be identified on MR images as increased striations of fat in the affected muscle [1]. This finding appears most clearly on T2-weighted images because of the signal intensity of normal muscle is lower than that of fat. Lastly, MR imaging reliably demonstrates the relationship of the tumour to the underlying healthy bone and muscle; this is particularly important when planning surgery because parosteal lipoma usually adheres firmly to the underlying cortex at the site of surface bone production.

The differential diagnosis with low-grade liposarcoma may be difficult, though there are no previous reports of either primary parosteal liposarcoma or degenerated benign parosteal liposarcoma in patients with these parosteal lipomas [24]. A differential diagnosis of parosteal lipoma should also be made with teratoma, dermoid, osteoma and myositis ossificans.

Parosteal lipomas are known to cause nerve compression, there being previous reports of radial, sciatic, ulnar and median nerve involvement [25,26]. Interestingly, the anchoring effect of the attachment site at the bone may predispose these paosateal lipomas to mass effect and nerve impingement, as opposed to the pattern of soft tissue lipomas, which expand along the path of least resistance [26].

Treatment of parosteal lipoma consists of complete surgical resection, with further exeresis of the bone and periosteal excrescence in cases with hyperostosis [20,27,28].

In cases with nerve entrapment, the tumor must be removed before the onset of irreversible muscle atrophy to maintain function [1,11,12]. The nerve must also be separated from the parosteal lipoma and spared during surgical excision. At surgery, parosteal lipomas are characteristically encapsulated and adhere firmly to the under-

lying periosteum The sites in which the tumors are most strongly attached to the underlying bone are those where osseous proliferation is most prominent. Owing to this peculiar characteristic, adequate surgical removal of a parosteal lipoma requires either subperiosteal dissection, an osteotomy to separate the lesion from the underlying bone, or segmental resection of bone, which is in contrast to the relatively easy dissection of a soft-tissue lipoma lying adjacent to bone [1,11,12]. Local recurrence is unusual but has been reported. There are no reports of malignant transformation.

In conclusion, parosteal lipomas are rare entities associated with periosteal involvement depending on their location. Current management should include CT scanning and MR. Surgery, which is mandatory treatment, requires particular attention to ensure that any periosteal involvement is removed completely.

Riassunto

Il lipoma è il più comune tumore e può essere trovato praticamente in ogni tessuto o organo. Tuttavia, il lipoma parosteo è una rara neoplasia benigna del tessuto adiposo che contrae un rapporto intimo con il periostio. L'incidenza di questo tumore è di 0,3% tra tutti i lipomi. Oltre 150 casi di lipoma parosteo sono stati precedentemente descritti in letteratura. A causa della rarità di questa condizione e le difficoltà incontrate nella sua diagnosi e trattamento, ne vogliamo segnalare quindici nuovi casi.

Abbiamo esaminato le cartelle cliniche di 15 pazienti sottoposti a intervento chirurgico per l'asportazione di un lipoma parosteo tra il novembre 2003 e il luglio 2009. La diagnosi di lipoma parosteo è stata confermata dai risultati dell'esame istologico, dalla presenza di tessuto adiposo nelle scansioni della risonanza magnetica o della tomografia computerizzata.

L'intervento chirurgico ha comportato in tutti i casi l'asportazione della lesione in blocco con il periostio. In tre casi, in cui era presente iperostosi, è stata eseguita un'ulteriore exeresi del tessuto osseo.

I lipomi parostei sono entità rare associate con la partecipazione periostale. L'attuale trattamento dovrebbe includere la scansione tomografica computerizzata e la risonanza magnetica. Il trattamento chirurgico, che è quello obbligatorio, deve richiede un'attenzione particolare per garantire che qualsiasi coinvolgimento periostale sia rimosso completamente.

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