Bifocal parosteal osteoma of femur: A case report and review of literature

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Abstract

Osteoma is a benign, slowly growing, asymptomatic, bone-forming tumor arising from cancellous or compact bone. Osteoma usually is a solitary lesion, but in patients with Gardner's Syndrome it may be multiple. osteoma may rarely have a parosteal localization. Parosteal osteoma has peculiar radiographic, histologic and clinical features. We describe a case report of a 51-years old man with a bifocal parosteal osteoma of the femur in a non-syndromic patient. This is the first described patient with a bifocal lesion. In literature only 24 cases of paraosteal osteoma are found. Our patient underwent surgery and the lesions were fully excised. At one year follow-up there was no evidence of recurrence.

Introduction

Osteoma is a benign, slowly growing, asymptomatic, bone-forming tumor arising from cancellous or compact bone.[1]

Osteomas are most commonly located in the skull (especially is the paranasal sinuses and jaw bones) and facial bones. Long bone involvement with osteoma is rare, with a prevalence of 0.03% of biopsied primary bone tumors.[2] There are a few reported cases of osteomas of the clavicle,[3] pelvis,[4,5] or long bones.[6,7] The tumor occurs most frequently in adults, and more than 78% of the patients are older than 40 years.

Usually osteoma is a solitary lesion, but in patients with Gardner's Syndrome they may be multiple and associated with intestinal polyps, fibromatous and other lesions of connective tissue, and epidermal cysts.[2,8]

Rarely, osteoma may have a parosteal localization. Parosteal osteoma (PO) has peculiar radiographic, histologic and clinical features. Radiographically osteoma presents as a solitary, long-standing uniform dense sclerotic lesion attached to the surface of the diaphysis or metadiaphysis.

Multiple parosteal osteomas appear to be exceedingly rare.

They may be incidentally identified as a mass in the skull or mandible, or as the underlying cause of sinusitis or mucocele formation within the paranasal sinuses

Three histological patterns are recognised:

- ivory osteoma, made of dense bone lacking Haversian system;
- mature osteoma resembles 'normal' bone, including trabecular bone often with marrow
- mixed osteoma, a mixture of ivory and mature histology.

The imaging shows very radiodense lesions, similar to the normal cortex, whereas mature osteomas may demonstrate central marrow.[9]

In the present study, we describe a case of multiple parosteal osteoma STS. We also provide a review of the literature about this topic.

Case Report

In the 2012 came to our attention a 51-years old man for a moderate painful swelling of the right knee and medial side of the right thigh. Swelling gradually increasing size in the last 15 years very slowly and in the last years appear worsening pain, both hip and knee. He was a bricklayer. The patient denied any history of recent or remote trauma at right leg or history of radiant exposure. He didn't smoke. He hadn't familiarity for cardiovascular diseases, cancers or other health problems. His sister had a similar painless mass at right knee. Nobody in the family suffered of Gardner's syndrome, fibromatosis, polyposis or poor eyesight. At observation of the right thigh, there was one hard mass in the right inguinal region and another hard mass on the medial side of the distal femur. No similar swelling noticed elsewhere in the body. The masses were no mobile and painful at palpation (VAS 6/10). The skin up to the masses was normochromic but hyposensitive. The ROM of the hip and knee were normal. The knee was stable. FABER and FADIR test, Posterior impingement test, log roll test, Thomas test were negative. There was no muscle wasting. The reflexes were normal. No constitutional symptoms were present. A routine blood exam was normal. An X-Ray of the knee showed two voluminous bone masses above the lesser trochanter and on the medial side of the distal femoral epiphysis. An incisional biopsy through a jamshidi needle was performed: the histological examination showed a surface osteoma.

The patient refused any excisional surgery. After three years, the patient came back to our attention for the worsening of the pain. A CT-scan (Figure 1) was performed and showed two voluminous exostotic formations with calcific hood. The first mass starting from the small trochanter measures 10x7 cm with an extension of 15 cm; this formation imprinted and displaced sartorial muscle. The second formation is located on the medial side of the distal femoral epiphysis (6x4 cm and extension of 8 cm), imprinted and displaced the vastus intermedius and medial muscles.

The patient underwent an excisional surgery through an ileo-inguinal approach for the proximal lesion and a medial knee

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approach for the distal lesion. The excision was particularly difficult, due to their hardness, and required special scalpels and saws; no prophylactic osteosynthesis was needed.[10] Macroscopically both lesions appeared as rounded, stone-hard masses. At histopathological level, they were composed of dense, compact, hypo cellular lamellar bone without cytological atypia. Neither macroscopic nor microscopic there was evidence of cartilage tissue or cartilage cap in the tissue specimens of both lesions.

The patient discharged after 3 days without complications. At 15-days after surgery stitches were removed, the wounds looked flat and regular. The VAS-score pain was 4/10. At one-month follow-up, X-ray control was performed; there were no evidence of pathological mass. The VAS-score was 2/10. After six months, the patient recovered autonomy in all his ADL (Activities of Daily Living) and IDAL (Instrumental Activities of Daily Living). At one-year follow-up there was no evidence of recurrence.

Discussion

Our search was performed on the PubMed and Cochrane databases using different key-words: parosteal osteoma, juxtacortical osteoma and surface osteoma. The examined range of time was between years 1951 and January 2018. Resulted 165 articles which 144 were excluded based on the language (non English articles) and title because they dealt with different topics, such as non-surface osteomas, syndromic osteomas, veterinary osteomas or other bone neoplasms. The resultant 21 articles were analyzed. In 1951 Geschickter and Copeland coined for the first time the term "parosteal osteoma" .[11] PO occur in early adult or in middle life.[11] The prevalence has been estimated to be 4.2 per 1000 patients.[12] Osteoma that involves the long bone is rare with a prevalence of 0.03 of 1000 patient undergoing bone biopsy for primary tumor.[12] PO are located mainly in the skull and the face (paranasal sinuses, jaw bones and facial bones). Multiple osteomas can be associated with Gardner's syndrome or tuberous sclerosis.[13] PO of bones other than skull and face involved lower extremities in the 86% of cases, both diaphyseal and metadiaphyseal region.[3] Histologically it consists entirely of dense sclerotic lamellar bone similar to cortical bone without fibrous stroma. In most cases the typical presentation is a swelling, hard, painful mass that gradually enlarges. PO grows very slow and can reach big size (in the Bertoni review[3] one patient has a PO of 20x2.8x3 cm). While PO is often clinically painful and palpable five asymptomatic cases have been reported.[3,14,15] Many other entities can simulate a PO; the differential diagnosis includes: melorheostosis, myositis ossificans, ancient osteochondroma developing in mature bone, secondary reactive bone change and parosteal osteosarcoma.[7,16,17] There are no international guidelines for treatment of PO. For his very low growth potential close clinically and radiographically follow-up will avoid extensive and sometimes debilitating treatment.[3] After complete surgical excision or surgical debulking the prognosis is excellent without a propensity for local recurrence.[3,11] Campanacci suggested a marginal excision without wide margins because the only debulking (intralesional resection) is usually uneffective and exposes the patient to pain,

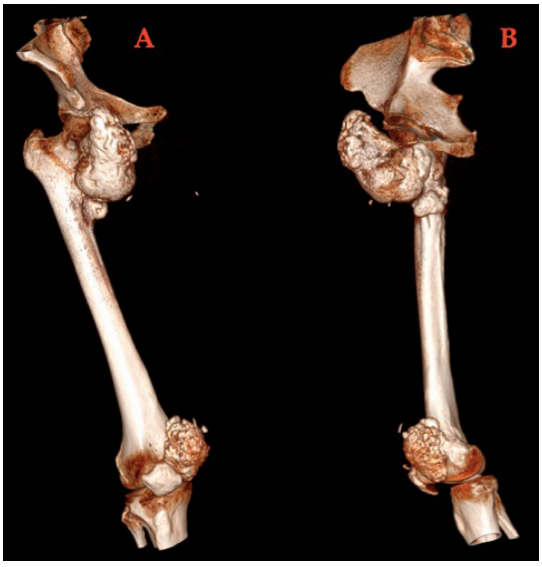

Figure 1. 3D TC reconstruction of right femur. In A frontal view of the lesions and the femur, in B lateral view.

Table 1. Overview of the available studies focused on the parosteal osteoma.

Authors	Year	Type	Sex	Age	Localization
Sundaram M *et al.*	1996	Case series	3 F 1 M	Mean age 37	1 scapula 2 femur 1 fibula
Bertoni F *et al.*	1995	Case series	8 M 6 F	Mean age 45	1 clavicle 1 humerus 6 femur 4 tibia 2 fibula
Hansford BG *et al.*	2014	Case report	F	45	Femur
Inokuchi T *et al.*	2014	Case report	F	51	Left clavicle
Yun SJ *et al.*	2012	Case report	M	66	Distal femur
Chikuda H *et al.*	2002	Case report	F	47	Ulna
Soler Rich R *et al.*	1998	Case report	M	33	Right iliac bone
Houghton MJ *et al.*	1995	Case report	F	47	Right pubic bone

to a secondary larger resection of the tumor and to the risks of a double surgery.[12,18]

In literature only 24 cases of PO are found (Table 1).[3,14,15,18-23] The mean age of incidence is 40 years. In 17 cases the PO is located on the lower limb, in 2 cases in the upper limb. In 5 cases the PO location is on flat bones (clavicle, scapula, pelvis). The most frequent localization is the femur (10 cases). All patients underwent surgery (biopsy or biopsy and later excision or primary excision). No complications related to surgery were described. None of the patients needed further surgery after the excision. No recurrence of the lesion was found.

Sundarman treated 4 cases with different localizations (Table 1) and after excision no recurrence of the lesion was found.[23]

Bertoni performed 9 resections, 4 incisional biopsies, 1 debulking. At the follow-up in 9 cases no evidence of the tumor was found.[3] In 2 cases tumor was stable after 10 years, 1 patient died for a metastatic myeloma e 2 were lost in the follow-up.

Hansford and Yun both performed a wide resection of the lesions.[15,22] Yun found an ostecondroma together with a parostal osteoma.[15] In both cases there is no follow-up. Sundarman performed a resection of the lesion[23]. After 2 years FU no clinical or radiological recurrence was found.

To the authors' knowledge, bifocal PO in non-syndromic patient has not been previously described in the literature.

Only Yun described a simultaneously parosteal osteoma and osteochondroma in the distal femur of a single patient.[15] Hansford described a strange case of PO not homogeneously dense and with a separate nodule in the soft tissues.[22]

Conclusions

PO is a rare, benign, slow growing lesion. An accurate diagnosis is important to exclude more aggressive and malignant neoplasms. We present the first case of a bifocal PO of the femur in a non-syndromic patient. The excision of the lesions guaranteed a fast recovery of the patient and a quicker relief from pain. No recurrence of the lesion was found.

References

1. Mubeen K, Vijayalakshmi KR, Abhishek PR. Peripheral ivory osteoma of the mandible in a young female patient. J Investig Clin Dent 2012;3:148-51.
2. J.M.Mirra, "Osteoma," inBone Tumors. Clinical, Radiologic and Pathologic Correlations, J. M. Mirra, Ed., pp. 174-182, Lea and Febiger, Philadelphia, Pa, USA 1989.
3. Bertoni F, Unni KK, Beabout JW, Sim FH. Parosteal osteoma of bones other than of the skull and face. Cancer 1995;75:2466-73.
4. Cervilla V, Haghighi P, Resnick D, Sartoris DJ. Case report 596: Parosteal osteoma of the acetabulum. Skeletal Radiol 1990;19:135-7.
5. Mirra JM, Gold RH, Pignatti G, Remotti F. Case report 497: Compact osteoma of iliac bone. Skeletal Radiol 1988;17:437-42.
6. Baum PA, Nelson MC, Lack EE, Bogumill GP. Case report 560: Parosteal osteoma of tibia. Skeletal Radiol 1989;18:406-9.
7. O'Connell JX, Rosenthal DI, Mankin HJ, Rosenberg AE. Solitary osteoma of a long bone. A case report. J Bone Joint Surg Am 1993;75:1830-4.
8. Unni K: "Osteoma," in Dahlin's Bone Tumors, pp. 98-101, Mayo Foundation for Medical Education and Research, Lippincott Williams &Wilkins, Philadelphia, Pa, USA, 6th edition, 2010.
9. Lambiase RE, Levine SM, Terek RM, Wyman JJ. Long bone surface osteomas: imaging features that may help avoid unnecessary biopsies. AJR Am J Roentgenol 1998;171:775-8.
10. Perisano C, Barone C, Stomeo D, et al. Indications for prophylactic osteosynthesis associated with curettage in benign and low-grade malignant primitive bone tumors of the distal femur in adult patients: a case series. J Orthop Traumatol 2016;17:377-82.
11. Geschickter CF, Copeland MM. Parosteal osteoma of bone: a new entity. Ann Surg 1951;133:790-807.
12. Peyser AB, Makley JT, Callewart CC, et al. Osteoma of the long bones and the spine. A study of eleven patients and a review of the literature. J Bone Joint Surg Am 1996;78:1172-80.
13. Gardner EJ, Plenk HP. Hereditary pattern for multiple osteomas in a family group. Am J Hum Genet 1952;4:31-6.
14. Houghton MJ, Heiner JP, De Smet AA. Osteoma of the innominate bone with intraosseous and parosteal involvement. Skeletal Radiol 1995;24:455-7.
15. Yun SJ, Jin W, Park YK et al. Simultaneously detected parosteal osteoma and osteochondroma in the distal femur of a single patient. Clin Imaging 2013;37:950-3.
16. Spinelli MS, Perisano C, Della Rocca C et al. A case of parosteal osteosarcoma with a rare complication of myositis ossificans. World J Surg Oncol 2012 29;10:260.
17. Pezzillo F, Maccauro G, Nizegorodcew T, et al. Resection of parosteal osteosarcoma of the distal part of the femur: an original reconstruction technique with cement and plate. Sarcoma 2008:763056
18. Campanacci M. Osteoma in Bone and Soft Tissue tumors, New York, Spinger. 1990; PP 349-354.
19. Soler Rich R, Martínez S, de Marcos JA et al. Parosteal osteoma of the iliac bone. Skeletal Radiol 1998;27:161-3.
20. Chikuda H, Goto T, Ishida T, et al. Juxtacortical osteoma of the ulna. J Orthop Sci 2002;7:721-3.
21. Inokuchi T, Hitora T, Yamagami Y, et al. Parosteal osteoma of the clavicle. Case Rep Orthop 2014;2014:824959.
22. Hansford BG, Pytel P, Moore DD, Stacy GS. Osteoma of long bone: an expanding spectrum of imaging findings. Skeletal Radiol 2015;44:755-61.
23. Sundaram M, Falbo S, McDonald D, Janney C. Surface osteomas of the appendicular skeleton. AJR Am J Roentgenol 1996;167:1529-33.