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Case report

Texture analysis in a rare case of tibial intraosseous lipoma

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Summary. Intraosseous lipoma is a very rare lesion, accounting for only 0.1% of all primary osseous tumors (1), first described in 1980 (2). This lesion is considered the rarest of benign bone tumors (3); probably it is not the actual incidence because these lesions are frequently asymptomatic and the introduction of cross-sectional imaging, especially MRI, seems to have increased the detection (4). The majority of intraosseus lipomas are in the lower limbs (70%) and the os calcis being the most frequently involved (32%). Most cases reported in literature have an age of 40 years (5). Tumor texture could be measured from medical images that provide a non-invasive method of capturing intratumoral heterogeneity and could potentially enable a prior assessment of a patient. Some Authors recently proposed Texture analysis to characterize musculoskeletal lesions (6). For the first time we measured the tumoral texture from Magnetic Resonance images in tibial intraosseous lipoma in a 29-years-old female. (www.actabiomedica.it)

Key words: texture analysis, intraosseous lipoma, musculoskeletal imaging

Introduction

Intraosseous lipoma is a very rare lesion, accounting for only 0.1% of all primary osseous tumors (1), this lesion is considered the rarest of benign bone tumors (3). Tumor texture could be measured from medical images that provide a non-invasive method of capturing intratumoral heterogeneity and could potentially enable a prior assessment of a patient; some Authors recently proposed Texture analysis to characterize musculoskeletal lesions (6). For the first time we measure the tumoral texture from Magnetic Resonance images in a tibial intraosseous lipoma in a 29-years-old female.

Patient information

A 32-year-old female presented to her general practitioner with a right severe pain of the knee after jogging; the patient reported a limited range of motion. There was no other prior pertinent medical history.

Clinical findings

Initial imaging was performed with radiography that demonstrated a large, lytic lesion causing prominent expansile remodeling of the proximal tibial diaphysis; the cortex was thin but well-formed and there was not an erosion (Fig. 1).

An MRI was performed (1,5 T, Philips) to further characterize the lesion. Pre- and post-contrast T1, T2, short tau inversion recovery (STIR) weighted and Dual Gradient-Echo In-Phase and Opposed and long-repetition-time fat-suppressed sequences were executed. Imaging showed in the proximal tibial diaphysis a well-defined T1-weighted and T2-weighted

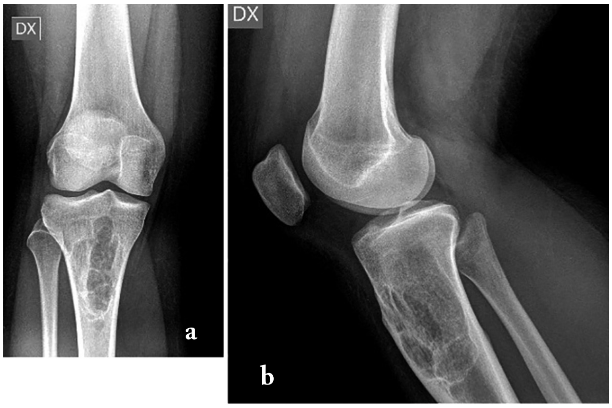

Figure 1. RX frontal (a) and lateral view (b). Lytic lesion causing prominent expansile remodeling of the proximal tibial diaphysis

high signal lesion (3,4 x 3,2 x 6,6 cm), mildly hyperintense relative to marrow in uninvolved adjacent bone and isointense relative to subcutaneous fat (Fig. 2).

Within the lesion multiple cystic areas with fluid signal were present (hyperintense on T2-weighted images and with low signal intensity on T1-weighted images) (Fig. 3).

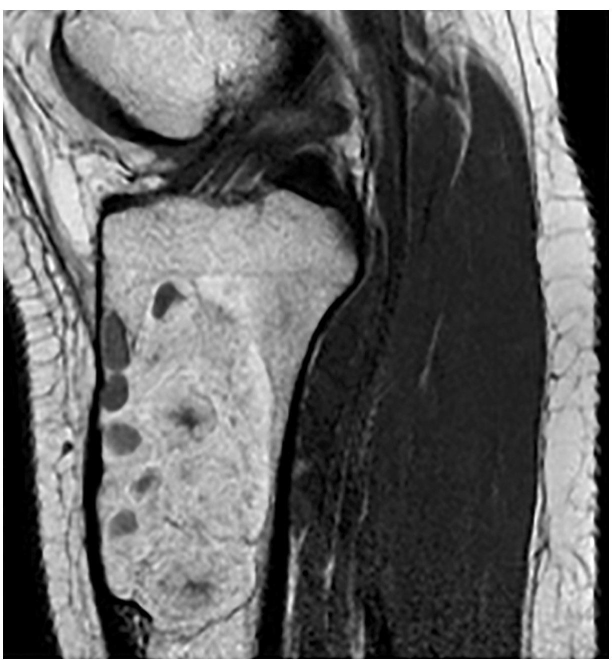

Figure 2. T1- weighted sequences (sagittal view): lesion with high signal intensity (same signal of adipose tissue) with multiple hypointense cystic areas. Cortex is thinned without erosion

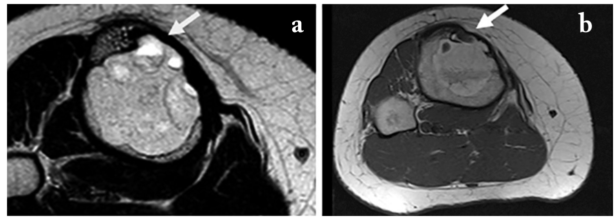

Figure 3. (Axial view) Cystic lesion lesion with high intensity signal in T2-weighted sequenze (a) and low intensity signal in T1-weighted sequenze (b)

Peripheral rimlike enhancement after gadolinium injection was found around the cysts. On long-repetition-time fat-suppressed images, the signal in regions with T1 high signal is isointense to that of subcutaneous fat (Fig. 4).

Subsequently, we analyzed the texture of the tumor using the open-source software ImageJ (version 1,46r, Wayne Rasband, National Institutes of Health, USA).

Radiologist with good experience and confidence with the use of Texture Analysis selected MRI slices and maximum diameter lesion were considered (Fig. 5). The slices were exported as a single TIFF image on a dedicated workstation and converted into 8 bit. The radiologist manually traced a ROI in the inner edge of

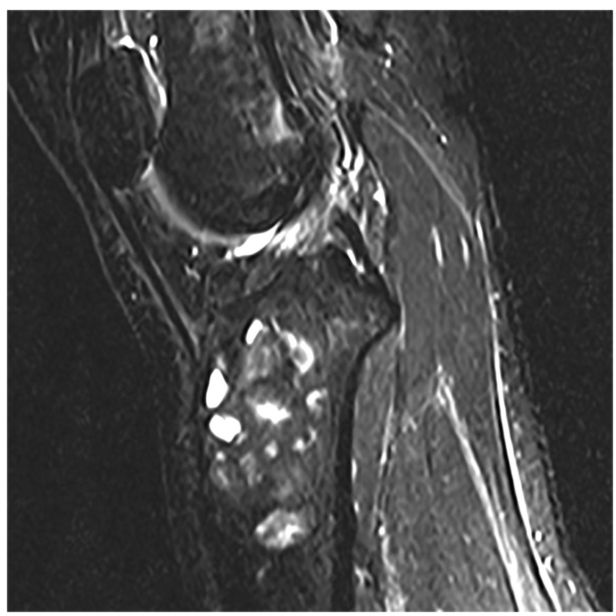

Figure 4. (sagittal view, STIR sequenze). The lesion presents suppression signal typical of adipose tissue

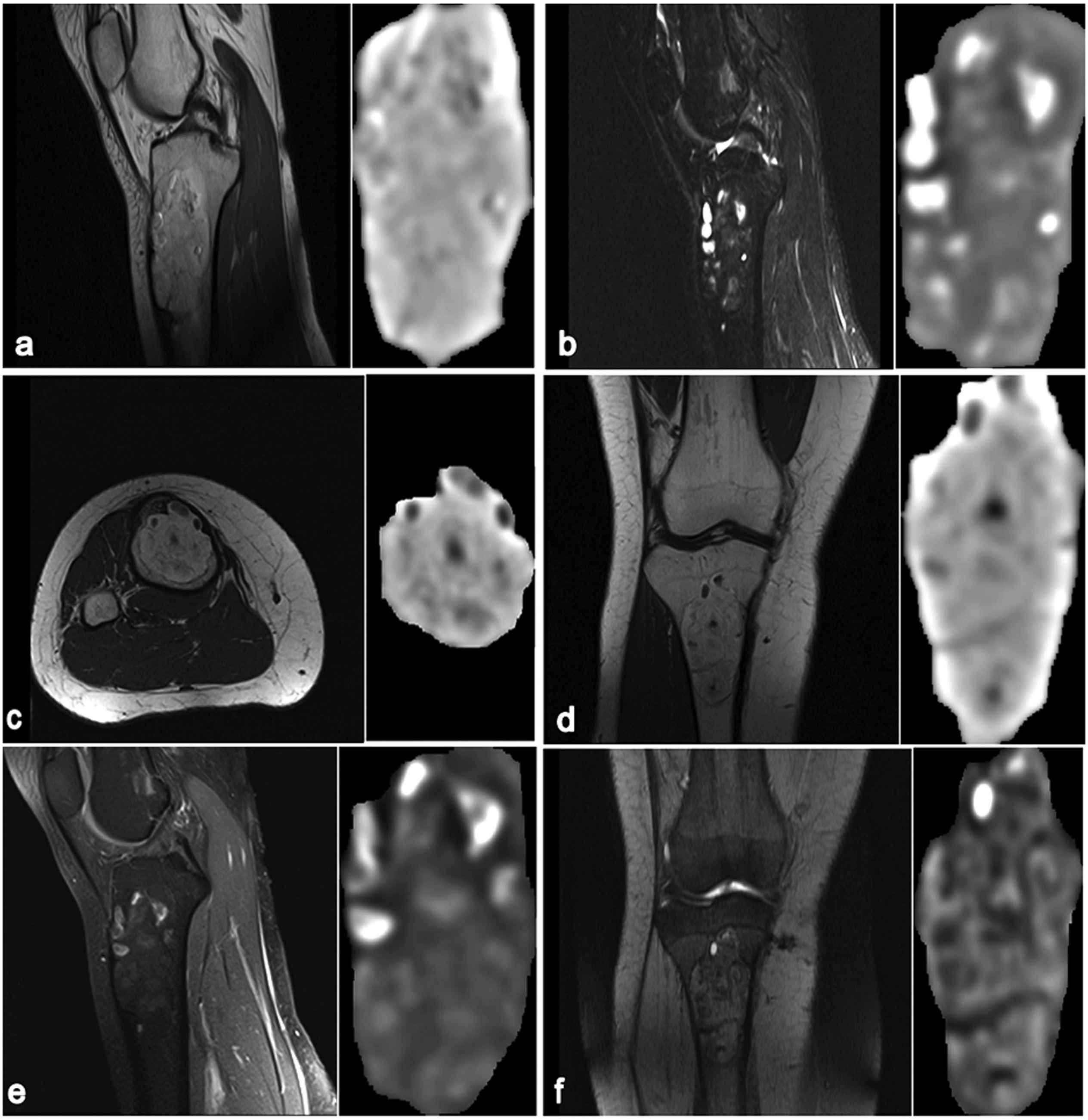

Figure 5. (a) Sagittal view PD T1-w (b) Sagittal view STIR (c) Axial view T1-w (d) Coronal view Spin-Echo T1-w (e) Sagittal view post contrast enhancement T1-w fat-sat (f) Coronal view, post contrast enhancement T1-w fat-sat. Near morphological image is shown the same lesion manually segmented after the application of a Spatial Scale Filter (sigma 4mm) and a Band-Pass Filter for the extraction of the textural feautures

the lesion. Then, the image was filtered and smoothed applying a band-pass filter and spatial filter (sigma 4mm); values obtained are reported in the following table (Table 1).

In our case, texture characteristics like the negative Skeweness and positive Kurtosi are typical for benign lesion (7-9). These data are completely original and, if confirmed by more effective studies, may help

Table 1.

	Pdt 1.2	Stir	t1 ax	t1 se cor	t1 sop	t2 gr
Mean	107.235	101.713	104.347	104.486	97.387	71.693
sd	23.307	33.623	23.470	30.580	34.440	52.218
skew	-0.066	-1.603	-0.608	-0.129	-1.873	-0.121
kurt	1.733	3.345	1.657	1.475	5.345	0.545
asm	0.040	0.052	0.077	0.084	0.045	0.085
contrast	135.411	134.487	214.234	191.819	88.332	103.250
correlation	1.998^{-4}	2.493^{-4}	1.881^{-4}	1.569^{-4}	3.885^{-4}	3.598^{-4}
idm	0.545	0.553	0.584	0.542	0.540	0.543
entropy	5.987	6.026	5.565	5.827	6.151	5.684

specialists in the future in the diagnosis and treatment planning of this disease.

CT biopsy was performed and showed mature adipose cells. No treatment were considered for the patient according with literature (4).

Discussion

Intraosseous lipomas are very rare bone tumours, with 200 cases reported thus far (5, 10, 11). Authors that presented the largest numbers of case were Milgram and Campbell et al. (61 cases, 35 of these were contributed by Members of the British Society of Skeletal Radiology) and carried out a meta-analysis of cases presented in the English literature since 1966 (5, 12).

Osseous lipomas are found in patients of widely divergent ages but are most commonly discovered in the 4th and 5th decades of life, with a slight predominance in males (male-to-female ratio 1.6:1)

Intraosseous lipomas in 70% of reported cases are associated with clinical symptoms (usually pain) (5). However, we think that the low incidence is determined by the lack of symptoms of injury.

Intraosseous lipoma often occurs in the lower limb (71%), usually within the calcaneus (about 30% of cases). Other frequent locations include: subtrochanteric femur, as well as femur and tibia in the region of the knee joint (5). A less common location is the upper limb (7%) and the axial skeleton (cranium – 4%, mandible – 3%, spine – 4%, pelvis – 5%) (5, 12-15).

A recent review of the literature report 14 cases of intraosseous lipoma (16) involving the spine; five cases (35%) occurred in the lumbar region, four (28%) in the sacral region, three (21%) in the cervical region, one (7%) in the thoracic region and one (7%) in the coccygeal region (17-26).

Histologically, these tumours are composed of mature adipose cells (slightly larger than the non-tumorous ones) and may include single spindle cells, regressive lesions (foci of fat necrosis, cystic spaces, and dystrophic calcifications) and bone trabeculae undergoing resorption (27-29).

Milgram classified these lesions into three stages, depending on the histopathological appearance (22).

Stage I includes lesions composed of mature lipocytes, similar to the cells of subcutaneous adipose tissue.

Stage II includes lesions whit necrotic foci, foamy macrophages and foci of reactive osteogenesis.

In stage III lesion is a completely necrotic with focal calcifications and cystic degeneration that resembles a bone infarct, the only features differentiating it from lipoma being lack of trabecular resorption or possible bone expansion (22, 28).

Some authors do not consider intraosseous lipomas true bone tumors (28, 29) because these lesions often localize in areas with physiologically low density of trabeculae and a higher yellow bone marrow content (ex. trochanteric region of the femur or the anterior part of heel bone). Another preferential localization is the Ward's triangle between the load-bearing trabeculae in the calcaneus (30, 31). In this area resorption of the trabeculae in unloaded area can determinate peripheral trabecular hypertrophy (5). Other authors consider the specific vascularization of the Ward's triangle as a predisposing factor to bone marrow infarction and development of regressive lesions (29).

Other authors thought that lipomas could constitute the last phase of involution of other focal lesions of bones (bone infarct for example) or regression of aneurismal cyst when this causes bone expansion (32).

Radiographic examination of these tumours present poorly delineated osteolytic lesions with a non-reactive bone degeneration or an accompanying malignant periosteal reaction and a large tumour in soft tissues; radiographic picture of intraosseous lipomas is usually uncharacteristic. There are many differential diagnoses based on RX findings includes non-ossifying fibroma, fibrous dysplasia, solitary cyst, giant-cell bone tumor, bone infarct and cartilaginous tumour (5, 28). Intraosseous lipomas usually present as cystic lesions with an increased radiolucency, surrounded by a sclerotic rim (74%) (5, 33). Lipomas located in the proximal femur may be surrounded by a relatively extensive area of sclerotisation (22). Lesions in the intertrochanteric area of the femur need to be differentiated from liposclerosing myxofibrous tumor, a very rare, benign fibro-osseous lesion with mixed histology, which may include components of lipoma (34). Inside the cysts, internal trabeculations may be present, while in lipomas with regressive lesions, foci of calcifications, usually located centrally (5, 28). Small bone expansion may be found in some cases of intraosseous lipomas and is usually of minor degree while greater expansion must be differentiate from aneurysmal cyst (27, 29).

MRI is essential for the correct characterization of this lesion because it is composed of adipose tissue and this method are actually diagnostic (35).

Fluid-filled spaces show a low-to-medium signal intensity in T1-weighted images, and a very high signal intensity in T2-weighted within intraosseous lipoma have been described in literature (34).

No contrast enhancement was present in the fat tissue of the intraosseous lipoma (35).

In many cases have been reported linear areas of low signal intensity on T1- and T2- weighted images correlated with areas of trabecular thickening and rim-like calcification.

Large fat-containing lytic lesion that demonstrated expansile remodeling and contained large areas of fat necrosis with peripheral calcification was diagnostic of an intraosseous lipoma. If MRI is contraindicated CT can be diagnostic (2, 35). CT examination revealed an extensive soft-tissue mass without macroscopically visible adipose tissue (5). In CT images fat tissue has low attenuation coefficient, ranging from –110 to –40 HU.

We analyzed the texture of the tumor using the open-source software ImageJ (version 1,46r, Wayne Rasband, National Institutes of Health, USA).

In our case texture characteristics like the negative Skeweness and positive Kurtosi are typical for benign lesion (7-9). These data are completely original and, if confirmed by studies more effective, may help specialists in the future in the diagnosis and treatment planning of this disease.

We believe, in agreement with the literature, that imaging of a tumour composed of adipose tissue proves its benign nature and concludes the diagnostic process (5). Additional proof for the validity of this approach is presented in papers analysing the presence of adipose tissue in benign and malignant bone tumours of various histological structures, which indicate that the existence of adipose tissue in malignant tumours is extremely rare (36, 37). A correct diagnosis is very important for the clinical prognosis. Some authors believe that no treatment should be considered for these tumors (38-44). Several others recommend, if there are any symptoms, carrying out currettage and filling the cavity with bone graft.

Teaching Point

Intraosseous lipoma is a very rare lesion, accounting for only 0.1% of all primary osseous tumors. MRI is essential for a correct diagnosis.

Texture analysis is a new technique that could help in the future in characterization of the lesion.

References

1. Mannem RR, Mautz AP, Baynes KE, Zambrano EV, King DM. AIRP Best Cases in RadiologicPathologic Correlation Intraosseous Lipoma. Radio Graphics 2012; 32: 1523-28, 10.1148/rg.325115031
2. Reig-Boix V, Guinot-Tormo J, Risent-Martinez F, et al.

Computed tomography of intraosseous lipoma of os calcis. Clin Orthop Relat Res 1987; 221: 286-91.
3. Unni KK, Dahlin DC: Dahlin's bone tumors: general aspects and data on 11,087 cases. Philadelphia: Lippincott-Raven, 1996.
4. Hassani M, Gharehdaghi M, MD, Khooei AR, Ghodsi E, MD, Nazarzadeh H. Bilateral intraosseous tumor of the calcaneus with imaging-pathologic discordance. A case report and literatures review. Arch Bone Jt Surg 2014; 2(3): 238-42.
5. Campbell RS, Grainger AJ, Mangham DC, et al. Intraosseous lipoma: report of 35 new cases and a review of the literature. Skeletal Radiol 2003; 32(4): 209-22.
6. Thornhill RE, Golfam M, Sheikh A, Cron GO, White EA, Werier J, Schweitzer ME, Di Primio G. Differentiation of lipoma from liposarcoma on MRI using texture and shape analysis. Acad Radiol 2014; 21(9): 1185-94. doi: 10.1016/j.acra.2014.04.005.
7. Miles KA, Ganeshan B, Hayball MP. CT texture analysis using the filtration-histogram method: what do the measurements mean? Cancer Imaging 2013; 13(3): 400-6. Doi: 10.1102/1470-7330.2013.9045.
8. Kierans AS, Rusinek H, Lee A, Shaikh MB, Triolo M, Huang WC, Chandarana H. Textural differences in apparent diffusion coefficient between low- and high-stage clear cell renal cell carcinoma. Am J Roentgenol; 203(6): W637-44. doi: 10.2214/AJR.14.12570.
9. De Falco Alfano D, Abousiam RN, Valpiani G, Florio A, Duati R, M. Totaro, Giganti M. Texture analysis of clear cell renal clear cell carcinoma. Journal of Healt & Medical Informatics http://dx.doi.org/10.4172/2157-7420.S1.006.
10. Schajowicz F. Tumors and tumorlike lesions of bone: pathology, radiology and treatment. 2nd ed. Springer-Verlag; 1994.
11. Dahlin DC, Unni KK. Bone tumors: general aspects and data on 8,542 cases. 4th ed. Charles C Thomas; Springfield IL: 1986.
12. Palczewski P, Swiątkowski J, Gołębiowski M, Błasińska-Przerwa K. Intraosseous lipomas: A report of six cases and a review of literature. Pol J Radiol 2011; 76(4): 52-9.
13. Chow LT, Lee KC. Intraosseous lipoma. A clinicopathologic study of nine cases. Am J Surg Pathol 1992; 16: 401-10.
14. Heir GM, Geron PR. Mandibular intraosseous lipoma related to trigeminal neuropathy. Clin Prev Dent 1983; 5: 13-5.
15. Miller WB, Ausich JE, McDaniel RK, et al. Mandibular intraosseous lipoma. J Oral Maxillofac Surg 1982; 40: 594-6.
16. Chaipond Teekhasaenee, Koji Kita, Kenji Takegami, Eiji Kawakita, Toshihiko Sakakibara and Yuichi Kasai. Intraosseous lipoma of the third lumbar spine: a case report. Journal of Medical Case Reports 2015; 9: 52. DOI 10.1186/s13256-015-0528-5.
17. Bin Z, Xiao-guang L, Zhong-jun L, Liao H, Liang J, Feng W. Intraosseous lipoma of adjacent upper cervical vertebral bodies with significant paravertebral expansion. Chin Med J 2010; 123(21): 3160-2.
18. Lin N, Ye Z-M, Li W. Intraoseous lipoma of the pelvis and spine: two cases reports. Orthop Surg 2009; 1: 326-9.
19. Kamekura S, Nakamura K, Oda H, Inokuchi K, Iijima T, Ishida T. Involuted intraosseous lipoma of the sacrum showing high signal intensity on T1-weighted magnetic resonance imaging (MRI). J Orthop Sci 2002; 7: 274-80.
20. Pande KC, Ceccherini AF, Webb JK, Preston BJ. Intraosseous lipomata of adjacent vertebral bodies. Eur Spine J 1998; 7: 344-7.
21. Williams CE, Close PJ, Meaney J, Ritchie D, Cogley D, Carty AT. Intraosseous lipomas. Clin Radiol 1993; 47: 348-50.
22. Milgram JW. Involuted intraosseous lipoma of sacrum. Spine 1991; 16: 243-5.
23. Ehara S, Kattapuran SV, Rosenberg AE. Case report 619. Intraosseous lipoma of the sacrum. Skeletal Radiol 1990; 19: 375-6.
24. Matsubayashi T, Nakajima M, Tsukada M. Case report 118. Skeletal Radiol 1980; 5: 131-3.
25. Hanelin LG, Sclamberg EL, Bardsley JL. Intraosseous lipoma of the coccyx. Report of a case. Radiology 1975; 114: 343-4.
26. Zorn DT, Cordray DR, Randels PH. Intraosseous lipoma of bone involving the sacrum. J Bone Joint Surg Am 1971; 53: 1201-4.
27. Gero MJ, Kahn LB. Case report 498: Intraosseous lipoma of the di end of the fibula with focal infarction. Skeletal Radiol 1988; 17: 443-6.
28. Greenspan A, Jundt G, Remagen W. Diagnostyka różnicowa w onkologii ortopedycznej. Medipage 2008: 438-42.
29. Barcelo M, Pathria MN, Abdul-Karim FW. Intraosseous lipoma. A clinicopathologic study of four cases. Arch Pathol Lab Med 1992; 116: 947-50.
30. Appenzeller J, Weitzner S. Intraosseous lipoma of os calcis. Case report and review of literature of intraosseous lipoma of extremities. Clin Orthop 1974; 101: 171-5.
31. Jensen NC, Madsen LP, Linde F. Topographical distribution of trabecular bone strength in the human os calcanei. J Biomech 1991; 24: 49-55.
32. Lagier R. Case report 128: lipoma of the calcaneus with bone infarct. Skeletal Radiol 1980; 5: 267-9.
33. Milgram JW. Intraosseous lipomas. A clinicopathologic study of 66 cases. Clin Orthop 1988; 231: 277-302.
34. Ragsdale BD. Polymorphic fibroosseous lesions of bone: an almost sitespecific diagnostic problem of the proximal femur. Hum Pathol 1993; 24: 505-12.
35. Blacksin MF, Ende N, Benevenia J. Magnetic resonance imaging of intraosseous lipomas: a radiologicpathologic correlation. Skeletal Radiol 1995; 24: 37-41.
36. Simpfendorfer CS, Ilaslan H, Davies AM, et al. Does the presence of focal normal marrow fat signal within a tumor on MRI exclude malignancy? An analysis of 184 histologically proven tumors of the pelvic and appendicular skeleton. Skeletal Radiol 2008; 37: 797-804.
37. Stacy GS. An analysis of malignant bone tumors for the presence of internal fat-signal-intensity on magnetic resonance examinations. Skeletal Radiol 2007; 36: 355-75.

38. Kozlowski K, Welshman R. What is it? Intraosseous lipoma in a 13-year-old boy. Br J Radiol 1991; 64: 855-6.
39. Bagatur AE, Yalcinkaya M, Dogan A, Gur S, Mumcuoglu E, Albayrak M. Surgery Is not always necessary in intraosseous lipoma. Orthopedics 2010; 33(5): 306.
40. De Filippo M, Gira F, Corradi D, et al. Benefits of 3D technique in guiding percutaneous retroperitoneal biopsies. Radiol Med 2011 Apr; 116(3): 407-16. doi: 10.1007/s11547-010-0604-2. Epub 2011 Feb 10.
41. Bertolini L, Vaglio A, Bignardi L, et al. Subclinical interstitial lung abnormalities in stable renal allograft recipients in the era of modern immunosuppression. Transplant Proc 2011 Sep; 43(7): 2617-23. doi:10.1016/j.transproceed.2011.06.033.
42. Pescarolo M, Sverzellati N, Verduri A, et al. How much do GOLD stages reflect CT abnormalities in COPD patients? Radiol Med 2008 Sep; 113(6): 817-29. doi: 10.1007/s11547-008-0284-3. Epub 2008 Jul 10.
43. Gafà G, Sverzellati N, Bonati E, et al. Follow-up in pulmonary sarcoidosis: comparison between HRCT and pulmonary function tests. Radiol Med 2012 Sep; 117(6): 968-78. Epub 2012 May 14.
44. Cataldi V, Laporta T, Sverzellati N, De Filippo M, Zompatori M. Detection of incidental vertebral fractures on routine lateral chest radiographs. Radiol Med 2008 Oct; 113 (7): 968-77. doi: 10.1007/s11547-008-0294-1. Epub 2008 Sep 13.

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