CASE STUDY

Myofibroblastic Sarcoma of the Bone: Two Cases

Author

Holgado-Moreno Esperanza [a], Ortiz-Cruz Eduardo José [a], Pozo-Kreilinger José Juan [b], Peleteiro-Pensado Manuel [a], Ortiz-Hernando, Raquel [c]

Affiliations

[a] Deparment Orthopedic surgery Hospital Universitario La Paz. Madrid
[b] Deparment of Pathology Hospital Universitario La Paz. Madrid
[c] Degree in Medicine and Surgery at CEU Universities-CEU San Pablo. Madrid

Correspondence

Holgado-Moreno Esperanza
Deparment Orthopedic surgery Hospital Universitario La Paz. Madrid
Email: eholgadomoreno@gmail.com

Abstract

Low-grade myofibroblastic sarcoma (L-G MFS) is a rare tumour that commonly affects the soft tissues of the head and neck. Its location in bone is very rare and there are few cases published in the literature, which sometimes leads to a wrong initial diagnosis. The following report will give an overview of the clinical, radiological and histological findings in two patients with a very unusual bone sarcoma, which are more often found in the soft tissues. Treatment options and outcome will be discussed.

On excision of any sarcoma tumor, surgeons should be aware of the potential risk for erroneous management of malignancy. If not, careless surgery may render the treatment protocol complicated and additional bed tumor resection with poor function and prognosis.

Low-grade myofibroblastic sarcoma (L-G MFS) requires a wide resection to avoid local recurrence and distant metastases.

We present two cases with an initial diagnosis of giant cell tumor and fibrous dysplasia respectively, which were diagnosed as low-grade bone myofibroblastic sarcoma after clinical, radiological y pathological studies.

Keywords: Sarcoma, myofibroblastic, low grade, femur

Introduction

The low-grade myofibroblastic sarcoma is a tumour defined recently (1998) by Mentzel et al. [1] and recognised as a new term by the World Health Organization (WHO) in 2002. It lies within the mesenchymal malignant tumours as a specific type of myofibroblastic tumour. Over one hundred cases are described in scientific literature [1-5].

Its incidence is higher in adults, with a preference for location in soft tissues, although in rare cases it can be located in bone [3, 4, 6-11]. We present two cases of intraosseous origin in the femur, which were initially treated in other hospitals with erroneous clinical judgement and were later forwarded to the Bone and Soft Tissue Tumour Unit of Hospital Universitario La Paz, in Madrid.

This review of our two cases and the literature is focused on the definitive anatomopathological diagnosis and the surgical treatments conducted.

Case 1

A twenty-year old woman presents an initial clinical history of one year of ongoing knee pain with spontaneous and occasional hemarthrosis (no history of trauma). Through plain radiographs an osteolytic intraosseous lesion could be observed, which occupied the entire central and posterior distal metaphyseal region of the femur. Its geographic margins (IC type) were relatively well defined with a slightly sclerotic rim in the basal area which disappears and is badly defined in the most proximal part (Fig. 1A). Magnetic resonance imaging (MRI) with gadolinium revealed a lesion infiltrated the posterior femoral cortical bone and there was no calcified matrix, nor periosteal reaction (Fig. 1B). The whole-body skeletal scintigraphy with Tc^{99} showed a solitary increase uptake in the distal femur. Analysis of imaging tests suggested a giant cell tumour of bone.

In her hospital two percutaneous biopsies were conducted, but no definite histological diagnosis could be established in either due to an insufficient sample. Without anatomopathological confirmation, it was decided that surgical intervention with clinical-radiological judgement of giant cell tumour should follow, conducting an extended intra-lesional tumour resection. Surgery consisted of making a wide window through the thinned cortex of bone, curettage, high-speed burring and followed phenolization of the resulting osseous cavity. The osseous defect was filled with allograft in the form of cancellous chips (Fig. 1C).

The anatomopathological study of the tissue obtained ruled out giant cell tumour and a diagnosis of a low-grade myofibroblastic sarcoma (L-G MFS) was suggested. It was then decided to refer the patient to our hospital, where after reviewing the anatomopathological sample, the diagnosis of low-grade myofibroblastic sarcoma was confirmed. A new MRI was conducted were post-surgical changes and probable tumour remains were observed (Fig. 1D), without metastasis in the extension study.

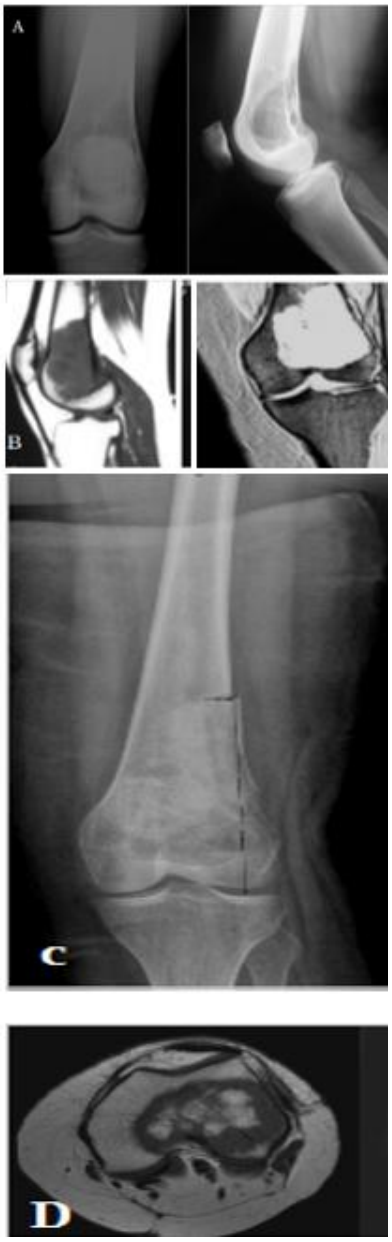

Figure. 1: A. Plain radiographs that shows IC type geographic lytic lesion which occupies entire central and posterior distal femoral metaphysis. It presents a slightly sclerotic basal rim which disappears and remains poorly defined at the top. Endosteal erosion with ridges without clear cortical tear.
B. The MRI confirms findings of plain radiography. Sagittal view Se-T1 with a hypo-intense metaphyseal lesion of slightly infiltrative rims which appears to invade the posterior cortical bone. Coronal T2 section shows the invasion of external cortical bone.
C. Secondary radiological changes after intra-lesional resection (curettage) of the lesion through external cortical window with cancellous filling.
D. Control MRI shows the likely persistence of tumour, plausible in the peripheral zones. Axial SE-T1 with hyper-intense granular areas which correspond to cancellous graft surrounded by hypointense substance.

After presenting the case in a multi-disciplinary sarcoma committee and due to the previous surgical treatment conducted on the patient, re-intervention was decided. The surgery consisted in wide intra-articular tumour resection of distal femur (14cm) and reconstruction with knee tumoral prostheses (Fig. 2 A)

The excision consisted in the distal segment of the femur, of 13.5 cm in length, without external deformity (Fig. 2 B). The sagittal section showed at intraosseous level an area of 9 x 4,5 x 4 cm of rarefaction yielding identification of individualised bone fragments presumably pertaining to the previous allograft separated from the host bone by a red or greyish peripheral rim.

In addition, in the external femoral condyle region, near the articular cartilage, a greyish area of about 1,5cm in diameter stood out, different from the macroscopic alteration previously described. The 2,6 cm of proximal femoral diaphysis were found free of macroscopic alteration.

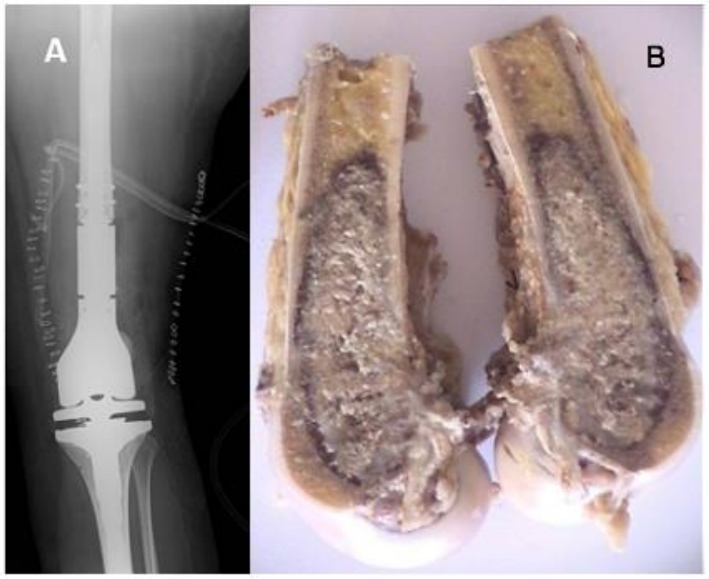

Figure. 2: A. Reconstruction with tumour prosthesis of distal femur. Radiological control. B. Distal femur resection piece. Sagittal section

CASE 2:

A twenty-one year old woman underway with a pathological proximal femur fracture with no previous traumatism. In the simple X-ray study, a pathological subtrochanteric fracture could be observed on an osteolytic, geographic lesion (Type 1C) (Fig. 3 A). In the computed tomography (CT scan) the pathological fracture is confirmed, presenting a thinning cortical, with no calcified matrix, nor localised periosteal reaction in the lesser trochanter and subtrochanteric region. The imaging test suggested a low-agressive lesion, bringing about differential diagnosis of simple bone cyst or fibrous dysplasia. A percutaneous biopsy was conducted in her referring hospital where no neoplasm was found, making the test inconclusive. In light of this, surgical intervention was carried out to conduct an excisional biopsy and osteosynthesis with a long intramedullary nail. During the postoperative period, the patient developed pain in the thigh which receded slightly with painkillers, and six months after surgery an increase in the soft tissues of the affected thigh was observed. Additionally, the X-rays showed a progression of bone destruction with complete resorption of the lesser trochanter as well as a periosteal reaction (Fig. 3 B), hence the patient was referred to our hospital.

An MRI was carried out where an osteolytic lesion could be observed in the proximal metaphysodiaphysary region associated to a great soft tissue mass (Fig. 3C). A new percutaneous core needle biopsy (CNB) was conducted which suggested it was a low-grade myofibroblastic sarcoma or fibrosarcoma, therefore an intra-articular wide resection of the proximal femur was considered and a reconstruction with tumoral prosthesis of the proximal femur (Fig. 3D). The diagnosis of low-grade myofibroblastic sarcoma (MFS) was confirmed in the definitive study of the resected tumorous piece.

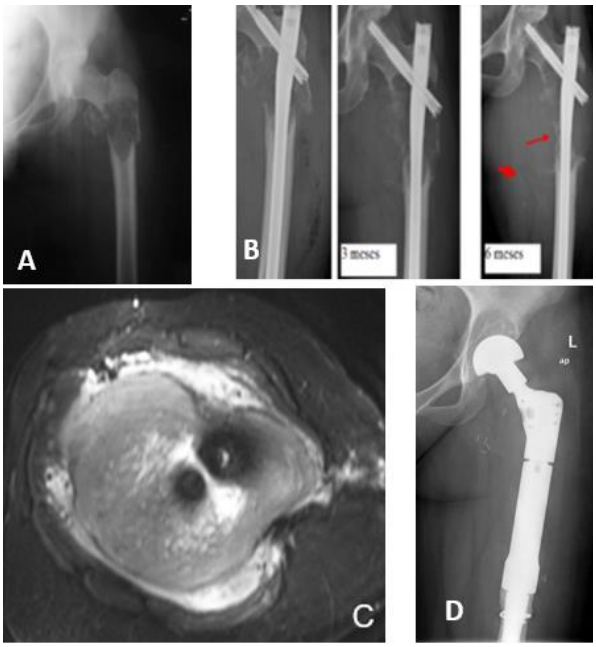

Figure. 3: A. Plain radiograh show a pathological fracture of proximal femoral metaphysis affecting the lesser trochanter. Geographic lytic lesion appears to contain calcified matrix in ground-glass opacity. IC type superior margin poorly defined.
B.Osteosynthesis with long intramedullary nail. Six months of monitoring showed progressive bone destruction with complete resorption of lesser trochanter and the appearance of a broken periosteal reaction (see thin arrow) as well as a mass of soft tissue toward the adductor region (see thick arrow).
C. MRI showing mass of soft tissue and the extension of the area affected by the tumour. Axial section in STIR with artefacts owing to osteosynthesis, showing a great mass of soft tissue surrounding the femur, with peritumoral edema displacing the muscle mass.
D. Radiological control of the tumoral prosthesis of the proximal femur, six months after surgery with no evidence of signs of local relapse.

The excision consisted in one third of the proximal femur, with a length of 20 cm (Fig. 4). A metaphysodiaphysal tumour of 16 x 13 x1 3 cm was identified, with an intraosseous epicentre and an extension to soft tissue where it appeared well defined but not encapsulated. When cut, the tumour was whitish, fascicular, of a firm consistency although not calcified, and contained myxoid areas together with others of cyst-like appearance.

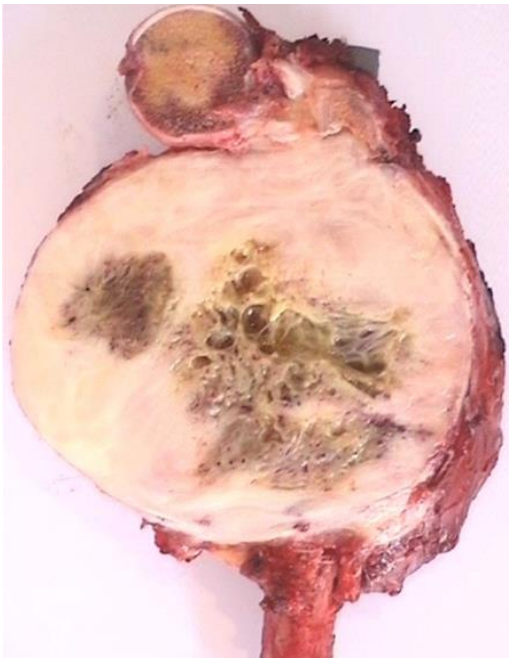

Figure. 4: Case 2. Excision of proximal femur with significant presence of soft tissue

In this patient, after four years of follow up, in a routine chest CT, three pulmonary nodules were identified: one in the right upper lobe of approximately 9 mm, another of 7 mm, and a third, the largest in size, of approximately 15 mm in left lower lobe in subpleural location (Fig. 5 A). After these findings, the case is presented at the Thoracic Surgery Committee, deciding to conduct a left posterolateral thoracotomy and resection of four pulmonary nodules, lung

segmentectomies, two in the left upper lobe and two in the left lower lobe, which are sent for anatomopathological study confirming as a result a pulmonary bilateral metastasis of low-grade myofibroblastic sarcoma in three of the four nodules (Fig. 5 B and C).

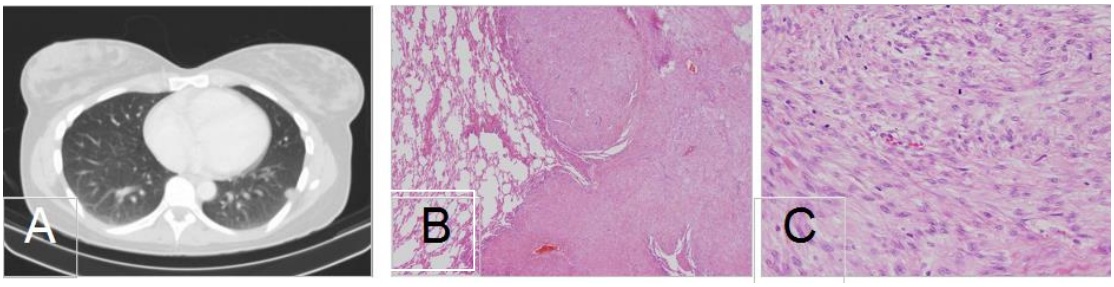

Figure. 5: A Subpleural nodule in left lower lobe. After resection and anatomopathological study, a metastasis of low-grade myofibroblastic sarcoma (MFS) was confirmed. B Image slightly magnified where lung parenchyma can be identified, infiltrated by a tumour of nodular architecture. C At high power field confirms metastatic nature of the lesion, with identical microscopic features to the original lesion in the proximal femur.

Follow up

In both cases there were no complications in the respective postoperative periods and the patients are currently monitored on an outpatient basis with no signs of relapse eleven years after surgeries. After being presented at the sarcoma committee, adjuvant treatments such as chemotherapy and radiotherapy were discarded in both patients.

Anatomopathological findings:

The histological tissue originating from the excision in both patients was microscopically similar, both show a homogenous mesenchymal fusocelular tumour, fascicular pattern and of moderate cellular density (Fig. 6A-D).

The neoplastic cells were elongated with scarce eosinophilic cytoplasm and an elongated slightly wavy and with discretely irregular point-like ends nucleus with a small nucleolus.

The mitotic activity in both cases was low, between 3-4 mitosis in ten high power fields. In the focal point of the residual tumour identified in the external femoral condyle of case 1, an infiltrative tumour pattern was observed among the trabeculae of the host bone. In case 2, small focal points of cartilaginous differentiation could be observed in a thinning cortical bone belonging to the fracture callus.

Immunohistochemically (Fig.6E), in both cases, the neoplastic cellularity expressed intense positivity for vimentin and alfa smooth muscle actin, and negativity toward desmin. Furthermore, in case 1, it presented a slight positivity toward calponin. The nuclear proliferation index (Ki-67) ranged between 5% in case 1 and 10% in case 2.

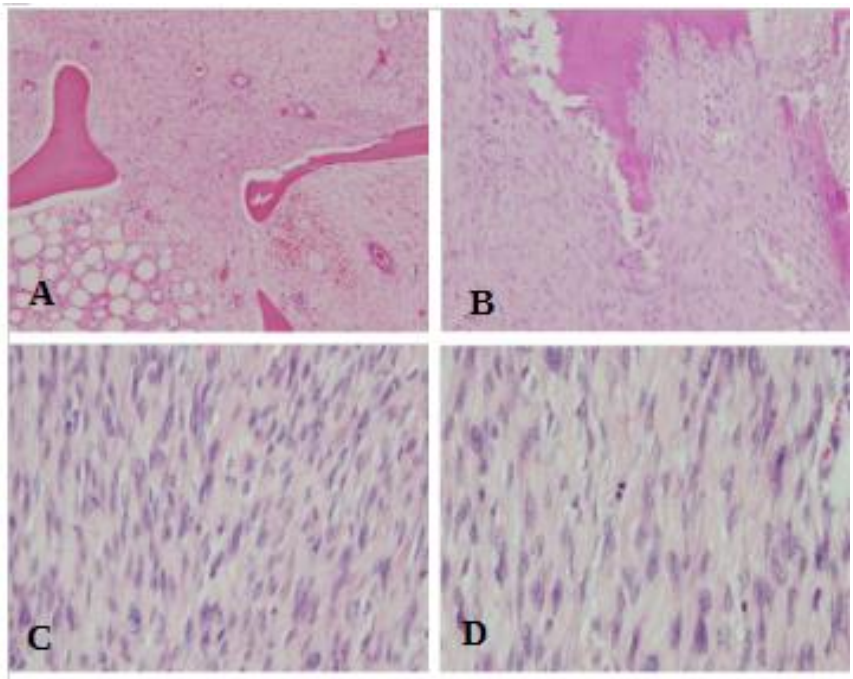

Figure. 6: A. Infiltrative pattern, B. Osteolytic spindle cell proliferation, C y D. Shows spindle tumour cells with low cytological atypia, with eosinophilic cytoplasms and an oval and wavy nucleus with sometimes blunt and other pointed ends, clumpy chromatin and occasional nucleoli

DISCUSSION

The low-grade myofibroblastic sarcoma (MFS) is a tumour with greater prevalence in adult patients [2, 6-7], although some cases have been documented in children [15, 16]. There is a slight prevalence in males [2, 4]. The most frequent location is soft tissue [4, 8], especially head and neck, including pharynx and the oral cavity, although it can also appear in other areas such as extremities and torso. The location of low-grade MFS in bone is rare [2] and that makes our two cases exceptional.

Both cases presented here have osseous origin affecting the femur. There are few published cases of low-grade MFS (Table 1.) Schenker et al. conducted a systematic review of a series of cases and case reports of low-grade MFS in extremities [18] which have been published and collated in the PRISMA website (Preferred Reporting Items for Systematic Review and Meta-Analysis).

Table 1. Diagram of published MFS cases of osseous location

Cases	**Age**	**Sex**	**Location**	**Clinical**	**Initial Treatment**	**Relapse**	**Definitive treatment**	**Mets**
Case 1	20	F	Distal femur	Pain	IR	No	WR	Lung

Case 2	21	F	Proximal femur	Pathological fracture	IR	No	WR	-
Watanabe et al. 2001	60 63 66 71	M F F F	Distal femur Distal femur Iliac fossa Illiac fossa		WR+CT IR WR+CT WR+CT	- twice - -	- WR - -	- - - Lung
Mongomeri et al. 2001	65	F	Tibia		IR	Local	WR	-
Bisceglia et al. 2001	24 49	M M	Jaw Jaw	Mass Mass	WR + RT WR + RT	No	-	- -
San Miguel et al. 2004	51	F	Distal phalanx ring finger	Painless mass	AMP	No	-	-
Fernández-Aceñero 2004	24	M	Distal femur	Mass	AMP	No	-	-
Meng et al. 2007	47 42 30 14 32 30	M F M M M F	Femur Femur Skull Femur Skull Femur	Painless mass	WR+CT WR+CT+ RT WR+CT WR+CT WR+CT	once once twice once once twice		- - - - - -
Niedzielska et al. 2009	54	M	Jaw	Mass	WR	No	-	-
Arora R et al. 2010	38	F	Femur	Painless mass	WR	No	-	-
Humphries et al. 2010	15	F	Sacrum	Mass	WR	No	-	-

Saito T et al. 2012	50	F	Femur	Painless mass	WR	No	-	-
Hadjigeorgiou G 2016	55	M	Thoracic spine	Paresis in MMII	WR	No	-	-
Wang L et al. 2019	NA NA NA NA NA	NA NA NA NA NA	Scapula Femur Shoulder Femur Femur	Mass Osteolytic lesion NA Osteolytic lesion Osteolytic lesion	NA NA NA NA NA	 Yes Yes		Lung Lung Lung Ósseous

Key: IR: intra-lesional resection, WR: wide resection, CT: Chemotherapy, RT: radiotherapy, AMP: amputation.

Clinically the L-G MFS usually manifests as a painless mass of progressive growth. In our cases the initial symptoms were pain in the first case and pathological fracture in the second case.

Osteolytic lesions are often observed [4,6,12] radiologically without peripheral sclerosis, lacking calcified matrix nor periosteal reaction. However, cortical thinning is common, as observed in our cases. For the diagnosis of this tumour, anatomopathological findings are essential.

Histologically, a mesenchymal fusiform-cell tumour is identified with variable cellular density arranged in a fascicular pattern [4, 6, 8]. Tumour cells have a scarce wavy and pale eosinophilic cytoplasm. The nucleous are small, uniform and ovoid, with vesicular chromatin, presenting on occasion a small nucleolus. The mitoses and pleomorphism are discrete, as are focal points of necrosis [2]. Trabecular bone infiltration is commonly observed, as well as formation of reactive bone tissue.

The stroma presents a collagenous matrix with occasional lymphocytes and plasmatic cells [2]. Inmunohistochemical study plays a key role in differential diagnosis with other fusocellular tumours. While leiomyosarcoma has an intense and diffused immunopositivity to alfa smooth muscle actin, desmin and caldesmon, MFS is positive, although not as intense and diffuse, to smooth muscle actin and calponin, and negative to caldesmon.
The fibrosarcoma does not show the inmunoprofile previously mentioned and displays a classic herringbone pattern.

Regard treatment [8,9,10,11], a wide tumour resection with the goal of obtaining free margins is the objective and it is not indicated neo or adjuvant treatments as chemotherapy or radiotherapy. On the other hand, for high-grade MFS it would be necessary to complement surgical treatment with adjuvant treatment. The local recurrences in this tumour are frequent, up to 33% according to Eyden's review [2], especially when the tumour is located in bone or an incomplete resection has taken place. Relapses can even take place after a long time with no symptoms,

which is why prolonged monitoring is required for these patients.
The metastases are rare (10%) [2,4,8] and are normally located in the lung [12]. Despite the radiological appearance of benign neoplasia, an oncological staging protocol of patients must always be followed and a reliable anatomopathological result of the biopsy must be obtained.

Initially, in both cases, the clinical radiological impression of benign led surgeons to inadequate treatments, as were the intra-lesional tumor resections in both cases, and the results of the percutaneous biopsies were not significant, having to clarify the diagnosis prior to surgery [13]. The Henry Mankin [14] studies and the MSTS (Musculoskeletal Tumour Society) show that greatest mistakes in biopsy take place in centres which are not qualified to conduct them.

In future, an effort will be made to research into genetic aspects, both at cellular and molecular levels, to deepen knowledge of this entity, as for example, changes in regions 12p11 and 12q13-q22 or ring chromosomes which have already been identified in low-grade sarcomas [2].

In sum, the low-grade MFS is a rare tumour and its bone location is exceptional. However, care must be taken to correct diagnosis and appropriate therapeutic treatment. The experience of the multidisciplinary team is essential.
Despite the low frequency of these tumours, the general orthopaedic surgeon must be familiar with these lesions and know when patients must be forwarded to a sarcoma specialist center. The clinical management includes diagnostic tests, adjuvant therapies, surgical treatment and analysis of the resected samples, which must be conducted by a group of specialists with specific training in sarcomas.

Conclusion:

- Because the differential diagnosis of this rare tumor includes bening tumors and others types of sarcomas, a pathological diagnosis is essential which requires a combination of complete histological and immunohistochemical examination.

- Due to the diagnostic difficulty from radiology and pathological anatomy point of view, it is essential that patients with a radiological diagnosis of suspected primary aggressive bone tumour as the L-G MFS has to be referred prior to the biopsy to expert sarcoma centers.
- Sarcomas are infrequent diseases and are classified as rare disease. We show in the article, that there are tumours such as low grade myofibroblastic sarcoma that are still very rare among the uncommon of this type of neoplasm and mainly when they are located in the bone.
- This makes us think that the support of skilled personnel working in a multidisciplinary way in expert sarcoma centres is essential and mandatory.
- An inappropriate pathological diagnosis leads to an indication of wrong surgery with consequences that can be fatal for a patient, which fortunately in our two cases were solved, but it does not always happen this way. In the two cases presented, they were initially treated in non-expert centers, with two major predictable errors. In the first one, an aggressive surgery should not be performed without a pathological diagnosis, and even less without

performing a biopsy. In the second case, despite having a previous biopsy, the tumour was not properly diagnosed, leading to erroneous treatment.

- For adequate diagnosis and therapeutic management, an assessment of clinical, radiological, and pathological characteristics of the case are required, with a fluid communication of those who make up the multidisciplinary team.

Credit authorship contribution statement

All authors have contributed significantly, and that all authors are in agreement with the content of the manuscript

Holgado-Moreno Esperanza: Writing - original draft, Writing - review & editing. Ortiz-Cruz Eduardo José: Conceptualization, Writing - review & editing, Formal analysis. Pozo-Kreilinger José Juan: Writing - review & editing. Peleteiro-Pensado Manuel: Writing - review & editing.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request

Conflict of interes

All author disclose any financial and personal relationships with other people or organizations that could inappropriately influence our work

References:

1. Mentzel T, Dry S, Katenkamp D, Fletcher CD. Low-grade myofibroblastic sarcoma: analysis of 18 cases in the spectrum of myofibroblastic tumors. *Am J Surg Pathol*. 1998 Oct;22(10):1228-38. DOI: 10.1097/00000478-199810000-00008.

2. Eyden B, Banerjee SS, Shenjere P, Fisher C. The myofibroblast and its tumours. *J Clin Pathol*. 2009 Mar;62(3):236-49. DOI: 10.1136/jcp.2008.061630. Epub 2008 Oct 17.

3. Montgomery E, Goldblum JR, Fisher C. Myofibrosarcoma: a clinicopathologic study. *Am J Surg Pathol*. 2001 Feb;25(2):219-28. DOI: 10.1097/00000478-200102000-00010.

4. Meng GZ, Zhang HY, Bu H, Zhang XL, Pang ZG, Ke Q, Liu X, Yang G. Myofibroblastic sarcomas: a clinicopathological study of 20 cases. *Chin Med J (Engl)*. 2007 Mar 5;120(5):363-9.

5. Huang G, Ye F, Fu M, Chen Y. [Clinical analysis of low grade myofibroblastic sarcoma in head and neck]. *Lin Chung Er Bi Yan Hou Tou Jing Wai Ke Za Zhi*. 2014 Aug;28(15):1148-50.

6. Watanabe K, Ogura G, Tajino T, Hoshi N, Suzuki T. Myofibrosarcoma of the bone: a clinicopathologic study. *Am J Surg Pathol*. 2001 Dec;25(12):1501-7. DOI: 10.1097/00000478-200112000-00005.

7. Bisceglia M, Tricarico N, Minenna P, Magro G, Pasquinelli G. Myofibrosarcoma of the upper jawbones: a clinicopathologic and ultrastructural study of two cases. *Ultrastruct Pathol*. 2001 Sep-Oct;25(5):385-97. DOI: 10.1080/019131201317101261.

8. San Miguel P, Fernández G, Ortiz-Rey JA, Larrauri P. Low-grade myofibroblastic sarcoma of the distal phalanx. *J Hand Surg Am*. 2004 Nov;29(6):1160-3. DOI: 10.1016/j.jhsa.2004.05.003.

9. Niedzielska I, Janic T, Mrowiec B. Low-grade myofibroblastic sarcoma of the mandible: a case report. *J Med Case Rep*. 2009 Aug 10;3:8458. DOI: 10.4076/1752-1947-3-8458.

10. Arora R, Gupta R, Sharma A, Dinda AK. A rare case of low-grade myofibroblastic sarcoma of the femur in a 38-year-old woman: a case report. *J Med Case Rep*. 2010 Apr 28;4:121. DOI: 10.1186/1752-1947-4-121.

11. Saito T, Mitomi H, Kurisaki A, Torigoe T, Takagi T, Suehara Y, Okubo T, Kaneko K, Yao T. Low-grade myofibroblastic sarcoma of the distal femur. *Int J Surg Case Rep*. 2013;4(2):195-9. DOI: 10.1016/j.ijscr.2012.11.017. Epub 2012 Nov 23.

12. Wang L, Li LX, Chen DQ, Yang L, Li SK, Cheng C. Low-grade Myofibroblastic sarcoma: clinical and imaging findings. *BMC Med Imaging*. 2019 May 2;19(1):36. DOI: 10.1186/s12880-018-0287-z.

13. Hadjigeorgiou GF, Samaras V, Varsos V. Low-grade myofibroblastic sarcoma of the thoracic spine: report of an extreme rare case. *Br J*

Neurosurg.2017Dec;31(6):731-733. DOI:10.1080/02688697.2016.1220505. Epub 2016 Aug 18.

14. M Fernández-Aceñero, A Sanz-Laguna, J Carrascoso-Arran, P López-Criado. Low-Grade Myofibroblastic Sarcoma Of The Bone. *The Internet Journal of Pathology*. 2004 Volume 4 Number 1.

15. Mankin HJ, Lange TA, Spanier SS. The hazards of biopsy in patients with malignant primary bone and soft-tissue tumors. *J Bone Joint Surg Am*. 1982 Oct;64(8):1121-7.

16. Mankin HJ, Mankin CJ, Simon MA. The hazards of the biopsy, revisited. Members of the Musculoskeletal Tumor Society. *J Bone Joint Surg Am*. 1996 May;78(5):656-63. DOI: 10.2106/00004623-199605000-00004.

17. Agaimy A, Wünsch PH, Schroeder J, Gaumann A, Dietmaier W, Hartmann A, Hofstaedter F, Mentzel T. Low-grade abdominopelvic sarcoma with myofibroblastic features (low-grade myofibroblastic sarcoma): clinicopathological, immunohistochemical, molecular genetic and ultrastructural study of two cases with literature review. *J Clin Pathol*. 2008 Mar;61(3):301-6. DOI: 10.1136/jcp.2007.048561. Epub 2007 May 18.

18. Eisenstat J, Gilson T, Reimann J, Sampson B. Low-grade myofibroblastic sarcoma of the heart causing sudden death. *Cardiovasc Pathol*. 2008 Jan-Feb;17(1):55-9. DOI: 10.1016/j.carpath.2006.12.006. Epub 2007 Feb 22.

19. Humphries WE 3rd, Satyan KB, Relyea K, Kim ES, Adesina AM, Chintagumpala M, Jea A. Low-grade myofibroblastic sarcoma of the sacrum. *J Neurosurg Pediatr*. 2010 Sep;6(3):286-90. DOI: 10.3171/2010.5.PEDS09289.

20. Astrid Schenker, Burkhard Lehner, Ewgenija Gutjhar et al. Low-Grade Myofibroblastic Sarcoma (LGMS) of the Extremities – A Systematic Review and Case Report, 24 July 2020, PREPRINT (Version 1) available at Research Square [+https://doi.org/10.21203/rs.3.rs-38829/v1+]

