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# Liposclerosing Myxofibrous Tumor in Tibia
## - A Case Report and Review of the Literature -

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Liposclerosing myxofibrous tumor (LSMFT) is a benign fibro-osseous lesion that is characterized by a complex mixture of histologic elements, including its fibrous dysplasia-like features and its lipoma, myxofibroma, xanthoma and pseudo-Paget's bone patterns. However, this lesion is considered by some researchers as a variant of fibrous dysplasia or as the non-specific end result of degenerative change, while it is considered by others as a definite clinicopathologic entity. Here, we report on a case of LSMFT occurring in tibia, which is a very uncommon location for this tumor, and we review the related literatures. The case presented here shares features with those described for LSMFT, except for the location of this tumor. We believe that more studies on a larger scale that compare LSMFT with other benign bone lesions, including fibrous dysplasia, are required to clarify the origin and behavior of this lesion.

**Key Words :** Liposclerosing myxofibrous tumor; Fibrous dysplasia; Tibia

Liposclerosing myxofibrous tumor (LSMFT) is a benign fibro-osseous lesion that has a marked predilection for the intertrochanteric region of the proximal femur.[1,2] It is characterized by a complex mixture of histologic elements that includes lipoma, fibroxanthoma, myxoma, myxofibroma, fibrous dysplasia-like features, cyst formation, fat necrosis, ischemic ossification, and, on rare occasions, the presence of cartilage.[3-5] Some consider LSMFT to be a genuine clinicopathologic entity[4,5] and the others regard it as a variant of fibrous dysplasia or as a nonspecific end result of degenerative changes in other benign bone lesions.[1,8] Herein, we report on the radiographic and histologic appearance of one case of LSMFT, and we also review the related literatures.

## CASE REPORT

A 61-year-old male was admitted to our hospital for an incidentally discovered mass that was detected on radiography and he complained of mild tenderness without any history of localized trauma to that area. He had no previous disease history. The mass was located in the proximal diaphysis of the left tibia, extending to the metaphysis. The radiographs revealed about a 10 cm-sized lytic and radiolucent lesion that displayed an irregular sclerotic rim, thinning of the cortices, an internal ground glass appearance, and multiple irregular nodular calcifications (Fig. 1A). This lesion showed a relatively homogeneous low T1 and high T2 signal intensity in most areas except for the nodular calcifications or trabeculae which displayed multiple internal irregular nodular enhancements (Fig. 1B). The patient underwent curettage of the lesion. The microscopic examination showed fragments of hypocellular myxofibrous tissue (Fig. 2A), and some of them entrapped fat and foam cells. In some of these fragments, fibrous dysplasia-like woven bone trabeculae without osteoblastic rimming were seen (Fig. 2B). There were also areas of sclerotic woven bone with mosaic cement lines (pseudo-Paget's pattern, Fig. 2C) and areas of ischemic bone containing coalescent calcifications (Fig. 2D).

## DISCUSSION

LSMFT has been described in the literature under various terms

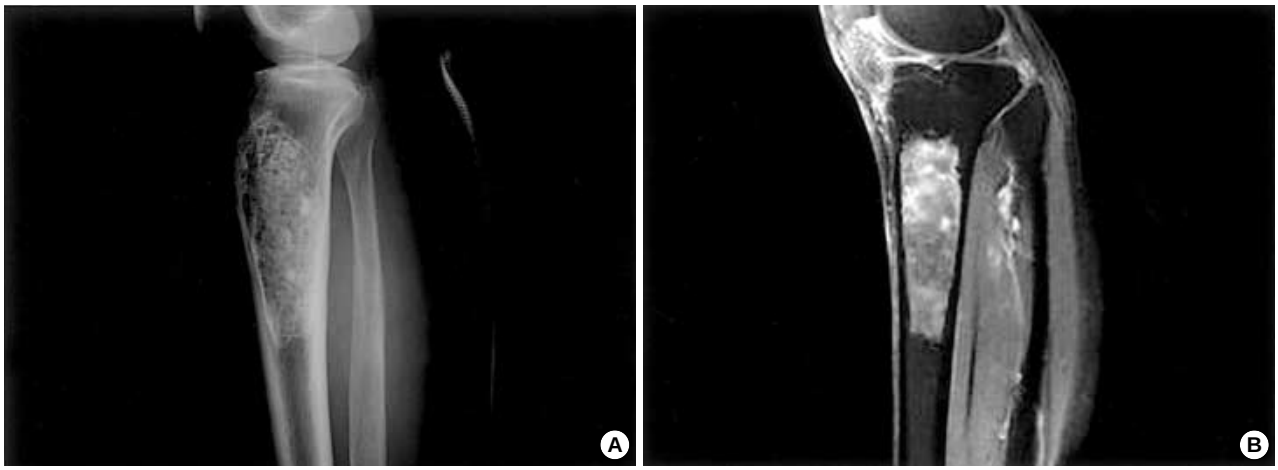


Fig. 1. (A) Anteroposterior radiograph of the left tibia reveals an about 10 cm-sized lytic and radiolucent lesion, which showed irregular sclerotic rim, thinning of the cortices, internal ground glass appearance, and multiple irregular nodular calcifications. (B) T2-weighted MR image shows a relatively homogeneous high signal intensity and multiple internal irregular nodular enhancement along the margins of calcification and trabeculations.

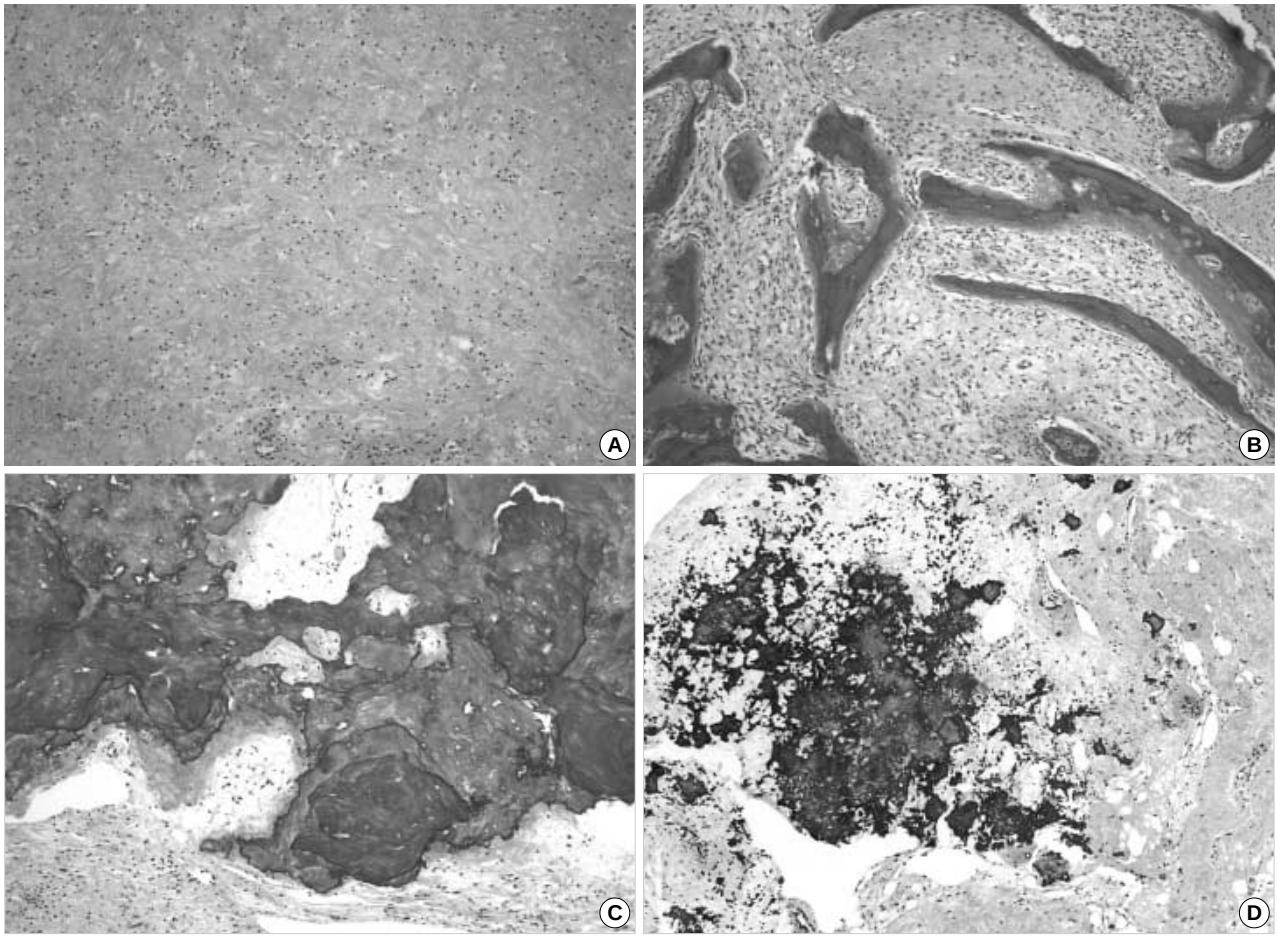


Fig. 2. (A) The stroma is hypocellular and myxofibrous. (B) Fibrous dysplasia-like woven bone trabeculae without osteoblastic rimming are seen. (C) Areas of sclerotic woven bone with mosaic cement lines (pseudo-Paget's pattern) are also found. (D) This lesion shows ischemic areas containing coalescent calcification.

including LSMFT, polymorphic fibro-osseous lesion, polymorphic fibrous dysplasia, and atypical fibrous dysplasia.[1] Their complex histologic components can be seen in varying proportions, and all of these lesions represent a common consultative problem.

The age range of the affected patients varies from the 2nd to the 7th decade of life; the mean age being about 40 years. There is an equal incidence of this lesion in both sexes.[6] Although the femur, ilium, humerus, and rib may be involved, it is widely accepted that the proximal portion of the femur is the most common location.[4]

The largest study that has been done on this entity has been provided by Ragsdale[5] and he described the complex histologic features such as lipomatous areas, the intralesional bone having lamellar, woven, pseudo-Paget's and ischemic patterns, the fibrous and myxofibrous areas, the curvilinear and circular ossicles similar to fibrous dysplasia, the fibroxanthomatous areas, the hypocellular fibrous areas with mineral concretions, the storiform patterns and the frequent cystic changes. However, Gilkey[3] has stated that a fatty component and cystic change were rarely encountered in his experience with 40 cases. Ragsdale also argues that the proximal femur, which is the most common site for LSMFT, is a very uncommon location for the fibrous dysplasia. Yet, the earlier literature has shown a contradictory result that the fibrous dysplasia is relatively common at this location and the incidence of monostotic and polyostotic fibrous dysplasia is about 23% and 90%, respectively.[7]

Kransdorf[4] has described LSMFT as a well-defined lytic bone lesion with sclerotic margins and mild expansile remodeling. He also admits that the fibrous dysplasia may show all the histologic components described for LSMFT, but it exceeds the usual features of fibrous dysplasia. Heim-Hall & Williams[1] have reported on four cases of LSMFT and all of the lesions were located in the proximal femur. The histologic features they described overlap with fibrous dysplasia to varying degrees, but the complex patterns noted for LSMFT exceeded the pattern that are typically seen in fibrous dyplasia. When researching 2 cases of LSMFT, Matsuba *et al*.[8] have recently disclosed the missense point mutation Gs alpha at the Arg201 codon, which has been identified in cases of McCune-Albright syndrome as well as polyostotic and monostotic fibrous dysplasia of the bone.[9-11] As a result, they suggested that a subset of LSMFT is a variant form of fibrous dysplasia. However, the relationship between LSMFT and fibrous dysplasia is still not clear.

The case presented here shares all the features that have been described in LSMFT except for the location of tumor. It is interesting that the present case shows an uncommon tumor location in the proximal diaphysis of the left tibia, extending to the metaphysis. Up to date, only two cases of LSMFT located in tibia have been reported in the literature by Ragsdale,[5] and one of those cases was diaphyseal in location.

There have been some questions and suggestions about the origin of LSMFT and also for the reason why the proximal femur is the site of predilection, but there are currently no demonstrable answers to these questions. Kransdorf[4] suspected that this tumor's origin is a combination of changes in the altered fat of a partially involuted lipoma or lipogenic lesion of the bone with superimposed proliferative changes. Ragsdale[5] suggested that this lesion is acquired as an aberration of the normal growth and development seen in childhood, and the precursor lesions seen in individuals younger than 20 years that show dominant cellular fibroxanthomatous components undergo internal modifications over the lifespan of the tumor. This theory is mainly based on the findings that the fully developed lesion showing all of the complex histologic elements is not found in young children and the radiographic appearance and histologic composition of the lesions are remarkably different between young and elderly adults. Heim-Hall & Williams[1] have concluded that some of the LSMFTs probably represent a traumatized variant of fibrous dysplasia because these lesions show similar histologic features, although only in part, and most of their cases were reported to be associated with incomplete fracture or stress fracture. They also mentioned that the increased mechanical stress unique to the proximal femur explains the predominant occurrence of LSMFT at this location.

Malignant transformation of these tumors to fibrosarcoma, malignant fibrous histiocytoma and other soft tissue malignancies has been well documented.[5] Several other studies[4,5] support the view that the malignant transformation is likely to be secondary to its involutional and ischemic changes and the tumor's malignant progression geneally occurs in the reactive border of the ischemic bone marrow. The malignant transformation reported in the literature constitutes about 10% of these tumors.[4,5,12] However, the percentages have not been fully demonstrated and the actual rate of malignant transformation could be lower or higher considering the circumstances of the individual studies.[4] Further, if LSMFT represents a variant of fibrous dysplasia, as according to the arguments by Heim-Hall & Williams,[1] then the risk of malignant transformation should be lower.

In summary, the lesion is considered by some researchers as a variant of fibrous dysplasia or as a non-specific end result of degenerative change, and other researchers see it as a definite clinicopathologic entity with its own characteristic radiographic features, a complex mixture of histologic elements and a potential

of malignant transformation at low (but not well-defined) rate. However, more large scale studies for comparing LSMFT with other benign bone lesions, including fibrous dysplasia, are mandatory to clarify the origin and behavior of this lesion.

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