Rare Massive Osteolipoma in the Upper Part of the Knee in a Young Adult

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abstract

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Lipoma is a common benign soft tissue tumor. This article describes a massive osteolipoma, an unusual lipoma that is fixed to the femoral periosteum. A 21-year-old man presented with a subcutaneous mass at the knee region, which had been present for more than 36 months, and a slight limitation of joint flexibility. On physical examination, a mass approximately 12×6×2 cm³ in dimension was palpable beneath the skin at the knee proximal medial area when the knee was in flexion. The mass was ovoid, hard, nontender, well demarcated, large, subcutaneous, and relatively fixed to the femur. Medical imaging examination showed that the femur had a well-demarcated mass with a basement. No prominent body weight loss was noted. The excisional mass of 16×12×10 cm³ was not well encapsulated by a thin, fibrous membrane and had an apparently osseous basal portion. Intraoperative rapid frozen section revealed that the tumor was derived from the adipose cells. The postoperative course was uneventful. The definitive pathologic diagnosis was intermuscular osteolipoma without evidence of malignancy. No recurrence was observed at 6-month follow-up.

Osteolipoma with independent bone and an osseous basal portion is rare, especially in young adults. Osteolipoma has the same prognosis as simple lipoma, and surgical excision is the recommended treatment. To the authors’ knowledge, such a massive osteolipoma has not been reported.

Figure: Intraoperative photograph showing the fringe of the tumor (green frame) and the border of the invasion portion (black frame) (A). Photograph of the excised tumor (12×6×2 cm³) (B).

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Lipoïme is a common benign soft tissue tumor composed of mature adipose cells without cellular atypia that predominantly occurs on subcutaneous regions of the upper back and neck, shoulder, and abdomen, especially in middle-aged adults. It presents as a soft or doughy asymptomatic, slow-growing, round or discoid mass. To the authors’ knowledge, this is the first report of an unusual osteolipoma relatively fixed to the periosteum of the distal femur.

**Case Report**

A 21-year-old man presented with a painless mass in the upper margin of the left knee. The patient had first noted the lesion approximately 3 years previously, but he had not brought it to the attention of a physician because it caused only slight discomfort when the knee was flexing.

The patient reported no history of trauma. Approximately 3 days previously, the discomfort became obvious, and knee flexibility was slightly limited. He had presented to several physicians, but a definite diagnosis had not been made. At presentation to the current authors, his pain was intermittent, related to physical activity, and did not radiate to other regions. On routine physical examination, he appeared healthy but was overweight (body mass index, 28.2 kg/m²). Gait pattern was normal. A giant mass was palpated when the left knee was in flexion, but it was not palpable when his knee was in extension. The mass was ovoid, hard, nontender, well demarcated, large, subcutaneous, and relatively fixed to the femur. No tenderness was present on his left lower leg. Neurovascular examination of the left lower extremity was within normal limits, and no lymphadenopathy existed. Laboratory findings were within normal limits. His medical history was unremarkable.

Computed tomography (CT) scan demonstrated an abnormally dense area in the anterior supracondylar femur with a connection to the femoral cortex (Figure 1). T2-weighted magnetic resonance imaging (MRI) of 3.0 T revealed a lobular contour with a heterogeneously increased signal intensity and fine decreased signal intensity septation (Figure 2A). Fat suppression short TI inversion recovery images showed geographically central increased signal intensity admixed with areas of low signal intensity (Figure 2B). T1-weighted MRI revealed a well-demarcated lobulated mass with heterogeneous signal intensity and focal areas of increased signal intensity, suggesting the presence of adipose tissue in the tumor (Figure 2C). The low signal intensity areas were interpreted as possibly representing ossification. The imaging findings were those of a heterogeneous soft tissue mass with calcification or ossification. The differential diagnoses included lipoma with calcification or ossification, ossifying fibromyxoid tumor, myositis ossificans, a cartilage tumor, and soft tissue osteosarcoma.

An intraoperative frozen section analysis identified that the mass was generated from adipocytes. The tumor was adherent to the periosteum as suggested by the CT images. The tumor was relatively avascular, and the intraoperative blood loss was approximately 100 mL. After proper hemostasis, the wound was closed with a drainage tube. The wound healed, and the patient returned to work in 2 weeks with no complications.
The oval-shaped excisional tumor was 16 cm at its greatest dimension. Grossly, the tumor was composed of yellow fatty tissue and surrounded by a thin fibrous capsule (Figure 3). Cut sections of the mass revealed mainly yellow soft tissue with numerous interlacing, thin, lamellar bony structures and surrounded with a thin fibrous capsule. Areas of bone formation were evident around the periphery and in the septa in the central aspect of the tumor. Microscopically, the tumor was composed of lobules of mature adipose tissue as seen in conventional lipoma (Figure 4). Many of these lobules contained nests of cells with round-oval, uniform, centrally placed nuclei and eosinophilic or multivacuolated cytoplasm. Some of the cells had vacuoles that indented the nucleus, indicative of lipoblastic differentiation. An amount of eosinophilic myxoid or hyalinized-appearing matrix was present between the cells. Many of the cells had well-defined cell borders. The matrix, vacuolated nature of the cytoplasm with a central nucleus and well-defined cell borders imparted a chondroid appearance to these areas. In regions with ossification, the bone was cortical-type bone composed of lamellar and woven bone, which was produced by osteoblastic cells on the outer surfaces of the rim and trabeculae of bone and not by tumor cells (metaplastic bone). The definitive pathologic diagnosis was intermuscular osteolipoma without evidence of malignancy. No recurrence was observed at 6-month follow-up.

**DISCUSSION**

Lipomas represent the largest group of mesenchymal tumors that occur in soft tissue, from superficial subcutaneous fat to as deep as the peristium. In the World Health Organization classification of human soft tissue and bone tumors, 14 types of benign tumors of adipose tissue exist, including lipoma, fibrolipoma, myxolipoma, angiolipoma, hibernoma, and chondroid lipoma, depending on the presence of variable amounts of other mesenchymal components that form an intrinsic part of the tumor, such as fibrous connective tissue in fibrolipoma or mucoid substances in myxoid lipoma. Cartilaginous or osseous metaplasia (eg, chondrolipoma or osteolipoma) is rarely described. Most lipomas are superficial (ie, subcutaneous) and present during middle age in the upper back, shoulder, abdomen, and extremities.

Lipomas containing osseous elements have been variously called ossifying lipoma and osteolipomas. Lipomas composed primarily of fat have been called ossifying lipoma, and those in which fat is not the primary component are called osteolipomas. In the current patient, the lesion was composed primarily of mature osseous elements and was, therefore, an osteolipoma. Eighteen cases of osteolipoma were reviewed by Kuyama in a period of 48 years.

To the current authors’ knowledge, the only previously reported case relating to the femur was an intramuscular osteolipoma without evidence of malignancy of the right medial proximal thigh in a 47-year-old man. However, in the current case, the largest nonpalpable osteolipoma was located in the distal femur in a young adult.

In the current case, the osseous element of the osteolipoma was composed of a periost, a cortex invasive portion, and an independent bone tissue portion. It is well known that longstanding tumors lying adjacent to a bone tend to cause bone erosion or a reaction. In the current case, the lipoma...
grew into the cortex; however, no evidence proved that the lipoma was sarcomatous degeneration. Although the bone reaction was not detected in the current case, early-stage bone reaction, such as bony hyperostosis, protuberance, erosion, and compressive changes, might be present.

Lipomas can undergo various changes, including ossification. Ossification does not represent a sinister change of the lipoma.\(^7\) The pathogenesis of osteolipoma remains unknown, although 2 main theories have been proposed. The first suggests that these tumors arise from multipotent mesenchymal cells;\(^6\) however, the favored hypothesis is secondary ossification from repetitive trauma.\(^8\)

Adipocytes and osteoblasts are derived from common mesenchymal stem cell precursors, and differentiation toward 1 cell lineage is usually at the expense of the other.\(^9\) Cytogenetic analysis revealed that the presence of the recurring t(3;12) (q27;q13-15) in osteolipoma and ordinary lipoma were the same.\(^10\) Although adipose-derived stem cells and lipoma-derived mesenchymal stem cells show different cell morphology and different biological properties,\(^11\) they both have the potential to differentiate into osteoblasts.\(^12\)

Adipocytes can promote osteoblastic proliferation and inhibit osteoclastic differentiation through adipocyte-secreted factors,\(^8,13\) therefore, the lipoma cells of osteolipoma may also have this feature. Osteoblastic activity has been proven to be influenced by local transforming growth factor-\(\beta1\).\(^14\) Transforming growth factor-\(\beta1\) can promote the proliferation of osteoblasts in low concentration and inhibit the proliferation of osteoblasts in high concentration.\(^15\) Among the lipoma tissue, local transforming growth factor-\(\beta1\) is in low expression.\(^16\) Cases in the literature have confirmed that the local transforming growth factor-\(\beta1\) expression macrophage-like cell infiltrates the vicinity of the ossification lesion.\(^17\) Taken together, osteoblastic activity is strengthened by the environment of lipoma tissue. This case is unique because the osteolipoma was relatively fixed to the periosteum of the distal femur. The reason that the osteolipoma grows adjacent to the bone has not been clarified. However, a possible reason is that repeated friction and trauma between the increased lipoma and the periosteum may cause locally enhanced activity of osteoblasts on the periosteum, which subsequently promote ossification.

The prognosis of lipoma with ossification is favorable, as is that of simple lipoma. The most appropriate treatment is surgical excision, and no recurrences have been reported. Although evidence is lacking in cytological atypia, the deep location of the lesions coupled with the presence of heterologous elements raised concern regarding atypical lipomatous tumor.\(^10\) In the current case, although the osteolipoma was fixed to the cortex of femur, no sarcomatous degeneration or other malignant changes existed.

**CONCLUSION**

Osteolipoma with independent bone and an osseous basal portion is rare. The pathogenesis of massive ossifying formation needs to be clarified.

**REFERENCES**