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Case Series

Intraosseous Liposarcoma. Case Report

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Abstract

Primary intraosseous liposarcoma is a rare and malignant type of tumor of the skeletal system, the first reports of primary bone liposarcoma were made by Ewing, Stewart and Barnard in the 1930s the first case was reported in 1931 by Stewart. Since 1980, only a small number of cases of primary liposarcoma affecting a long bone have been described in the English literature, 2 male patients aged 68 and 25 years with a diagnosis of round cell liposarcoma and primary pleomorphic liposarcoma of bone were presented staging, biopsy, and once the lesion was diagnosed, amputation of the affected limb was performed and follow-up by the multidisciplinary team led by oncology.

Keywords: Intraosseous, Liposarcoma, Skeletal

Introduction

Primary intraosseous liposarcoma is a rare and malignant type of tumor of the skeletal system [1-5], the first reports of primary bone liposarcoma were made by Ewing, Stewart and Barnard in the 1930s [6] the first case was reported in 1931 by Stewart [2], Catto and Stevens, reviewing the world literature in 1963, could only find 15 cases of primary bone liposarcoma; but most of them corresponded to pleomorphic sarcomas and only 1 of the one reported by Dawson in 1955 coincided with the diagnosis of primary bone liposarcoma [3], Since 1980, only a small number of cases of primary liposarcoma affecting a long bone have been described in the English literature [2]. According to the World Health Organization (WHO) classification in its updated edition in 2013, four defined subtypes are defined: atypical lipomatous tumor/well-differentiated liposarcoma, (ii) dedifferentiated liposarcoma, (iii) myxoid liposarcoma, (iv) pleomorphic liposarcoma [4].

Case 1

A 68-year-old male patient with a history of apparent health complained of pain and increased volume in the right leg for 3 months that did not relieve with the use of conventional analgesics, which made it difficult for him to walk, so he came to our clinic where it was decided to admit him for study and treatment

Computerized axial tomography

In the middle third of the right leg, an osteolytic image with low density was observed, with cortical rupture, and the presence in soft tissues of a well-defined ovoid image measuring between 7.7 x 1.2 cm.

Anatomical pathological findings

- **Macro:** Several irregular tissue fragments are received in Pathological Anatomy, the largest of 5 x 4 x 1cm, firm con-

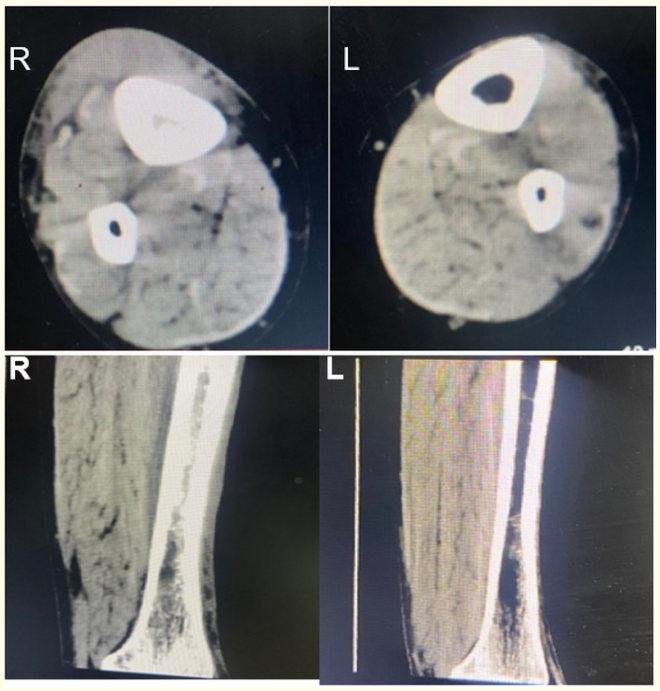

Figure 1

sistency to the touch. When cut, light brown surface, same consistency.

- **Micro:** In the microscopic study, the presence of hypercellular areas composed of adipocytic cells with differentiation into round cells, which show increased nuclear grade and elevated mitotic activista, immersed in a myxoid stroma, is observed.
- **Immunohistochemistry:** This cell population was found to be S100 positive. Which led us to confirm our diagnosis of Round Cell Liposarcoma.

Case 2

A 25-year-old male patient with a history of Gilbert's syndrome, chronic gastritis, hiatal hernia, who complained of pain and increased volume in the region of the middle third of the left thigh, which limited his gait, so he came to our clinic where it was decided to admit him for study and treatment, and during admission he presented a pathological fracture at the level of the left femur.

Computerized axial tomography

The tomographic study showed an osteolytic image at the level of the middle third of the left femur with loss of bone cortical continuity solution and infiltration of soft tissues, it was interpreted as a possible osteoblastic osteosarcoma.

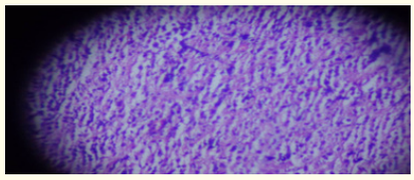

Figure 2: Round Cell Liposarcoma Panoramic View.

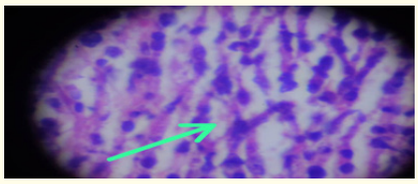

Figure 3: Adipocytic cell vacuolated cytoplasm.

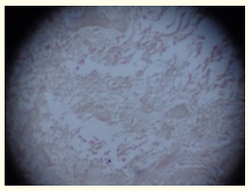

Figure 4: Primary pleomorphic liposarcoma of bone. Panoramic view.

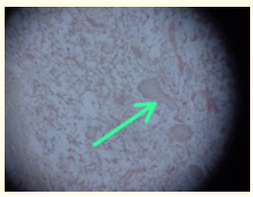

Figure 5: Bone trabecular immersed in pleomorphic liposarcoma.

Anatomical pathological findings

- **Macro:** Multiple grayish, irregular soft tissue fragments were received in Pathological Anatomy, which together make 3cc, coming from the spinal canal, according to the operative report, with some fragments of bone tissue that were sent to be decalcified.
- **Micro:** Microscopic study shows the presence of large lipoblasts with hyperchromatic nuclei and prominent nucleoli in a multivacuolated cytoplasm. In the company of bone trabeculae of normal histological appearance. These characteristics led us to the diagnosis of primary pleomorphic liposarcoma of bone.
- **Immunohistochemistry:** Lipoblasts were strongly positive for the S100 protein, which reinforced our diagnosis. HMB45, CD68 and CD45 negative.

Discussion

Liposarcomas are classically soft tissue tumors that most commonly affect the upper and lower limbs, In most reported cases, bone liposarcoma has been observed to develop in the major long tubular bones, including the femur, tibia, and humerus, with most cases located in the lower extremity [1-3,6]. Well-differentiated liposarcoma is the most common subtype, accounting for 40-45%. It is notable for its tendency to local relapse, onset in adulthood [4,7] the primary bone location of liposarcomas remains rare; in fact, liposarcomas account for only less than 0.1% of primary bone cancers [1,4,5,7] their diagnosis is based on clinical and radiological elements and is confirmed by pathological anatomy, In addition, immunohistochemistry and molecular biology techniques [1,2] as with other malignant bone tumors, patients have a history of pain, swelling, decreased range of motion, and an expansive and osteolytic lesion as demonstrated by radiographs. Clinically, it is possible to confuse a primary intraosseous liposarcoma with osteosarcoma, Ewing sarcoma, plasmacytoma, or lymphoma. Comments [2].

In the case of round cell liposarcoma, it is necessary to take into account other entities that may present pseudolipoblasts, such as myxofibrosarcoma, low-grade fibromyxoid sarcoma and myofibroblast neoplasms, among others [8]. Myxofibrosarcoma is a multinodular growth tumor with incomplete fibrous septa. Presence of alternating hypo- and hypercellular areas in an abundant myxoid stroma. Characteristics not observed in our case in question. Low-grade fibromyxoid sarcoma features a spindle-shaped cell population in a densely collagen stroma with abrupt transition to myxoid areas. Therefore, it is also ruled out in our histological diagnosis [9].

All high-grade pleomorphic sarcomas should be considered in the differential diagnosis of pleomorphic liposarcoma. The diagnosis of undifferentiated pleomorphic sarcoma was excluded precisely because of the presence of lipoblasts that are not present in this entity. Clear cell sarcoma was ruled out because the HMB45 stain was negative, since the cells of this sarcoma are S100 and HMB45 positive. Other tumors to consider in the differential diagnosis are those containing osteoclast-type giant cells such as Soft Tissue Giant Cell Tumor, Giant Cell-Rich Sarcoma, and Undifferentiated Giant Cell Sarcoma. In general, these tumors have osteolytic growth that invades neighboring structures and destroys the bone. Their cells express CD68 and CD45, antibodies that were negative in our case study [10].

There is no defined standard treatment for primary intraosseous liposarcoma due to the low incidence of these, surgical resection remains the primary treatment method to reduce recurrence [2]. The outcome after surgery alone for primary intraosseous liposarcoma is relatively poor, and there are few data on the use of adjuvant or neoadjuvant chemotherapy or radiotherapy [3], Metastatic disease is the main cause of death in patients with primary intraosseous liposarcoma, and the most affected target organ is the lung [2,6] indicating that this type of tumor is associated with a poor prognosis [2] and once the lesion was diagnosed, amputation of the affected limb was performed and follow-up by the multidisciplinary team led by oncology.

Conclusion

Primary intraosseous liposarcoma is a rare tumor that most frequently affects adults, mainly located in the extremities. It must be proven that the tumor has originated mainly within the bone, that is, that it is not a metastatic deposit and that it is not of periosteal origin, affecting the cortex and medulla secondarily. it is necessary to establish a clinical diagnosis, Radiológica and histological studies for adequate management, focusing on the principles of tumor lesion management, always seeking to determine if there is the presence of secondary lesions that may modify the course to be followed.

Conflict of Interest

None to declare.

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