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Adamantinoma of Tibia: A Case Report and Literature Review

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Abstract **Case Report**

Adamantinoma of long bones is a slow-growing, low-grade primary malignant bone tumor. It is a rare entity and accounts for less than 1% of all primary bone tumors, and in most cases, it occurs in the mid-axis of the tibia of adolescents and young adults. In this report, we describe the case of a 55-year-old woman with a diagnosis of adamantinoma of the left tibia.

Keywords: Adamantinoma, primary bone tumor, adolescents, diagnosis.

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INTRODUCTION

Adamantinoma is a very rare primary bone tumor located almost exclusively on the anterior surface of the tibia. Adamantinomas occur most frequently in adolescents and young adults with a slightly greater prevalence rate in men than women. The clinic is reduced to local pain, tenderness and/or swelling. The objective of this work is to report the case of an adamantimoma of the tibia in a young woman and to recall the radiological semiology X-ray standards CT and MRI).

CASE PRESENTATION

A 55-year-old woman with no particular pathological history who presented with leg pain evolving for 4 days. Standard X-ray showed an unifocal osteolytic, eccentric and expansive lesion, affecting the cortex and marrow of the distal third of the diaphysis of the left tibia with cortical disruption. We note that the lesion is not associated with peripheral osteosclerosis or periosteal reaction.

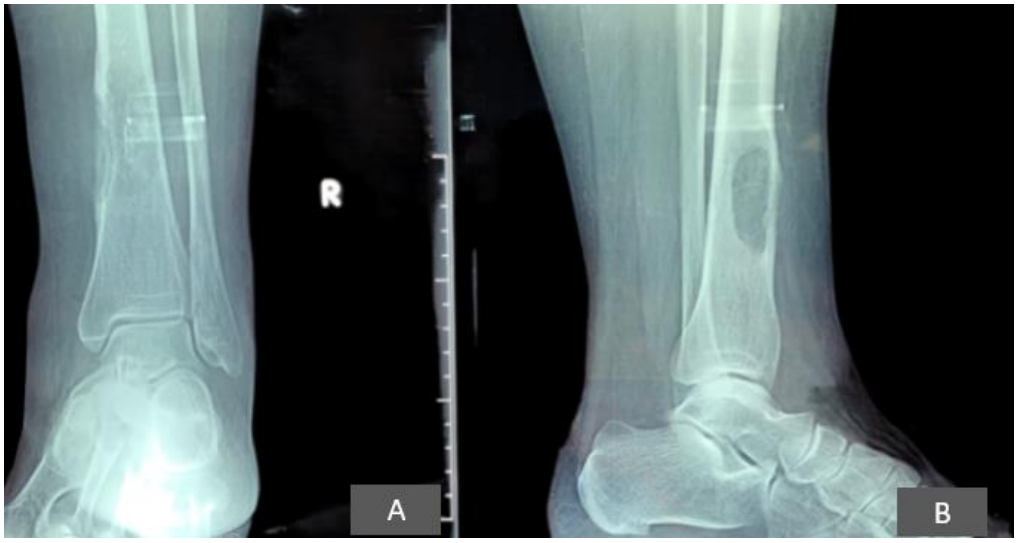

Figure 1: An AP (A) and lateral (B) view of the left ankle: an unifocal osteolytic, eccentric and expansive lesion, affecting the anterior cortex and marrow of the distal third diaphysis of the left tibia, associated to a cortical disruption and without a peripheral osteosclerosis or periosteal reaction

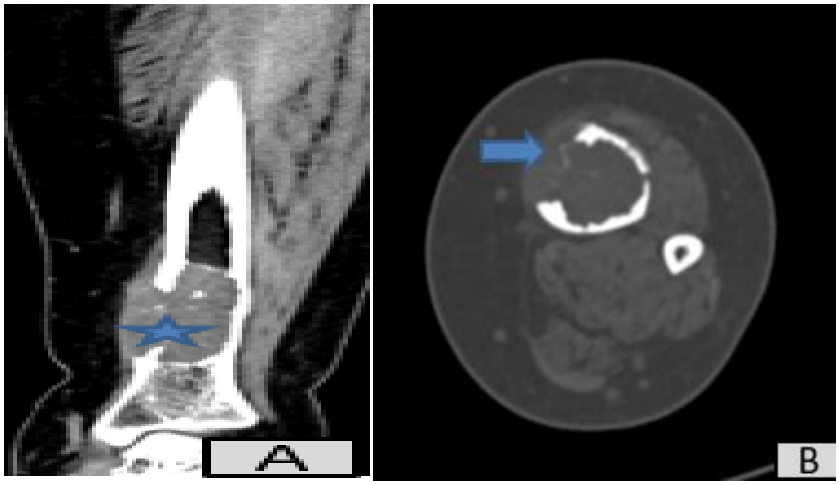

Figure 2: CT scan of the left leg centered on the lesion: axial in bone window (A), reconstruction sagittal in soft tissue window in post contrast product (B): unifocal lesion located at the distal third of the diaphysis of tibia with cortical disruption (arrow), involvement of the medullary canal and surrounding soft tissue extension (asterix) with enhancement after injection of the product contrast

The MRI showed a lesion in low signal T1, heterogeneous hyigh signal T2 and STIR, intensely enhanced after injection of PDC with involvement of the medullary canal and surrounding soft tissue extension. The biopsy confirmed the diagnosis of an adamantimoma. The patient subsequently had a CT of the chest, abdomen, and pelvis performed, which revealed no distant metastasis. The patient subsequently underwent en bloc resection of the tumor with healthy margins.

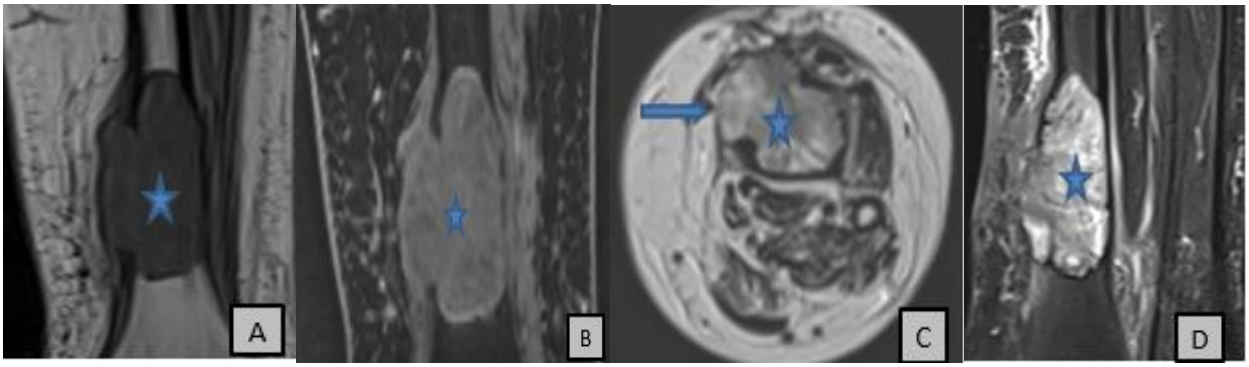

Figure 3: MRI of the left leg, A: sagittal T1, B: sagittal T1 FAT SAT post contrast product injecting, C: axial T2, and D: sagittal STIR

A well-limited lesion on the anterior side of the tibia cortex (asterix) with low signal T1, high signal T2 and STIR and significantly enhancing after intravenous injection of gadolinium. This lesion involves almost all of the medullary cavity and surrounding soft tissues (arrow).

DISCUSSION

Adamantinoma is a very rare primary bone tumor located and it represents 0.1 to 0.5% of all primary bone tumors [1]. The anterior cortex of the tibial diaphysis is involved in the vast majority of cases [2]. The preferred location of adamantinoma is the tibia: the diaphysis is affected in 60.4% of cases, the proximal metaphysis in 13.9% of cases and the distal metaphysis in 24.8% of cases [1, 6]. Other long bones can be affected much more rarely (femur, fibula, tibia and ipsilateral fibula) [1-6].

Involvement of a synchronous lesion in the ipsilateral fibula has been reported in approximately 10% of cases [4]. Adamantinomas occur most frequently in adolescents and young adults with a mean age of 25–35 years [6]. Previous series have identified a slight male predominance [8, 12]. Given the indolent nature of this malignancy, it typically has a long, progressive clinical course characterized by swelling, pain, and deformity prior to diagnosis [7].

From an anatomopathologic point of view, adamantinoma is a tumor characterized by the presence, in variable proportions, of epithelial cells within an osteofibrous tissue. There are two histological subtypes: adamantinoma of the osteofibrodysplastic type (predominance of the osteofibrous contingent) and classic adamantinoma (predominance of the epithelial contingent) [6]. The osteofibrous dysplasia, osteofibrodysplastic-type adamantinoma, and classic

adamantinoma are usually considered to be separate (but possibly related) entities with decreasing prognosis. To adapt the therapeutic management, it is essential to differentiate osteofibrous dysplasia from adamantinoma. Indeed, osteofibrous dysplasia is a benign bone tumor, more frequently encountered in children. Conversely, adamantinoma, more frequently encountered in adults, presents marked local aggressiveness; it can recur locally and metastasize distantly (lung, bone) [4].

Adamantinoma characteristically affects the anterior cortex of the tibial shaft. On radiography, the incipient adamantinoma appears as a lesion eccentric cortical, unifocal or more rarely bifocal (ipsilateral tibia and fibula), small size, without associated periosteal reaction [1, 2]. At a more advanced stage, the tumor takes on a "soap bubble" appearance [1, 2]. Peripheral osteosclerosis is common and indicative of slow growth. The cortical disruption and periosteal reaction may be associated 7. The lesion is longitudinally oriented with an average length in one series of 11.0 cm [8]. MRI is the examination to be performed in first intention to determine the extension of the tumor in the bone marrow and surrounding structures. Two types of presentation have been described: a lobulated, solitary or sometimes bifocal lesion, or multiple small nodules grouped in one or more foci [2]. The signal of the lesion and its enhancement are not specific but the topography (anterior tibial cortex) is more evocative. It is impossible to differentiate on imaging an osteofibrodysplastic adamantinoma from a classic adamantinoma [2] but some criteria could point to osteofibrous dysplasia or adamantinoma [2]. A lesion extended in height (more than 10 cm on average), "moth-eaten" pattern of bone disruption, and invasion of the medullary cavity would be more in favor of adamantinoma [2, 3, 5].

En bloc resection of the tumor with healthy margins followed by reconstructive surgery is considered the most appropriate treatment for adamantinoma. None radiotherapy or chemotherapy has not proven its efficacy. With appropriate surgical treatment, the prognosis of adamantinoma is excellent (survival rate of 87.2% at 10 years [6]). However, prolonged clinical and radiological monitoring is necessary because of the risk of local recurrence or metastasis which can occur many years after the initial treatment [4]. This risk is directly related to the quality of the surgical resection and therefore requires early diagnosis and precise locoregional extension assessment. Imaging is therefore essential for optimal management of these patients.

CONCLUSION

Adamantinoma is a rare, low-grade malignant tumor of the bone and typically occurs in the diaphysis of long bones, particularly in the tibia and associated with an indolent course due to its slow growth. En bloc resection with wide opeerative margins followed by limb reconstruction is the preferred modality for managing this malignant tumor and producing good functional outcomes along with high rates of limb preservation and overall survival.

Conflicts of Interest: The authors declare that they have no conflicts of interest.

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