

Adamantinoma of the Tibia: A Case Report

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What to Learn from this Article?

Slow growing by locally aggressive tumor of tibia. differential diagnosis and management

Abstract

Introduction: Adamantinoma is a bone tumor which is commonly showed in mandible. Its occurrence in long bones constitutes a rare affection and a difficult histological diagnosis This rare malignant tumor of mesenchymal and epithelial origin was discovered in the tibia of a male patient.

Case Report: The authors report a case of adamantinoma of the tibia in a 36- years old. He presented with slowly progressing leg swelling that evolved over 2 years without significant clinical manifestation. Complementary examination and investigation did not help in differential diagnosis and confirmation was only possible after histological examination. There was increase in size of lesion with increased pain and patient presented with pathological fracture. Local extension of the tumor indicated an amputation of leg. The histological exam of the amputation specimen confirmed the results of the initial biopsy by showing cells tumorales in favour of an adamantinoma. After a good medium-term follow up, the patient died secondary to lung metastases. The authors discuss their method of diagnosis and therapeutics in front of such tumor.

Conclusion: Adamantinoma is characterized by a slow clinical development marked by the prevalence of lung metastases and local recurrences. After the diagnosis, there must be a complete and sufficient surgery to avoid a local recurrence or metastasis.

Keywords: Adamantinoma, Tibia, Tumor.

Author's Photo Gallery



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Introduction

Adamantinoma is an intermediate malignancy tumor of fibroblast origin. Its preferential location in the mandible makes its discovery at the level of long bones a rare affection and a difficult histological diagnosis. It typically occurs during second decade and histogenesis remains controversial[1] Since its first description by Fischer[2] in 1913. The seat of the tumor at the level of the limbs varied, but remains predominantly in the tibia (80%). It represents less than 0.4% of all malignant bone tumors[3]. Adamantinoma is characterized by a slow clinical development marked by the prevalence of lung metastases and local recurrences. We report a case of adamantinoma of tibia in a young adult with special reference to difficulty in diagnosis and final outcome after radical surgical approach.

Case Report

Thirty six year old unemployed male patient presented with swelling of right leg that was gradually increasing since last two years. there was no history of significant trauma and the general status was good. Clinical examination revealed a firm steady swelling, tender on palpation, sitting in the antero lateral face of the leg. It measured 15 cm in its long axis. There was no collateral venous circulation and skin appeared normal. The x-ray of the leg showed a diaphyseal cortical erosion image associated with an aspect of soft parts densification (Fig.1). Computer tomography examination revealed a tumor process with involvement of the medullary cavity and extending to the anterior muscle (Fig.2). Furthermore, abdominal ultrasound and chest

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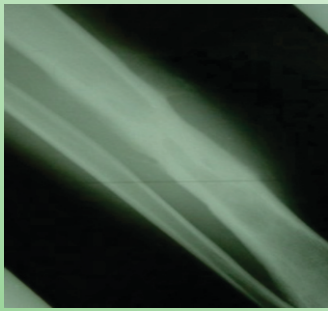


Figure1: Bone scan of bilateral tibiae demonstrates focal increased radiotracer uptake along the medial aspect of the left tibia at the concavity of the curve.

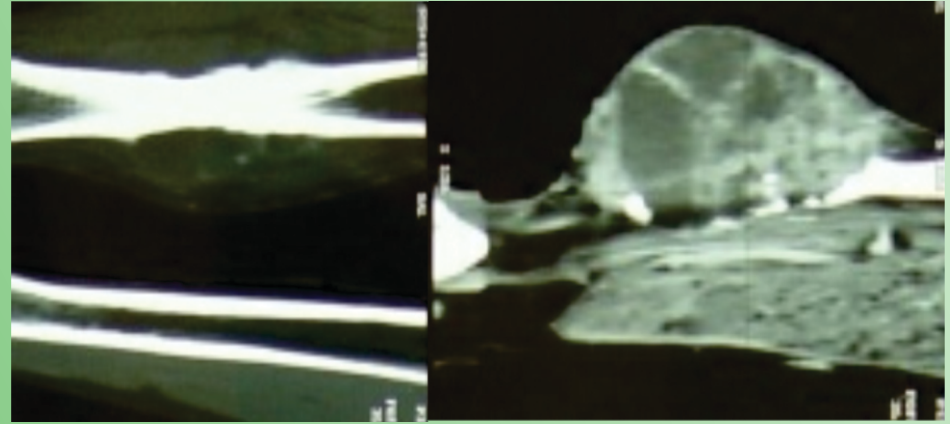


Figure 2: CT Scan of the leg: confirmation of cortical lysis with tumor infiltration of the soft tissues.

radiography revealed no abnormal findings. The histological examination after biopsy revealed a bone adamantinoma. The patient was reviewed two months later and had an overall swelling distorting the whole leg associated with severe pain.

Significant lysis and a pathological fracture of the tibia were discovered on x-ray (Fig. 3). Radical treatment was indicated and amputation in the upper third of the leg was done. Histological analysis of the lesion confirmed the biopsy result by showing a tumor proliferation made of lobules with palisade edges and large fibrous focus rich in vessels. The immediate postoperative course was uneventful and the patient was regularly followed. The evolution was marked by the installation of a massive pleurisy and worsening of the general status leading to death in 17 months.

Discussion

Adamantinoma is a neoplasm described the first time in 1921 by Fischer. This designation by the author was based on histological similarities of the tumor with that of the mandible usually called ameloblastoma. It is a tumor in the young patients between ages 20 to 40. Apart from the tibia, the other long bones are rarely affected but the axial skeleton is very rarely involved [4]. This pathology has a non-specific clinical manifestation, pain is the only main symptom. In our case the pain had been evolving for eight months. The radiograph appearance is that of a diaphyseal osteolysis well limited by a thin edge of osteosclerosis and osteocondensation focus [4]. This aspect of classic tumorous bone lysis was observed in our case at the anterior part of the tibial shaft. According to Desai et al [5], the tibial site and the intra cortical development are two indicative arguments for an adamantinoma. CT scan and magnetic resonance imaging are not specific. Histology on the biopsy

or excision sample provides definitive diagnosis based on the presence of epithelial cells arranged in islands in a fibrous and fibroblast proliferation medullary tissue. Immunohistochemistry confirms the dominance of epithelial cells that result in a progressive abandonment of the notion that it could have a synovial origin.

Fischer [2] invoked an in-utero inclusion at the origin of the presence of epithelial cells in the bone while Ryrie [6] advanced a traumatic inclusion by epithelial migration into the underlying bone. In our case immunohistochemistry was not performed. Braud and al [7] showed that there are many histological features and defined classically four types, which are sometimes associated: basaloid, fusiform, malpighian or tubular. Our case is the basaloid type because of the palisade position of basal cells. Dorfman and Czerniak [8] divided adamantinoma into two groups: a classical form, characterized by an intra periosteum radiographic destruction. On the histological plan, this form is dominated by basophil and tubular tumor cells. And a differentiated form characterized by the presence of an osteofibrodysplasia (OFD) with small epithelial isolated cells and an intra cortical and multifocal location on the radiological plan. Our case described is the classical form. Desai et al [5] insisted on five major injuries that can be confused with an adamantinoma: metastasis, epithelial tumor, a vascular tumor, a fibrodysplasia and an osteofibro-dysplasia. All these injuries are to be eliminated on clinical, radiological and histological arguments. Thus, the tibia is no preferential seat for bone metastases, and metastases occur rather in elderly subjects.

Myoepithelial tumours predominate in soft tissues even if some adamantinomas were exceptionally found in these [9]; in this case the immunohistochemical study allows diagnosis [5]. This diagnosis will also help eliminate vascular tumors like hemangioendothelioma that affects all ages with

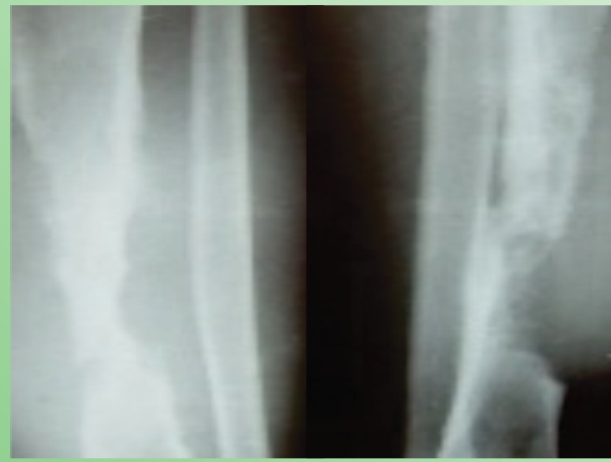


Figure 3: Radiograph of the leg, third month after biopsy: Note the worsening cortical lysis with a pathological fracture of the tibia.

a histological aspect close to an adamantinoma. Bone dysplasia can affect young patients is marked by recurrence and metastasis, Cytokeratin research through immunohistochemistry must be the rule.

The therapeutic protocol in the management of adamantinomas is not formally established. Braud et al [7] identified three methods (curettage, resection and amputation) used without asserting the superiority of one or another. Curettage is the source of almost constant recurrences; wide resection and amputation allow to avoid them. The recurrence rate following an incomplete excision may reach 30% [4]. Hoshi et al [10] advocate a personalized attitude based on the histological nature. If the first biopsy identifies a classic adamantinoma, wide outright resection is performed. Curettage is performed in case of an OFD differentiated form, the recurrence potential of which does not virtually exist. The evolution of the tumor is assessed based on the diagnosis delay and the type of treatment. Long-term surveillance is required. Classically, lung metastases are about 15-31%. Ganglion metastasis and local recurrences represent about 30% whereas mortality is 13-25% after 10 years backward step [7]. In our case death occurred in a context of recurrent pleurisy confirming the lethality of lung metastases of adamantinoma.

Conclusion

Adamantinoma is a quiescent malignant evolution bone

tumor and deadly by its complications. The histological, microscopic and immunohistochemical analysis provides confirmation diagnosis and eliminates associated forms. After the diagnosis, there must be a complete and sufficient surgery to avoid a local recurrence or metastasis.

Clinical Message

Because of the slow evolution, adamantinoma is usually found late. Consequently, in front of any painful swelling of the tibia, the surgeon must investigate thoroughly with biopsy for the histological diagnosis or refer the patient to a specialized center for a suitable treatment. This attitude would avoid the death due to pulmonary metastases as reported in our study.

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