Case Report/Olgu Sunumu

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A rare case of adamantinoma in tibia and radiological features

Nadir bir tibia'da adamantinoma olgusu, radyolojik özellikleri

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ABSTRACT

Adamantinoma is a rare low grade primary malignant bone tumor usually found in the diaphysis of the tibia. The tumor is locally aggressive and can metastasize. As it is resistant to chemotherapy and radiotherapy, the main treatment is total excision of the lesion with wide surgical margins. Although it is difficult to distinguish from other bone lesions (such as osteofibrous dysplasia) radiologically, there are some helpful features. With this presentation of a typical case, we hope to draw the attention of radiologists to this lesion.

Keywords: Adamantinoma, tibia, bone neoplasms, radiology, MRI, osteofibrous dysplasia

ÖZ

Adamantinoma, nadir görülen düşük dereceli primer malign kemik tümörüdür ve genellikle tibia diyafizinde bulunur. Tümör lokal olarak agresiftir ve metastaz yapabilir. Kemoterapi ve radyoterapiye dirençli olduğu için asıl tedavi lezyonun geniş cerrahi sınırlarla total eksizyonudur. Radyolojik olarak diğer kemik lezyonlarından (osteofibröz displazi gibi) ayırt etmek zor olsa da bazı ayırt edici özellikleri vardır. Tipik bir vakanın bu sunumu ile radyologların dikkatini bu lezyona çekmeyi umuyoruz.

Anahtar Kelimeler: Adamantinoma, tibia, kemik neoplazmları, radyoloji, MRG, osteofibröz displazi

INTRODUCTION

Adamantinoma is a rare low grade primary malignant bone tumor. The Greek word 'adamantinos'means 'very hard'. It was first observed in ulna in 1900 by C. Maier (1,2). It was named as "primary adamantinoma of the tibia" by Fischer in 1913 because of its histological resemblance to the jaw ameloblastoma (3,6). The tumor is usually located in the diaphysis of tibia especially in 2'nd and 3'rd decades, mildly more frequently in males. Patients present to the physician with a complaint of mild pain that has been going on for years and a swelling that gradually becomes apparent. Sometimes the complaint may be a pathological fracture (4). In this article, we aimed to share the magnetic resonance imaging (MRI) and radiographic findings of this rare entity.

CASE

A 28 year old male patient admitted orthopedics and traumatology department with pain on left leg. There was a painful contour deformity on left tibia in physical examination. The patient had no other known diseases. Laboratory findings were within normal limits. Direct radiography showed a translucent lesion in the left tibia diaphyseal region, which was located in the anterior cortex with sclerotic margin (**Figure 1**). MRI revealed a mass lesion in the anterior diaphyseal section of the left tibia, that caused destruction in cortical bone. The lesion had an axial diameter of 28x17 mm with craniocaudal extension of 70 mm, extending into the tibialis anterior muscle. Significant contrast enhancement was observed in the lesion (**Figure 2**). There was also an increase in

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signal intensity from proton density to T2 sequences, and with cystic area showing a hyperintense fluid-like signal (**Figure 3,4**). No signal suppression in the lesion was observed on the fat saturation sequences. It was recommended to excise the lesion in terms of possible malignant lesion due to the exophytic extension of the bone, causing obvious destruction in the bone. A biopsy was performed and histology revealed features typical of epithelial and osteofibrous components of adamantinoma. Metastasis were not observed in other imaging techniques. The lesion was resected with a margin of normal bone by the surgeon.

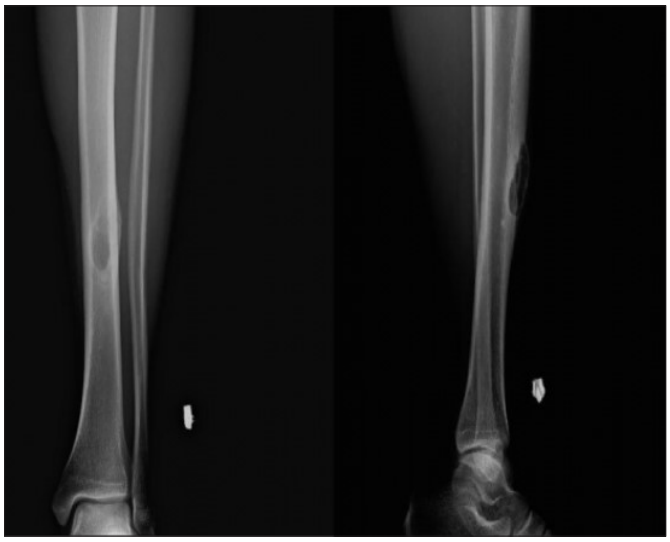

Figure 1. Anteroposterior and lateral radiographs of the left tibia and fibula demonstrate an osteolytic lesion is primarily cortical based and has a well-defined sclerotic margin on its inner aspect. Note the narrow zone of transition, which correlates with the indolent nature of the lesion.

DISCUSSION

Adamantinoma is locally aggressive tumors with slow progression and metastasis potential (2,4,7,8). Since it is not sensitive to chemotherapy and radiotherapy, the treatment is the carefully removing of the lesion with a wide surgical margin (9). Inadequate surgical procedures significantly increase the risk of local recurrence and lung metastasis (5,10-13). Therefore, preoperative imaging and staging are important for appropriate treatment. Evaluation with only radiographic findings may make the actual size of the tumor smaller than it is. MRI is the preferred imaging technique for staging musculoskeletal tumors due to its high soft tissue contrast resolution and its ability to perform multiplanar imaging (9).

Some features of the tumor such as its classic location and characteristic appearance are findings that help radiological diagnosis. In general, as in our patient, multiloculated radiolucent lesions are seen in the tibia in the form of central or eccentric localized, sclerotic limited appearance and osteolytic lesions with slightly large, sharp or insufficiently circumscribed. The lesion may be surrounded by a prominent sclerotic border that indicates slow growth (5,12). Although diaphysis is the most common location, rarely metaphyseal extension or isolated metaphyseal involvement can be seen. When metaphyseal involvement occurs, it becomes difficult to make a diagnosis since other tumors with metaphyseal location are also included in the differential diagnosis (2). Tumors involving the anterior tibial cortex, peripheral sclerosis and tumors with well-defined lesions including septation, may have multifocal involvement in the same bone. These multifocal radiolucencies surrounded by

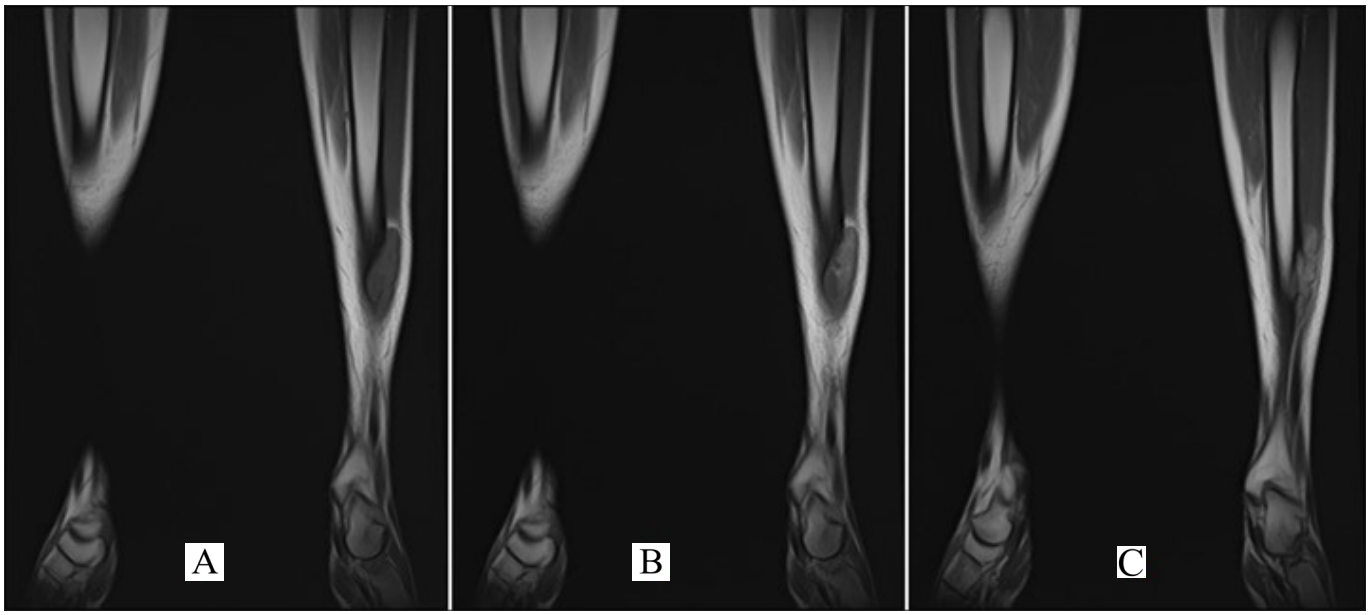

Figure 2. (a) Coronal T1-weighted image reveals anterior of tibia presence of tumor with intermediate signal intensity showing soft-tissue extension. (b-c) Coronal T1-weighted image obtained after administration of contrast medium shows enhancement, cortical destruction and soft-tissue extension.

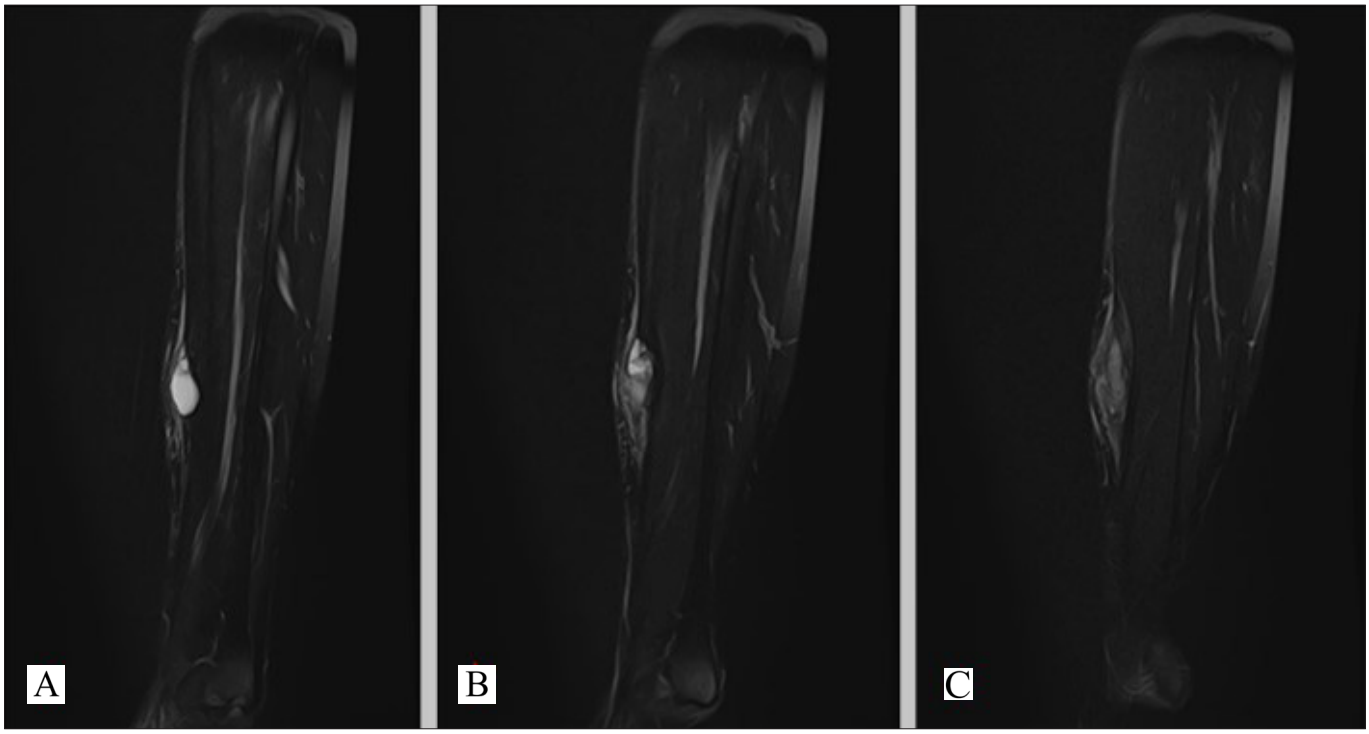

Figure 3. Cystic areas showing a hyperintense fluid-like signal in the sagittal T2-weighted images

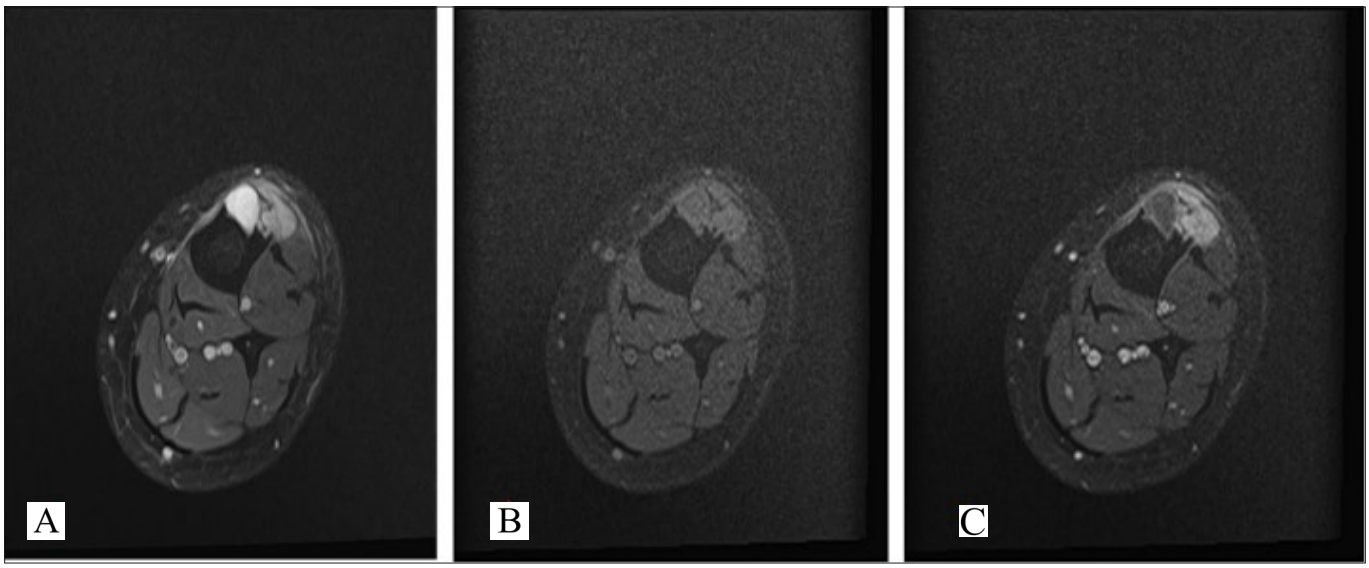

Figure 4. (a) Axial proton density imaging shows a lesion with a cystic component. (b) On axial T1-weighted fat saturated section, an intermediate signal intensity lesion with soft tissue extension destructing the anterior bone cortex was seen. (c) On axial T1-weighted images with fat-selective presaturation after contrast medium administration, lesions show intense enhancement and are well demarcated. The cystic compartment does not show enhancement.

ring-shaped densities form the characteristic "soap bubble" appearance (5). The lesion is usually located intracortically, but in some cases, there may be cortex destruction and involvement in extra cortical soft tissues. Different degrees of periost reaction can be seen (5,11).

Radiologically, the differential diagnosis of adamantinoma should include osteofibrous dysplasia and fibrous dysplasia. In direct radiographs, "soap bubble appearance" can be observed both in osteofibrous dysplasia and differentiated adamantinoma (6).

Although it is difficult to distinguish between osteofibrous dysplasia and fibrous dysplasia from adamantinoma, there are some helpful features. Osteofibrous dysplasia involve the bone cortex without medullary involvement and has a ground-glass appearance (5). Fibrous dysplasia presents with a homogeneous low signal intensity in T1- and T2-weighted images because of its histological composition of mainly fibrous material (6). On the other hand, the appearance of single or multiple nodular lesions with frontal cortex involvement located in the diaphysis and extending into the bone

marrow approaches the diagnosis of adamantinoma (5). Adamantinoma shows a more heterogeneous pattern with cystic areas (high signal intensity in T2), sometimes with hemorrhagic content (high signal intensity in T1), surrounded by a fibrous mass (low signal in all sequences) (6). Other lesions that should be kept in mind in the differential diagnosis including angiosarcoma, hemangioendothelioma, aneurrysmal bone cyst, giant cell tumor, chondrosarcoma, nonossifying fibromas, epithelial metastasis (2).

Although computed tomography (CT) can show bone cortex involvement and soft tissue extension, it cannot clearly show the intraosseous extension of the lesion. CT is often used routinely to screen for lung metastases.

MRI plays a critical role in demonstrating extension of intramedullary and soft tissue, distant cortical foci and tumor-free margins. It is seen in low signal intensity in T1-weighted images and high signal intensity in T2-weighted images. Because these appearances are also typical of most tumors, these findings are nonspecific (2). Nuclear imaging methods can be used also in the evaluation of adamantinoma (14).

CONCLUSION

The main treatment in adamantinoma is total excision of the lesion with wide surgical margins. Evaluation of the true size of the lesion and the soft tissue component in the preoperative period is important in the success of the surgical treatment. As a result, the possibility of local recurrence-residual tumor tissue and distant metastasis is reduced in the postoperative period. MRI is the most important imaging technique in cases like our case due to its high soft tissue contrast resolution and its success in showing the true limits of the tumor to guide surgical treatment correctly.

ETHICAL DECLARATIONS

Informed Consent: Written informed consent was obtained from all participants who participated in this study.

Referee Evaluation Process: Externally peer-reviewed.

Conflict of Interest Statement: The authors have no conflicts of interest to declare.

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Author Contributions: All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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