CASE REPORT

Osteosarcoma Arising from a Solitary Osteochondroma of the Tibia

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Abstract

We present a case of osteosarcoma arising from an osteochondroma of the right tibia in a 71 year old man. The radiographic studies were suggestive of a malignant lesion. Histologic examination showed a conventional osteosarcoma that eroded the cartilagenous cap. The patient received postoperative chemotherapy with no evidence of metastasis until three years following the operation. The occurrence of osteosarcoma out of osteochondroma is an extremely rare event and very few cases have been reported.

Keywords: Osteochondroma; Malignant transformation; Osteosarcoma

Introduction

Solitary osteochondroma is the most common benign tumor of the skeleton.[1,2] Jaffe estimated that although as many as 10 to 20% of the patients with multiple hereditary osteochondromatosis experienced malignant transformation, about 1% of the solitary ones undergo malignant changes to chondrosarcoma.[3] In a solitary osteochondroma, the occurrence of a sarcoma other than chondrosarcoma is exceedingly rare and only sporadically reported. It is usually in the form of the osteosarcoma.[4,5] We present a case of osteosarcoma that arose from a slowly growing solitary osteochondroma.

Case Report

A 71 year old man was referred to our institution with a history of a large mass on the lateral aspect of proximal part of the right leg for 29 years. Radiographs taken 29 years ago showed a typical osteochondroma of the right tibia. This had recently increased in size from three months prior to admission. On physical examination, there was a large, firm, bony fixed mass of about 15-Cm in its largest dimension on the proximal lateral aspect of the right leg. The overlying skin was thin but moved freely over the tumor. It was not painful and no neurologic or circulatory deficits were evident.

The plain X-ray (AP & LAT) of the knee and proximal leg revealed a large heterogeneous expansile dense bony mass arising from the proximal tibia, with no significant soft tissue component, but with mild irregularity of the surface (Figure 1). Axial CT scan showed a large expansile bone tumor with the foci of the anterior cortical erosion and mild soft tissue swelling over its anterior (Figure 2).

A biopsy was taken. Microscopic studies showed high-grade sarcoma with malignant osteoid formation. Upper knee amputation was done. An irregularly shaped 15X10X5 Cm bony mass partially covered by a bluish cartilagenous cap was found over the upper portion of the tibia. On cut surface, it was mainly made of dense bone of cancellous appearance. The cartilagenous cap was uneven with maximum thickness of 1 mm.

Microscopically, the lateral and most of the upper surface was covered by cortical bone and it was only focally covered by a cartilagenous or fibrous cap. The underlying spongy bone was filled by conventional osteosarcoma. The tumor focally interrupted the surface cortical bone. The tumor cells were spindle shaped with hyperchromatic, pleomorphic nuclei and frequent

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Received: March 20, 2008 Accepted: July 15, 2008

mitotic figures. They produced a fair amount of osteoid but no cartilagenous matrix (Figure 3 and 4). Some osteoclastic giant cells were also present. The resection margins of the bone and soft tissue were free of tumor. Chemotherapy was started for the patient and after three years there was no evidence of metastasis.

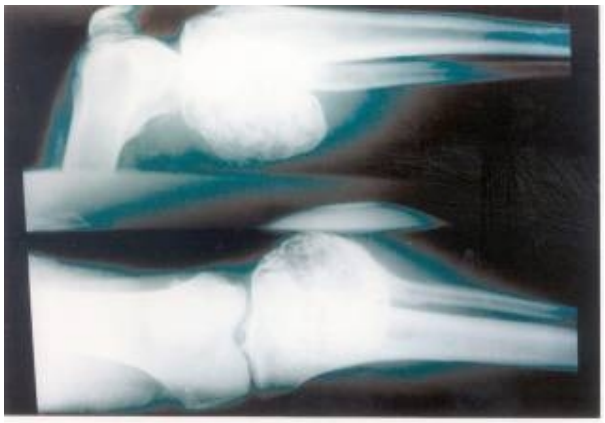

Fig 1: Plain X–ray (AP & LAT) of Knee and proximal leg bony large mass arising from proximal part of tibia.

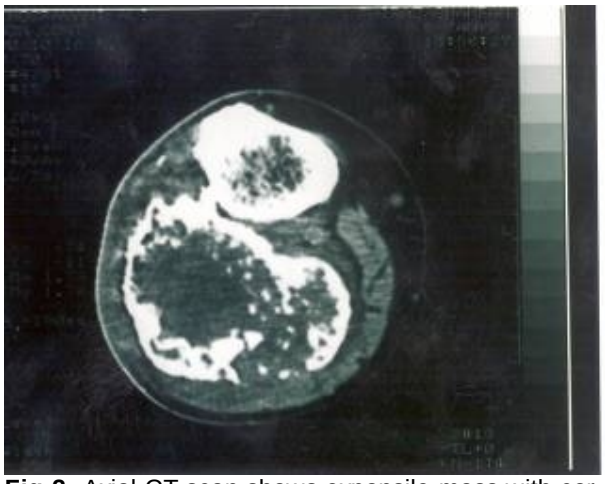

Fig 2: Axial CT scan shows expansile mass with cortical erosion.

Discussion

Malignant transformation of a solitary osteochondroma is a rare event, occurring in about 1 to 2 % of cases.[1] The malignant transformation is almost exclusively in the form of chondrosarcoma.[1,2,6,7] Reports of other sarcoma arising from an osteochondroma are rare.[8] Osteosarcoma is an unusual complication of an osteochondroma. Very few cases have been reported.[9]

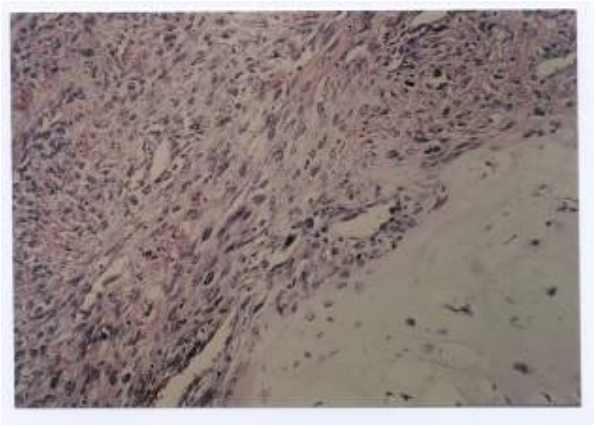

Fig 3: Microscopic examination shows tumor cells with pleomorphism, high N/C ratio, hyperchromatic nuclei adjacent to cartilaginous cap (H&E, X1000).

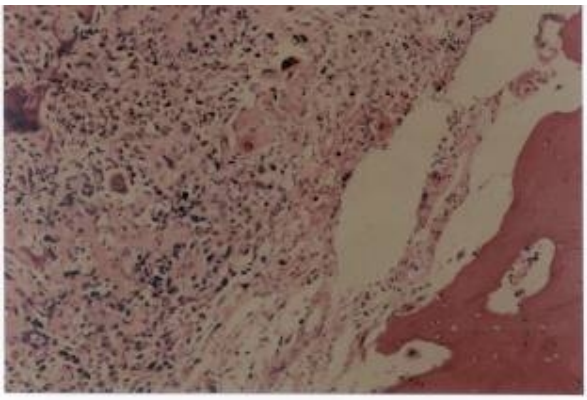

Fig 4: Malignant osteoid produced by tumor cells (H&E, X1000)

In review of the literature till 2007, nine cases of osteosarcoma arising in the solitary chondrosarcoma were reported. Dahlin reported two such cases.[9] In his patients, osteosarcoma arose from the osteochondroma of the femur. The complete files of the Mayo Clinic contain only three such cases, in two of which the patients had a solitary osteochondroma and the third occurred in a patient with multiple exostoses.[10] The rest of the published cases were single case reports.[4,11-13,15] Other reports of malignant transformation to osteosarcoma occurring in multiple osteochondromatosis are also present which are not related to our discussion.[5,14]

In our case, we were not able to obtain radiographic studies of the lesion done 29 years ago. However, the radiographs of the current lesion and histologic data were those of osteochondroma with malignant trans-

formation. The recent accelerated growth of the tumor is also in favor of malignant transformation. With regard to prognosis, the follow up in most cases as well as ours was relatively short. However, it is suggested that osteogenic sarcoma arising from an osteochondroma have a better prognosis than the usual kind.[5,9]

Conflict of interest: None declared.

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