

Case Report

Metallic implant-related osteosarcoma*,**

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ABSTRACT

Orthopedic metallic prostheses are commonly used in the current practice of orthopedic surgery. Although, biomaterials used in these implants are generally considered to be biologically inert, there have been consequences of foreign body reactions and potential carcinogenesis. Majority of implant-related malignancies are high grade, and develop in bone or soft tissue around the implant site. No clear association has been identified between the biomaterial implanted and the type of sarcoma. We report the case of a 36-year-old male who underwent intramedullary nailing of femur for mid-shaft fracture secondary to trauma and presented with osteogenic sarcoma of femur 14 months later.

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Introduction

The use of metallic prosthesis for repair of skeletal deformities, fracture fixation, and replacement of poorly functioning or painful joints is common in current practice of orthopedic surgery. The implanted materials are usually biologically inert but some components may have a carcinogenic potential [1]. Often cementless implants are chosen as they have a porous surface, allowing a greater interface between the parent bone and the synthetic device for better stabilization. However, this has the disadvantage of exposing larger amounts of the recipient tissue to a foreign material, thereby enhancing biological incompatibility and the likelihood of carcinogenesis [2].

Most implant-related malignancies developing in bone or soft tissue adjacent to the implant site are high grade. The most reported is pleomorphic sarcoma (malignant fibrous histiocytoma) followed by osteosarcoma. Others include angiosarcoma, chondrosarcoma, Ewing sarcoma, epithelioid sarcoma, epithelioid hemangioendothelioma, malignant peripheral nerve-sheath tumor, fibrosarcoma, synovial sarcoma, and

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lymphoma [1]. No correlation has been established between the biomaterial implanted and the histologic type of sarcoma [3].

We report the case of a 36-year-old male who underwent intramedullary nailing of femur for mid-shaft fracture secondary to trauma in an otherwise normal bone and presented with osteogenic sarcoma of femur 14 months later.

Case report

A 36-year-old male presented to the Emergency department following a road traffic accident. His initial X-ray showed left femoral mid-shaft fracture (Fig. 1A) in an otherwise normal bone. The patient underwent surgery, and an intramedullary nail was placed in the fractured femur (Fig. 1B). Early follow up radiograph was unremarkable. He presented 14 months later with left thigh swelling for 2 months. The radiograph showed interval formation of new bone around the fracture site along with elevation of the periosteum and soft tissue swelling (Fig. 2). Magnetic resonance imaging (MRI) of thigh was done which revealed abnormal signals almost involving the entire femur and a large enhancing soft tissue component (Fig. 3) which showed increased tracer uptake on bone scan (Fig. 4). Biopsy was performed which confirmed osteosarcoma (Fig. 5). Whole body Computed tomography (CT) was negative for metastatic disease. The patient received 4 cycles of neo-adjuvant chemotherapy comprising methotrexate, doxorubicin, and cisplatin (MAP protocol) followed by resection of left femur with wide margins (Fig. 6A) and total hip and knee replacement (Fig. 6B). Postoperatively the patient was vitally and hemodynamically stable. He was gradually mobilized and



Fig. 2 – Frontal radiograph 14 months later shows intramedullary nail in left femur. New bone formation in sunburst pattern around the fracture site (blue arrows) and along proximal left femoral shaft (white arrow) with triangular elevation of the periosteum (black arrow).



Fig. 1 – (A) Frontal radiograph shows transverse fracture mid shaft of left femur with mild displacement. (B) An intramedullary nail has been placed showing good alignment of fracture fragments.



Fig. 3 – (A) Coronal postcontrast MRI image shows artifact from intramedullary nail (white arrow), abnormal enhancement along the shaft of femur (yellow arrows). (B) Sagittal postcontrast MRI image shows enhancing soft tissue component (white arrows).

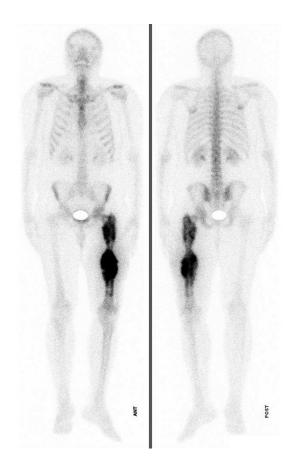


Fig. 4 – Bone Scan shows intense tracer uptake with bony expansion over proximal and middle one-third of left femoral shaft.

discharged a week later in stable condition. One year later, patient is ambulatory and has no distant metastasis on recent CT scan.

Discussion

Malignancy arising secondary to an implanted biomaterial, although rare, is a grave complication. Prostheses implicated in human carcinogenesis are those containing metals such as chromium, nickel, manganese, iron, stainless steel, and silicon. Another plausible cause for development of malignancy following implant placement is implant induced osteonecrosis [4,5]. The reported mean age of implant-related sarcomas is 50 years, but it has been seen in both extremes of age, without any gender preference. The sarcoma can manifest from anywhere between 6 months to 30 years after implant placement, with a mean time of 9 years [1,3]. Our patient presented approximately 14 months post implant placement. Also, femur is the most reported site as this the most common bone undergoing implant treatment.

The annual incidence of primary osteosarcoma, which is the most common malignant tumor of the bone, is 3.4 per 1 million population [6,7]. Secondary osteosarcoma arises in an already abnormal/diseased bone [8] or rarely in those with a metallic implant as seen in our case. The presentation is nonspecific, common symptoms being pain and swelling which may be mistaken for postoperative or inflammatory changes, leading to a diagnostic delay. Our patient remained well for almost a year following intramedullary nail placement and then developed new onset swelling for which he re-sought medical advice and was found to have osteosarcoma. The clinical course is usually aggressive, and significant number of pa-

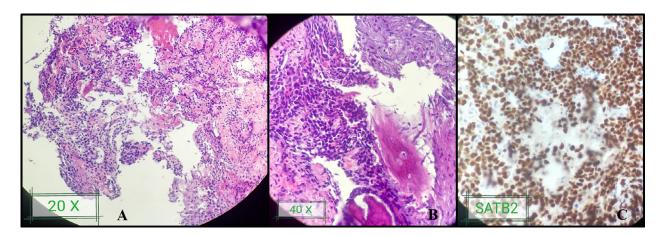


Fig. 5 – (A) Tumor cells composed of round to spindle cells arranged in nest (H&E stain; $20 \times \text{magnification}$). (B) Pleomorphic hyperchromatic nuclei with eosinophilic cytoplasm and focal osteoid matrix (H&E stain; $40 \times \text{magnification}$). (C) Immunohistochemical stain SATB2 positive in tumor cells.



Fig. 6 – (A) Radiograph of the resected left femur. (B) Frontal radiograph showing total left femur and left knee replacement.

tients develop metastasis. The risk of mortality is quite high even with surgery, chemotherapy, and radiotherapy [3]. Fortunately, our patient did not have any metastatic disease and is doing well.

Conclusion

Although implant-related sarcoma is extremely rare, it is imperative to consider this possibility when any new symptoms develop in patients with metallic orthopedic implant placement.

Patient consent

Written informed consent for medical information and images to be published in this case report was provided by the patient.

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