



# Local recurrence of parosteal osteosarcoma adjacent to a prosthesis after 20 years: A case report

TW Lau, JWK Wong, DKH Yip, EP Chien

Department of Orthopaedics and Traumatology, The University of Hong Kong, Queen Mary Hospital, Hong Kong

TWH Shek

Department of Pathology, The University of Hong Kong, Queen Mary Hospital, Hong Kong

LLS Wong

Department of Radiology, The University of Hong Kong, Queen Mary Hospital, Hong Kong

---

## ABSTRACT

We describe the treatment of an uncommonly late presentation of a recurrent parosteal osteosarcoma of the distal femur. The osteosarcoma had originally been detected 20 years earlier, and had been treated with wide excision and mega-prosthesis to reconstruct the femur. The tumour recurred in close proximity to the femur prosthesis and encased half the femoral stem. Because there was a large piece of metal at the site of recurrence, which might have interfered with computed tomography and magnetic resonance imaging, ultrasonography was used to locate the lesion. The tumour was successfully treated with wide local re-excision. This case emphasises the importance

of the long-term follow-up of patients with parosteal osteosarcoma.

*Key words:* osteosarcoma; prosthesis; recurrence

---

## INTRODUCTION

Parosteal osteosarcoma—a well-differentiated malignant bone-forming tumour arising on the surface of bone—is an uncommon tumour.<sup>1,2</sup> The most frequent site of occurrence is the posterior aspect of the distal femur. Being a generally low-grade malignant tumour, parosteal osteosarcoma carries a better prognosis than conventional intramedullary

osteosarcoma.<sup>3</sup> After inadequate excision, however, parosteal osteosarcoma tends to recur locally.<sup>1,2,4</sup> In this article, we report a case of recurrent parosteal osteosarcoma in the distal femur 20 years after its initial excision, and its successful treatment with wide local re-excision.

## CASE REPORT

A 35-year-old woman presented to the Queen Mary Hospital in November 1996 with mild pain in her left thigh. At the age of 15 years, the patient had experienced pain and swelling in the left thigh; a bone lesion had subsequently been found in the posterior aspect of the distal left femur, which had been diagnosed as fibrous dysplasia according to excisional biopsy results. At age 17 years, she had undergone another excision of the lesion because of local recurrence. At the age of 18 years while being a student in the United Kingdom, she was seen at a national orthopaedic tumour centre because the swelling in the thigh had recurred; X-ray and histological examinations had led to the diagnosis of parosteal osteosarcoma of the distal femur. One year later, the parosteal osteosarcoma had been treated with wide excision and the femur had been reconstructed with a prosthesis. The procedure had yielded excellent results.

At presentation, the patient reported a 2-year history of intermittent left-knee pain, swelling, and instability. The pain and swelling was brought on by prolonged walking, and could be relieved by taking rest and non-steroidal anti-inflammatory drugs. On examination, she had a lateral longitudinal surgical scar over the left knee. There was moderate effusion, 10° recurvatum, flexion to 120°, and mild laxity during the valgus and varus stress test. Radiographs of the left femur and knee showed the cemented prosthesis without evidence of loosening, as well as a faint calcified area close to the femoral component at the postero-medial aspect. A bone and thallium scintiscan showed a moderate hotspot medial to the prosthesis in the lower portion of the left femur, without increased thallium uptake. This finding was suggestive of mild reactive changes in the site of the prosthesis—namely, ectopic calcification—rather than recurrence of osteosarcoma. The calcified area was monitored with serial radiography. Meanwhile, her knee pain worsened, which was attributable to the wearing of the polyethylene axial bushings of the prosthesis. Two years later (at age 37 years, or 18 years after initial definitive surgery), the bushings were changed and were found grossly worn down. She



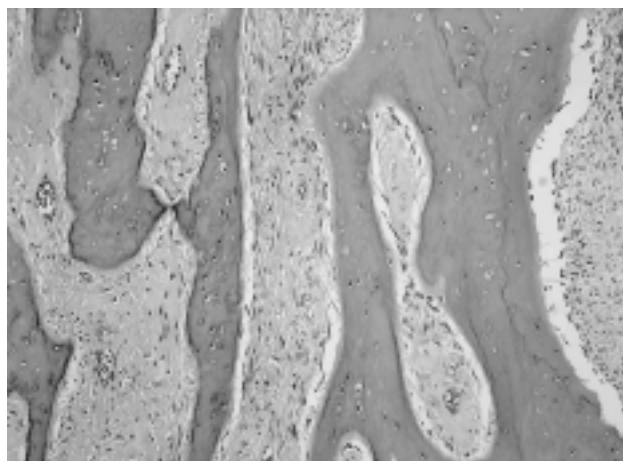
**Figure 1** Anteroposterior radiograph of the femur and knee showing the calcified soft tissue mass adjacent to femoral prosthesis.

recovered from this operation uneventfully and the knee pain, swelling, and instability improved significantly.

However, subsequent serial radiography at age 39 years showed that the calcified area adjacent to the femoral prosthesis had increased (Fig. 1). The area



**Figure 2** The tumour recurred in close proximity to the femur prosthesis, encasing half the femoral stem.



**Figure 3** The recurrent parosteal osteosarcoma was composed of low-grade fibroblastic cells between parallel spicules of bone (H&E, x50).

became palpable at the medial thigh and was slightly tender. Because the large metal prosthesis would have interfered with computed tomography (CT) and magnetic resonance imaging (MRI), ultrasonography was used to locate the calcified lesion, and showed that it was well-defined and deep in the vastus medialis, abutting the femoral prosthesis. In addition, a mixed cystic and solid lesion was visible proximal to but separate from the calcified lesion, which also involved the vastus medialis. A trucut biopsy of the lesion was performed under ultrasound guidance. Histological examination showed fibrogranulation tissue with silica-induced foreign-body reaction, but no malignancy was found. Incisional biopsy on the calcified lesion subsequently showed that the parosteal osteosarcoma had recurred. Bone scanning and CT of the thorax and abdomen did not show evidence of metastasis.

Wide excision of the recurrent osteosarcoma was thus performed, during which the lesion was found to encase half the femoral prosthesis (Fig. 2), suggesting slow growth of the tumour in the soft tissue. Histological examination confirmed parosteal osteosarcoma recurrence, showing areas of both grade 1 and grade 2 tumours (Fig. 3). There was no evidence of dedifferentiation. The more proximal mixed solid cystic lesion that had been detected on the ultrasound scan was confirmed histologically to be a synovial-lined cystic structure with features of metallosis and chronic inflammation. The patient regained muscle strength and range of motion of the knee 6 months after surgery. Three years later, she remained asymptomatic and had excellent knee function.

## DISCUSSION

This case report illustrates very well the point that parosteal osteosarcoma without features of dedifferentiation indicates a good prognosis. Despite its propensity to recur locally after inadequate excision, parosteal osteosarcoma rarely results in metastasis.<sup>1-4</sup> The first 2 excisions had obviously been inadequate, because fibrous dysplasia had been the initial diagnosis. The tumour inevitably recurred shortly after the operations. When the correct diagnosis was finally made, wide excision and careful reconstruction with prosthesis led to an excellent outcome.

The mega-prosthesis indeed served this patient very well: for more than 20 years, the patient had undergone just one change of the polyethylene bushings and had been asymptomatic for most of the time. We believe this case report may be the same patient as that in case 14 in the series described by Kavanagh et al.<sup>2</sup> and thus describes the long-term follow-up of that case. The long-term outcome (>20 years) of the mega-prosthesis is not well documented in the literature, and it would be worth reporting because many patients with this tumour undergo the surgery at a young age.

Defining the anatomical location of the tumour recurrence is sometimes difficult in patients who have undergone mega-prosthesis, because the prosthesis may distort the images of the conventional CT and MRI scans. An ultrasound investigation may be an alternative approach when these imaging studies cannot be performed. Ultrasonography may require a skilful radiologist with relevant expertise.

The recurrence of parosteal osteosarcoma 20 years after initial definitive surgery with wide excision and prosthetic replacement is very rare. Okada et al.<sup>4</sup> reported that the longest time from definitive surgery to local recurrence in their series of 226 patients was 15 years. Campanacci et al.<sup>1</sup> reported the longest time was 13 to 14 years in an analysis of 41 cases. Furthermore, Temple et al.<sup>5</sup> reported a mean of local recurrence at 7.6 years from surgery in a series of 38 patients. The time to recurrence may reflect the adequacy of the surgery and the length of

follow-up. Therefore, vigilance is needed in looking for recurrence when physicians follow up patients with low-grade malignant tumours.

#### **ACKNOWLEDGEMENTS**

We thank Mr SR Cannon and Dr Jean Pringle of Royal National Orthopaedic Hospital, Brockley Hill, Stanmore, England, for providing invaluable information of the medical history on this patient.

#### **REFERENCES**

1. Campanacci M, Picci P, Gherlinzoni F, Guerra A, Bertoni F, Neff JR. Parosteal osteosarcoma. *J Bone Joint Surg Br* 1984;66:313–21.
2. Kavanagh TG, Cannon SR, Pringle J, Stoker DJ, Kemp HB. Parosteal osteosarcoma. Treatment by wide resection and prosthetic replacement. *J Bone Joint Surg Br* 1990;72:959–65.
3. Unni KK, Dahlin DC, Beabout JW, Ivins JC. Parosteal osteogenic sarcoma. *Cancer* 1976;37:2466–75.
4. Okada K, Frassica FJ, Sim FH, Beabout JW, Bond JR, Unni KK. Parosteal osteosarcoma. A clinicopathological study. *J Bone Joint Surg Am* 1994;76:366–78.
5. Temple HT, Scully SP, O'Keefe RJ, Katapurum S, Mankin HJ. Clinical outcome of 38 patients with juxtacortical osteosarcoma. *Clin Orthop* 2000;373:208–17.