CASE REPORT

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Reactivation of tumor after 19 years? A case of local recurrence in clear cell chondrosarcoma

Accepted: 28 February 2000

Abstract Local recurrence of a clear cell chondrosarcoma of bone is reported arising 19 years after resection of the original tumour

Résumé Un cas de recidive local d'un chondrosarcome a cellules claires est rapporté 19 ans apres resection de la tumeur d'origine et reconstruction par prothèse massive.

Introduction

A 43-year-old man presented in 1977 complaining of aching in the right thigh. Radiographs showed a lytic lesion involving the upper femur and epiphysis. (Fig. 1). Biopsy suggested this was a low-grade chondrosarcoma. He underwent resection and insertion of a custom-made endoprosthesis. He subsequently returned to near-normal activities but was unable to play any sports.

In 1996, 19 years after the original operation he complained of aching in the thigh and radiographs suggested both components were loose. He underwent revision of both components and histological examination of curettings taken from the acetabulum showed polyethylene wear debris. He returned to normal function but 6 months later he developed groin pain.

Radiographs now showed a destructive lesion above the acetabulum involving the ilium and computerized tomography (CT) scan confirmed extensive destruction involving the entire ilium. Biopsy showed this to be a clear cell chondrosarcoma; identical to the tumor resected 20 years previously (Figs. 2, 3). Further staging studies showed no signs of tumor elsewhere in the body and he underwent complete resection of the ilium (Fig. 4) and reconstruction of the hemipelvis with a temporary combination of pins and cement attached to the side of the

Fig. 1 The initial radiograph in 1977 showing an epiphyseal tumor

sacrum. Two years later he remains disease free but with modest function of the affected limb. He has returned to part-time work.

Retrospective review of the 1996 radiographs taken at the time of the revision showed no abnormality within the ilium.

Discussion

Clear cell chondrosarcomas are rare, representing 1.5%–5% of all chondrosarcomas [2,4]. Only 100 or so have been described in the literature worldwide. They typically affect the epiphyseal area of a long bone and

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Fig. 2a Photomicrographs of the tumor resected in 1977. Area of clear cell chondrosarcoma showing large, non-cohesive tumor cells with no associated cartilage matrix around a reactive bone trabecula. **b.** Photomicrographs of the tumor resected in 1977. Area showing similar tumor cells situated in lacunae of abundant, focally mineralized, cartilage matrix

Fig. 3 Photomicrographs of the tumor resected in 1997. Area of tumor lacking cartilaginous matrix showing identical features to those seen in Fig. 2. Photomicrographs of the tumor resected in 1997. Area showing similar features to those seen in Fig. 2b. There is abundant tumor produced cartilage matrix and endochondral ossification

this diagnosis must be entertained in any adult with a tumor at this location. They can be difficult to diagnose histologically and can be confused with other tumors such as aneurysmal bone cyst, giant cell tumor and chondroblastoma [2]. Unni et al. first described the tumor in 1976 [5] and its relative novelty as an entity precluded the correct diagnosis being made at the time of initial resection in 1977.

Treatment of these tumors is by wide local excision, which is usually curative. Inadequate excision leads to a high risk of local recurrence and metastatic disease. Local recurrences have been reported in this condition as arising up to 12 years following the original diagnosis and in one case metastatic disease to other bones has been reported 23 years after initial diagnosis [1]. The re-

Fig. 4 The transected specimen of ilium in 1997, showing recurrent tumor arising from the iliac blade directly above the cemented acetabular cup

currence of tumor in this case is most unusual. It is quite possible that there was a “skip” lesion in the ilium, which was undetected initially and lay dormant for 20 years. It was not apparent radiologically at the time of revision, which was due to straightforward loosening – a well-recognized risk in proximal femoral replacements [3]. It would appear that the revision itself “awoke the sleeping beauty” and caused it to become active. It is possible that alterations in blood supply combined with increased growth factors stimulated the tumor to become active – a mechanism that may possibly be of relevance for local recurrence in a multiplicity of tumors. This case report has highlighted the need for continued awareness of possible tumor recurrence in low-grade tumors, especially after surgical intervention for whatever cause.

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