On-line Case Report

Local recurrence of osteosarcoma after 17 years

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ABSTRACT

The recurrence of osteosarcoma is rare. Local recurrence occurs in 4–10% of patients following effective treatment. So far, recurrences as late as 15 years have been reported in the literature. We report a unique case of local recurrence of intramedullary osteosarcoma 17 years from initial diagnosis and treatment. Regular long-term follow-up of patients with this diagnosis is crucial to ensure early detection and treatment of potential recurrences.

Keywords: Osteosarcoma – Recurrence – Long-term follow-up

Survival of patients with high-grade osteosarcoma has significantly improved with combined multi-agent chemotherapy and aggressive local surgical control. However, despite modern therapy, approximately one-third recur and these represent a greater surgical and oncological challenge. We report a unique case of local recurrence of osteosarcoma 17 years from initial diagnosis and treatment, highlighting the importance of regular long-term follow-up of patients with this diagnosis.

Case report

A 41-year-old woman presented with a painful right hip and was shown on biopsy to have an intramedullary osteosarcoma with both chondroblastic and osteoblastic differentiation of the right ilium in November 1989 (Fig. 1). There were no metastases or skip lesions.

She received one cycle of neo-adjuvant combination chemotherapy (platinum, ifosfamide and adriamycin [PIA]) prior to a right hemipelvectomy in April 1990 (Fig. 2). This operation was complicated by postoperative internal iliac vein thrombosis and pulmonary embolism, which required anticoagulation and insertion of an inferior vena caval filter.

Figure 1 Plain anteroposterior radiograph demonstrating osteosarcoma of right ilium.

At 6 weeks post-excision, she underwent a hemipelvic replacement with cemented Stanmore total hip arthroplasty (Fig. 3). She received a further five cycles of PIA chemotherapy. She developed a postoperative infection resulting in the formation of a discharging sinus. Despite multiple washouts and a debridement with

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Figure 2 Postoperative radiograph demonstrating hemipelvectomy.

Figure 3 Postoperative radiograph demonstrating hemipelvic and proximal femoral replacement.

external oblique rotational flap coverage, the sinus continued to discharge and the femoral and pelvic prostheses were removed in March 1994. This was complicated by an iatrogenic femoral fracture on prosthesis extraction.

She was able to mobilise with the use of two crutches and functioned extremely well. She was not keen for re-insertion of a prosthesis and remained on yearly follow-up until 2000 (11 years post diagnosis).

In June 2007, she presented to her general practitioner with dull right iliac fossa pain, radiating down her right leg. She was referred back to our service and examination revealed a mass in the right iliac fossa, confirmed on CT scan (Fig. 4). This was biopsied and histology demonstrated locally recurrent osteosarcoma. Staging investigations revealed no metastatic disease but did find a contralateral renal cell carcinoma.

She had excision of the recurrent osteosarcoma in September 2007 and has returned to her previous level of function. Histology from this resection demonstrated incomplete excision with residual tumour within iliacus. She went on to have a left laparoscopic nephrectomy. Postoperative abdominal CT scan demonstrated a new lesion within the right psoas muscle. She, therefore, had re-excision and is currently undergoing radiotherapy.

Discussion

The recurrence of osteosarcoma is rare. Local recurrence occurs in 4–10% of patients following effective treatment. The large Cooperative Osteosarcoma Study Group (COSS study), which ran between 1980 and 1998, found that 23 (1.4%) of 1702 osteosarcoma patients relapsed after 5 disease-free years. Of these patients, the longest disease-free period was 14 years.¹

Figure 4 CT scan demonstrating local recurrence of osteosarcoma.

This is the first occasion that local recurrence of intramedullary osteosarcoma has been described in the literature as much as 17 years from initial diagnosis and treatment. Whilst large-scale studies have reported recurrences as late as 15 years,^{2,3} only Lau *et al.*⁴ have reported an isolated case of recurrence 20 years following initial presentation. This, however, was a parosteal osteosarcoma which is recognised to carry a more favourable prognosis.

The prognosis of such late recurrence is uncertain. Mankin *et al.*³ studied survivorship data for 638 osteosarcoma patients. Of these patients, 68% survived the disease at an average follow-up time of 5.9 years. However, when the disease recurred, only 30% of patients survived. Work by Strauss *et al.*² highlighted several factors that influence the prognosis of recurrent disease. These factors include the disease-free time period, site of recurrence, histological response to treatment and the ability to attain complete surgical excision.

Notably, it seems that a prolonged disease-free period is associated with improved outcome. Patients with a disease-free interval of longer than 2 years have a significantly better post-relapse survival rate than those with a shorter interval (40% versus 7%).² However, Strauss *et al.*² also suggest that patients with very late relapse may fare worse, although such cases are so rare that any statistical analysis becomes difficult.

This case serves as a useful clinical reminder. Whilst 17 years is longer than most documented cases, patients do have relapses after 5 years. Vigilance must be encouraged for both patients with a history of osteosarcoma, and medical professionals from all disciplines dealing with these patients. High clinical suspicion must be maintained for all osteosarcoma patients presenting with unexplained symptoms. This case report may also serve to fuel consideration of the duration of current osteosarcoma follow-up protocols.

References

1. Bielack SS, Kempf-Bielack B, Delling G, Exner GU, Flege S, Helmke K *et al.* Prognostic factors in high-grade osteosarcoma of the extremities or trunk: An analysis of 1,702 patients treated on neoadjuvant cooperative osteosarcoma study group protocols. *J Clin Oncol* 2002; **20**: 776–90.
2. Strauss SJ, McTiernan A, Whelan JS. Late relapse of osteosarcoma: implications for follow-up and screening. *Pediatr Blood Cancer* 2004; **43**: 692–7.
3. Mankin HJ, Hornicek FJ, Rosenberg AE, Harmon DC, Gebhardt MC. Survival data for 648 patients with osteosarcoma treated at one institution. *Clin Orthop* 2004; **429**: 286–91.
4. Lau TW, Wong JW, Yip DK, Chien EP, Shek TW, Wong LL. Local recurrence of parosteal osteosarcoma adjacent to a prosthesis after 20 years: a case report. *J Orthop Surg (Hong Kong)* 2004; **12**: 263–6.