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CASE REPORT

Postradiation soft tissue sarcoma of the shoulder : A case report

Philippe DEBEER, Bart Van de MEULEBROUCKE, Jos STUYCK, Raf SCIOT, Ignace SAMSON

From the University Hospital Pellenberg, Pellenberg, Belgium,
the OLV Hospital Aalst, Belgium,
and the University Hospital Gasthuisberg, Leuven, Belgium

We present a case of a postradiation soft tissue sarcoma of the shoulder in a patient with a hemiarthroplasty of the shoulder. Initially the patient was treated for an infection of the hemiarthroplasty but subsequent removal of the loose prosthesis and biopsy revealed the presence of a malignant tumour.

Keywords : postradiation sarcoma : shoulder : tumour ; hemiarthroplasty ; infection.

CASE REPORT

A 62-year-old woman was referred to our institution five months after implantation of a right CTA humeral prosthesis (DePuy). The previous medical history included an ovariectomy in 1975, a radical mastectomy of the right breast in 1984 followed by radiotherapy and a radical hysterectomy in 1998. She was also treated for type 2 diabetes and hypertension. Eight years previously, she fell with her bike and sustained a fracture of the greater tuberosity of her right humerus that was treated with open reduction and fixation with a cerclage wire (fig 1a). Because of persisting pain a CTA humeral prosthesis (DePuy International Ltd, Leeds, UK) was implanted. Initially there was a good evolution after the implantation of this prosthesis. A few weeks after the operation, however, the patient complained of increasing pain, swelling of the anterior aspect of the shoulder and limited range of motion. Aspiration of the joint was performed but cultures remained sterile at that stage. After the aspiration the swelling progressively increased and the patient was referred to our institution. At that time she complained of pain and limited function of the shoulder. The pain was localised over the lateral aspect of the deltoid and the anterior shoulder region, and extended as far as the interscapular region. On examination there was no active motion in the right shoulder. Passive abduction and flexion were very painful and limited to 30°. Passive external rotation was -20°. There was a localised subcutaneous swelling at the anterior aspect of the

■ Philippe Debeer, MD, PhD, Consultant Orthopaedic Surgeon.
■ Jos Stuyck, MD, Consultant Orthopaedic Surgeon.
■ Ignace Samson, MD, Consultant Orthopaedic Surgeon.
Department of Musculoskeletal Science, Division of Orthopaedics, University Hospital Pellenberg, Pellenberg, Belgium.
■ Bart Van de Meulebroucke, MD, Consultant Orthopaedic Surgeon.
Department of Orthopaedics, OLV Hospital Aalst, Aalst, Belgium.
■ Raf Sciot, MD, PhD, Consultant Pathologist.
Department of Pathology, University Hospital Gasthuisberg, Leuven, Belgium.
Correspondence : Philippe Debeer, MD, PhD, Department of Orthopaedics, University Hospital Pellenberg, Weligerveld 1, B-3212 Pellenberg, Belgium.
E-mail : philippe.debeer@uz.kuleuven.ac.be.

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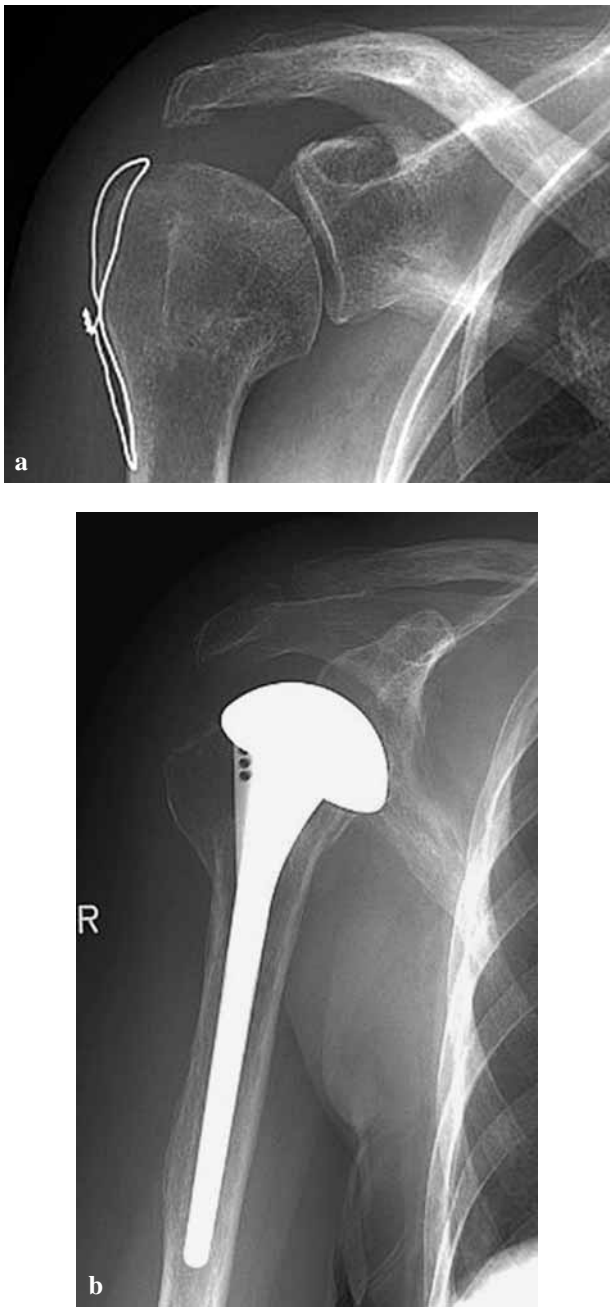

Fig. 1. — a. Radiograph of the right shoulder demonstrating the cerclage wiring of the previous greater tuberosity fracture ; b. Radiograph of the right shoulder taken 3 months after insertion of a CTA humeral head prosthesis. No radiolucencies are visible in acromion, distal clavicle, glenoid or humerus ; c. Radiograph of the right shoulder taken four and a half months after implantation of the CTA humeral head prosthesis. Radiolucencies can be seen in the glenoid, the acromion, the distal clavicle and the proximal humerus.

shoulder and global swelling of the rest of the right shoulder as compared to the contralateral side. Radiographs demonstrated the presence of an uncemented CTA prosthesis with radiolucencies in the humeral canal, the glenoid, the acromion and the distal clavicle (fig 1c). There was soft tissue swelling around the shoulder. These radiolucent changes were not as clear on the radiographs taken 6 weeks earlier (fig 1b). Laboratory investigations showed a CRP level of 30.5 mg/L (normal value < 5 mg/L) and a white cell count of 23.7/mcL (normal value between 4.0 and 10.0). Because of the suspicion of an infection, it was decided to take deep biopsies to identify the pathogen since previous (superficial) aspirations were sterile. The previous deltopectoral incision was used but no structures could be identified. The prosthesis was entirely loose and was removed without difficulties. The humeral canal, the glenoid, the subacromial space and the distal clavicle were all invaded by abnormal tissue. Multiple biopsies were taken and because of the malignant appearance of the soft tissues we decided not to implant a spacer. The

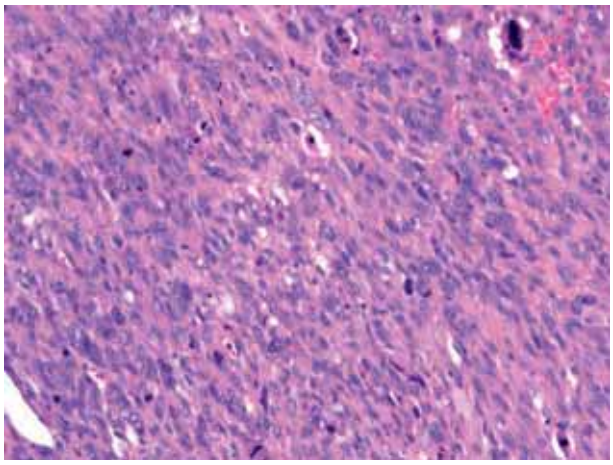

Fig. 2. — Histologic analysis of the biopsy specimens revealed the presence of a tumour consisting of atypical spindle cells with hyperchromatic nuclei and multiple mitotic figures. (Haematoxylin & Eosin stain, ×450).

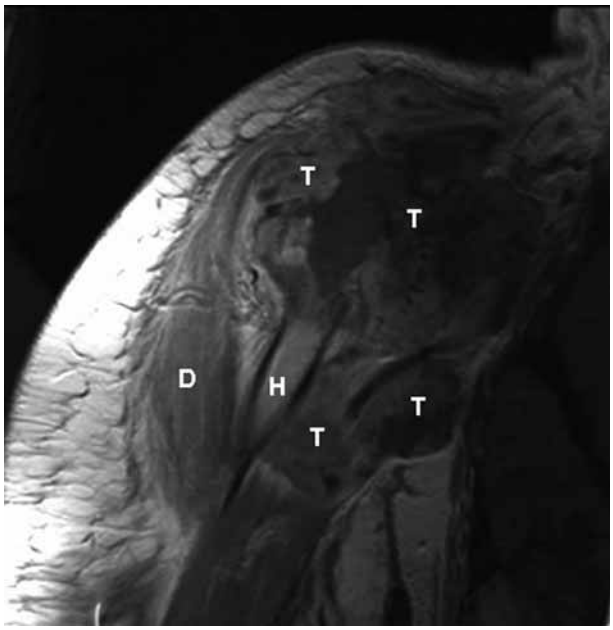

Fig. 3. — The postoperative MRI scan of the shoulder shows the large tumoral mass (T) extending towards the chest and the lower part of the upper arm (H = humeral shaft ; D = deltoid).

cultures demonstrated the presence of *Propionibacterium acnes* and coagulase negative *Staphylococcus*. Histological examination showed a highly cellular and pleomorphic sarcomatous proliferation, consisting of plump to spindly cells with atypical nuclei and numerous mitotic figures, some of which being atypical (fig 2). Numerous multinucleated giant cells were present. Immunohistochemically some myogenic differentiation was established in view of the scattered expression of desmin and alpha smooth muscle actin. MRI scan of the right shoulder region, taken after removal of the prosthesis, demonstrated the presence of a large tumoral soft tissue mass ($13 \times 11 \times 16.5$ cm) in the upper right shoulder region extending from the anterior aspect of the shoulder to the axilla and the pectoral muscles anteriorly and between the chest wall and the scapula posteriorly, with ingrowth in the chest wall (fig 3). A CT scan of the chest also showed this large tumoral mass in the right shoulder. There was a small pleural effusion and a small metastasis in the right lung. A CT scan of the abdomen was normal. A technetium scan showed increased tracer uptake in the right shoulder region extending halfway into the diaphysis of the humerus. There was no evidence for other bony metastases.

Since we did not expect the diagnosis of a malignant tumour in this patient we reviewed the radiographs and the CT scan of the shoulder that were taken five months earlier, just before the implantation of the shoulder prosthesis. There was no evidence for any tumour on these images. Moreover, during implantation of the shoulder prosthesis, no tissue abnormalities were observed. The patient was considered to be inoperable because of the extensive local invasion of the tumour and the presence of lung metastases. She was referred to the Oncology Department for further chemotherapy, which she declined. She died one month later.

DISCUSSION

The development of postradiation sarcomas is a well-known but rare complication after irradiation of soft tissues. Postradiation sarcomas can arise in bone (postradiation osseous sarcoma) or in soft tissues (postradiation soft tissue sarcoma). The latter account for 0.5%-5.5% of all sarcomas (*3, 5*). The shoulder is particularly vulnerable because it is

often exposed to X-rays after ipsilateral removal of breast tumours. Radiation-induced soft tissue sarcomas exhibit a wide range of histological types. Malignant fibrous histiocytomas are most commonly seen (*4, 6, 9*). Leiomyosarcomas seem to be underdiagnosed and are frequently misclassified as fibrosarcomas or unclassifiable tumours. Specific immunohistochemical stainings and electronmicroscopy can demonstrate smooth muscle differentiation in these types of sarcomas as shown in the present case (*2, 7*). Postradiation soft tissue sarcomas usually occur in older patients and this is mainly due to the long latency period. Several studies have shown that the mean latent period between the initial radiation and the clinical diagnosis of radiation-induced neoplasms is 10-20 years (*1, 3, 5-8*). The prognosis of radiation-induced soft tissue sarcomas depends on its location. Localisation in the spine, the pelvis and the shoulder girdle seems to be associated with a poorer prognosis (*10*). Other factors associated with a poor prognosis include large size of the primary tumour, histopathological differentiation and incomplete surgical removal. Wide surgical resection remains the treatment of choice, but frequently the disease is too advanced to achieve this.

The case presented here is interesting for two reasons. First, it illustrates that postradiation sarcomas should be suspected in patients who develop pain and/or swelling in previously irradiated regions, even after a period of 20 years. Second, infection can mask the presence of a tumour. When we see a patient with a painful shoulder arthroplasty, usually an infection or aseptic loosening is suspected. However, when dealing with previously irradiated areas we should include postradiation sarcomas in our differential diagnosis. We should also keep in mind that, as illustrated in this patient, infections and tumours may occur together. A careful medical history and a good clinical examination, together with proper technical investigations should allow to differentiate between an infection or a tumour.

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