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Case report

Fibrosarcoma in association with a total knee joint prosthesis

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Summary. A case of fibrosarcoma of the proximal tibia, arising at the site of a cemented Richards total knee joint replacement with cobalt chrome alloy is reported. The patient, an 80-year-old man, received the endoprosthesis because of osteoarthritis of the knee 4 years ago. The literature on malignant tumours occurring in association with endoprostheses is summarized and briefly discussed.

Key words: Fibrosarcoma – Knee joint replacement – Endoprosthesis

Introduction

In the past 15 years, total knee joint replacement has become the most successful surgical therapy for osteoarthritis of the knee. Today the most important problem in long-term follow-up of patients with knee joint replacement is aseptic loosening of the implant. Only 1 case of a malignant tumour in association with such a prosthesis has been reported (Weber 1986). In comparison with this, 13 cases of malignant tumours occurring next to a hip joint endoprosthesis and 10 next to other metallic implants have been published (Table 1). In an editorial Apley (1989) emphasized the importance of reporting all cases of the association of malignancy and joint replacement, assuming that the reported cases are the "tip of an iceberg". This idea is supported by Moore (1991), who suggested a registry of foreign-body-associated tumours with preservation of tissues for further analyses. In accordance with Apley (1989) we assume that the case reported by Weber (1986) and the present are only the first cases in a series of publications. The importance of this problem may be illustrated by the assumption that about 300,000–400,000 joint replacements are performed worldwide every year (Goldring et al. 1983; Tait et al. 1988).

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Case report

In January 1987 a 76-year-old man was admitted to a peripheral hospital because of severe osteoarthritis of the left knee joint. A Richards total knee joint replacement was implanted. Three weeks after the operation the tibial tuberosity tore out and was fixed with plates. Some weeks after this revision it came out again and was re-attached with wires and screws. Post-operatively, a *Pseudomonas aeruginosa* wound infection developed and the skin wound became necrotic. After treatment with ofloxacin the wound healed and the patient was discharged.

He was admitted to the University Hospital in August 1987 because of a fistula from the tibial tubercle to the skin at the site of the knee replacement. Pre-operative radiography of the chest showed uniform ventilation on both sides without infiltrate. Radiography of the knee joint showed no signs of a malignant tumour. The plates, screws, wires and a sequestered bone fragment with tissue from underneath were removed. Histological examination showed chronic non-specific osteomyelitis without any sign of a malignant tumour. The wound healed up and the patient was discharged. The patient felt well until November 1990 when he was admitted again to the University Hospital with indigestion and diarrhoea. Colonoscopic examination showed diverticulosis. Additionally he complained of pale green expectoration but without shortness of breath since October 1989. Radiography of the chest showed multiple nodules in the lungs and massive effusions in both pleural cavities. The pulmonary masses were suspected to be metastases of an unknown tumour. Because of the patient's age and his physical condition no further extensive investigations were performed. At the beginning of February 1991 the patient died of respiratory failure.

The most important autopsy findings were multiple solid, greyish-white, partly haemorrhagic tumours in both lungs, visceral pleura, parietal pleura, and diaphragm. Small- and medium-sized pulmonary arteries of both lungs contained multiple thrombo-emboli. The right pleural cavity contained 1500 ml and the left 800 ml of haemorrhagic effusion.

Intensive examination for the primary tumour disclosed only at the site of the total knee joint replacement a soft, greyish-white lobulated mass in the area of the tibial tubercle. The tumour measured about $3 \times 3 \times 2$ cm and was intraosseous, adjacent to the cement of the implant (Fig. 1). The complete tumour in the tibia next to the implant was embedded and 20 histological sections were made. Although no other evidence of a primary tumour was found at post-mortem examination, an unrevealed tumour of microscopical size cannot be excluded with certainty.

Histological examination of the sections shows numerous rather uniform, large, spindle-shaped cells. They vary little in size and

Table 1. Reported cases of malignant tumours associated with orthopaedic implants

Author	Age/sex	Site	Time lapse	Histology
McDougall (1956)	42/M	Humerus	30 years	Ewing's sarcoma
Delgado (1958)	40/M	Tibia	3 years	Osteosarcoma
Castleman and McNeely (1965)	56/M	Femur	3 years	Giant-cell rich sarcoma
Dube and Fisher (1972)	84/M	Tibia	26 years	Haemangioendothelioma
Tayton (1980)	11/F	Femur	7 years	Ewing's sarcoma
McDonald (1981)	48/M	Tibia	17 years	Malignant lymphoma
Dodion et al. (1983)	49/M	Femur	1 year, 9 months	Immunoblastic lymphoma
Bago-Granell et al. (1984)	77/F	Femur	2 years	Malignant fibrous histiocytoma
Swann (1984)	63/M	Iliac crest and femur	3 years, 4 months	Malignant fibrous histiocytoma
Lee et al. (1984)	44/M	Femur	15 years	Malignant fibrous histiocytoma
Penman and Ring (1984)	75/F	Iliac fossa and femur	5 years	Osteosarcoma
Weber (1986)	76/F	Knee	5 years	Epithelioid sarcoma
Bauer et al. (1987)	66/F	Femur	10 years	Osteosarcoma
Hughes et al. (1987)	42/M	Femur	30 years	Malignant fibrous histiocytoma
Ryu et al. (1987)	40/M	Pelvis	1 year, 4 months	High-grade soft-tissue sarcoma (periacetabular)
Lamovec et al. (1988)	62/M	Hip	12 years	Spindle-cell sarcoma of the synovia
Martin et al. (1988)	60/F	Hip	10 years	Telangiectatic osteosarcoma
Tait et al. (1988)	56/F	Hip	11 years	Malignant fibrous histiocytoma
Van der List et al. (1988)	72/F	Hip	11 years	Malignant epithelioid haemangioendothelioma
Haag and Adler (1989)	69/F	Femur	9 years	Malignant fibrous histiocytoma
Brien et al. (1990)	60/F	Hip Femur	8 years	Osteogenic sarcoma
Nelson and Phillips (1990)	72/F	Femur	8 months	Malignant fibrous histiocytoma
Ward et al. (1990)	65/F	Femur	9 years	Osteogenic sarcoma
Scully et al. (1991)	18/M	Femur	11 years	Osteogenic sarcoma
Presented Case	80/M	Knee	4 years	Fibrosarcoma

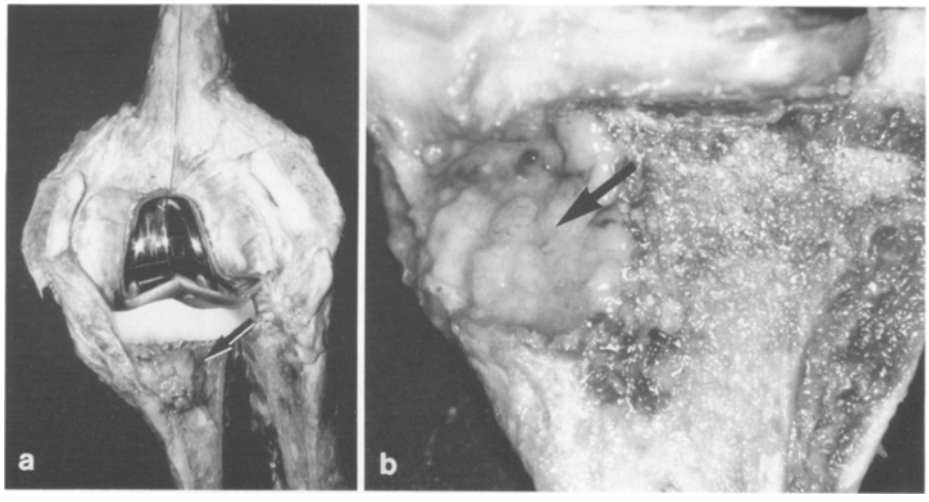

Fig. 1 a, b. Macroscopic findings. **a** Left knee joint with implanted total joint prosthesis. A longitudinal section through the specimen was performed. The tumour (*arrow*) was adjacent to the artificial polyethylene socket and the metallic stem of the tibial implant. **b** Close-up of the region of the tuberositas tibiae. The tumour (*arrow*) showed a soft, greyish-white cut surface and extended from the implanted stem to the bone surface

shape and are arranged in typical "herring-bone" patterns, embedded in a matrix of collagenous fibres (Fig. 2). From this uniform appearance the tumour is considered to be a well-differentiated fibrosarcoma. There is no particulate debris of bone cement or the metallic implant in, or next to, the tumorous tissue. Histological examination of the pulmonary nodules shows similar findings to the tumour in the leg leading to the assumption that they are metastases of the primary tumour close to the endoprosthesis. Multiple immunohistological stainings on formalin-fixed tissue from the primary tumour and the pulmonary nodules with antibodies against cytokeratin, S-100 protein and vimentin are negative in this post-mortem tissue.

Discussion

This is believed to be the first reported case of fibrosarcoma in association with joint replacement and the sec-

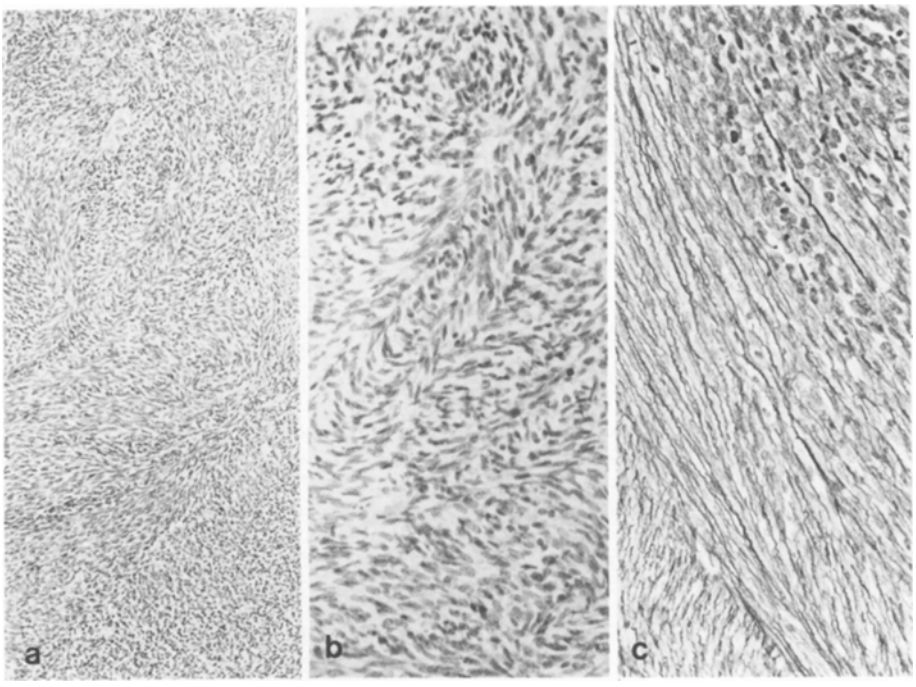

Fig. 2a–c. Microscopic findings. **a** Histological examination of the tumour from Fig. 1 showed a well-differentiated fibrosarcoma with regular and rather monotonous cells arranged in typical "herring-bone" patterns. H & E, ×63. **b** Cells and nuclei with a great uniformity arranged in a characteristic fascicular pattern. H & E, ×160. **c** Argentophil fibres aligned parallel between the tumour cells. Gomori, ×160

ond report of a malignant tumour next to a total knee prosthesis. Weber (1986) reported the case of an epithelioid sarcoma in association with total knee replacement. Fibrosarcomas next to artificial implants were reported by Burns et al. (1972), Herrmann et al. (1971) and O'-Connell et al. (1976), who described fibrosarcomas occurring in the neighbourhood of a vascular prosthesis made of woven Teflon-Dacron or Dacron threads. Within the known limitations (Enzinger and Weiss 1988) for the differential diagnosis the present tumour was diagnosed as a fibrosarcoma because of its characteristic histological appearance and the lack of other differentiation in multiple tissue sections.

The knee implant used in the present patient was a cemented Richard's maximum contact prosthesis. It is tri-compartmental and made up of cobalt chrome alloy and high-density polyethylene. The tibial implant is polyethylene with a metal endoskeleton and a metal fixation stem (Laskin 1986).

Various authors have shown the occurrence of several malignant tissue tumours at the site of total hip replacements or metallic osteosynthetic material (Table 1). The ages of the patients were from 11 to 84 years (mean 57 years). Time lapse from implantation to diagnosis of the tumour varied from 16 months up to 30 years (mean 10 years). The reported survival time of 13 patients ranged from 0.5 to 36 months with a mean of only 9.1 months. Eight authors (Delgado 1958; Castleman and McNeely 1965; Bago-Granell et al. 1984; Lee et al. 1984; Bauer et al. 1987; Tait et al. 1988; Van der List et al. 1988; Brien et al. 1990) lost their patients to follow-up or gave no further details; Scully et al. (1991) described a patient who is still in treatment and only McDonald (1981) reported a patient with malignant lymphoma who was asymptomatic after chemo/radiotherapy. These reports show that malignant tumours in association with joint prosthesis are very rare events, but if they occur they have a poor prognosis.

The incidence of malignancy after implantation of prostheses is uncertain. Gillespie et al. (1988) showed that after hip replacement the risk of developing a "lymphoreticular" neoplasm increased from 2 per 1000 over 10 years in the population as a whole to 6 per 1000 in the same period in the operated group. Visuri and Koskenvuo (1989) reported that patients with a chrome-cobalt-molybdenum prosthesis have a cancer morbidity that differs from that of the normal population. They reported an increase in the risk of leukaemias and lymphomas but a decrease in the risk of breast cancer.

Hamblen and Carter (1984) and Apley (1989) writing in editorials, called for the publication of all cases of malignancy associated with artificial implants. We have seen two further unpublished cases of malignant tumours developing next to orthopaedic prostheses diagnosed in our Department of Pathology during the last years. The first case was an 81-year-old man with an angiosarcoma next to a total hip joint prosthesis, the second a 76-year-old woman with a malignant lymphoma (Burkitt-type) adjacent to a total knee prosthesis. Unfortunately no further clinical data were available from these patients. This suggests strongly that there must be a higher prevalence of malignant tumours in association with joint replacements and orthopaedic implants than published in the literature so far. Rarity and the reported long mean time lapse (more than 10 years) between implantation and a possible malignant tumour may be reassuring for all patients with an artificial joint or implant.

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