Introduction: Paget’s disease of bone is a pathologic condition marked by increased bone resorption and formation. The exact etiology remains to be fully understood, however a slow paramyxovirus infection occurring in-patients with a genetic predisposition is suspected (1-4). Among the different location, the proximal part of the femur and the acetabulum are involved in up to 80 percent of patients with Paget’s disease. Also, osteoarthritis of the hip joint is frequently associated with Paget’s disease with medial or concentric wear patterns. The authors report a case of a monostotic Paget’s disease of the pelvis transferred to the uninvolved ipsilateral mid femur following total hip arthroplasty.

Methods: A sixty-six-year-old man was referred to our department in 1993 for an intractable pain in the left groin related to an osteoarthritis of hip. This patient had an untreated monostotic Paget’s disease involving the left hemi-pelvis. No other location of the disease, notably in the ipsilateral femur, was present on plain radiographs. The patient underwent a cemented total hip arthroplasty. The femoral medullary canal was blocked using a bone plug made of cancellous bone retrieved from the acetabulum and the proximal femur (Fig. 1). Macroscopically, usual lesions of osteoarthritides were noted. Specimens retrieved from the capsule and the synovial membrane showed usual degenerative fibrosis with no inflammatory cells on a microscopic scale. One year after hip replacement, an asymptomatic distal lytic lesion of the femur was observed below the tip of the stem (Fig. 2). The stem had not migrated, and wear of the acetabular component was undetectable on plain radiographs of the pelvis. Laboratory tests, including C-reactive protein level and erythrocyte sedimentation rate, showed no sign of infection. The level of the alkaline phosphatase was twice the normal. A bone scan made with technetium 99-labeled methylene diphosphonate showed intense tracer uptake in the left hemi-pelvis, but also in the mediodiaphyseal region of the left femur around the tip of the stem (Fig. 3). No other sites of involvement were apparent.

To avoid a fatigue fracture of the femur below the stem, the patient was revised through a transtrochanteric approach in 1996. The acetabular component was left in place as it showed no sign of macroscopic wear and was not loosened. The femoral stem that was judged as stable by mean of intra-operative examination was removed. Multiple specimens were harvested from the distal femur, including the bone plug, with the patient’s informed consent obtained prior to the procedure. The biopsy specimens were fixed in 10 percent formalin and embedded in paraffin. Sections 5 µm thick were obtained and surface stained with haematoxylin and eosin for standard and polarized light microscopy. A long-stemmed femoral component was used to by-pass the ostelytic region of the mid-diaphysis.

Results: There was no evidence of deep infection, and all the cultures remained sterile. Histological analysis of the specimens revealed typical aspect of Paget’s disease of bone with a mosaic pattern associated with multinucleated osteoclasts (Fig. 4). Under polarized light, no birefrgent particles suggesting polyethylene wear debris were observed. The patient underwent antipagetic treatment, including calcitonin (100 International Units 3 times a week) during two months. However, the level of the alkaline phosphatase remained 1.78 times over the normal level. Although no further extension of the osteolytic lesion was noted, there was no radiologic bone remodeling indicating new bone formation. A bisphosphonate treatment was therefore started, using etidronate at a dose of 400 milligrams per day for five months. The alkaline phosphatase level decreased regularly and remained normal afterwards. The lytic lesion of the femur was progressively replaced by non-pagetic newly formed bone on a radiographic scale. The hip continued to be graded as excellent, up to five years after femoral revision. No other location of the disease had appeared.

Discussion: A number of data have supported a viral etiology for Paget’s disease of bone. Indeed, it has been proposed that the localized disorder of bone remodeling occurred as a result of a viral infection of osteoclasts in pagetic bone. Inclusions similar to nucleocapsids have been described in the nuclei and cytoplasm of osteoclasts at pagetic sites, but not in non-pagetic osteoclasts from the same patients. A current unifying hypothesis has suggested that the functionally hyperactive osteoclasts in pagetic bone were a product of a virus-mediated increase in cell fusion between osteoclasts and osteoclasts progenitor cells that migrated to pagetic sites. In the current case, it is highly probable that the occurrence of the pagetic femoral lesion in a previously uninvolved bone is related to the use of autogenous pagetic bone graft that was retrieved from the pelvis. This hypothesis is supported by the fact that in most instances, sites affected with Paget’s disease when the diagnosis is made are the only ones that will show pagetic changes over time. Although progression of the disease within a given bone may occur, the sudden appearance of new sites of involvement years after the initial diagnosis is uncommon. Moreover, the extensive analysis of the dynamic radiologic patterns of Paget’s disease have shown that in long bones, Paget’s disease usually began at one end of the involved bone, and subsequently extended towards the other end. Therefore, although the possibility of a sudden pagetic site in the mid-uninvolved femur, as been observed herein, related to the extension of the disease, cannot be completely eliminated, this hypothesis is not the most likely.

To the best of the authors’ knowledge, transfer of Paget’s disease through autograft bone has never been described in the literature as a complication of total hip arthroplasty for Paget’s disease. The authors believe that this observation support at least a partially viral etiology for Paget’s disease through a contagious infectious process of osteoclasts. Moreover, the current reported case indicates that orthopaedic surgeons should be aware of the risk of transfer of Paget’s through the use of pagetic autograft bone. This complication has not been reported with the use of deep frozen or irradiated allograft bone, although pathological lesions other than osteoarthritias, including Paget’s disease, can be present in osteoarthritic femoral heads that are considered suitable for bone-bank donation.

References