

# Cementless total hip arthroplasty in Paget's disease of bone: a retrospective review

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**Abstract** Paget's disease of bone (PDB) is a localised chronic osteopathy leading to bone deformities, bone hypervascularity, structural weakness and altered joint biomechanics. The pelvis and upper femur are frequently involved, resulting in disabling hip disease, and total hip arthroplasty (THA) may be required. We performed a retrospective study on the management and the outcome of 39 uncemented hydroxyapatite fully-coated THA in patients with PDB of the hip. The follow-up averaged 79.4 months (range 24–194). Functional scores improved significantly and, using the Harris hip score, 84% of patients had an excellent clinical outcome at the latest follow-up. Despite one case of an uncemented acetabular component with probable loosening, no implant revision had been required at our latest follow-up. Signs of implant loosening were found to be significantly more frequent in patients with active disease. For this reason, we advocate the use of pre-operative medication with bisphosphonates to reduce disease activity. Another benefit of this treatment is the significant decrease of intra-operative blood loss.

Provided the control of disease activity in the pre-operative period with bisphosphonates is achieved, good outcome of cementless THAs can be expected. Bisphosphonates reduced the risk of implant loosening and excessive intra-operative blood loss.

## Introduction

Paget's disease of bone (PDB) is a localised chronic osteopathy characterised by an osteoclastic dysfunction with increased resorption and subsequent compensatory formation of new woven bone with an anarchic microstructure [28]. This structural change produces bone that is expanded in size, more vascular and may lead to osseous deformities, structural weakness and altered joint biomechanics [29]. Prevalence of PDB has been shown to increase with age and the most commonly involved sites include the pelvis, femur, spine, skull and tibia. The pelvis and upper femur are involved in 20–80% of patients with PDB resulting in frequent disabling hip disease [9]. Deformities such as coxa vara, femoral bowing, acetabular protrusio and bony enlargement can be observed and may lead to Paget's arthropathy [15]. The increased bone turnover and remodelling is associated with elevated levels of serum alkaline phosphatase (sAP). Such a marker allows assessment of the activity of the Paget's disease and monitoring of the effectiveness of the medical treatment by bisphosphonates [2, 4].

Although total hip arthroplasty (THA) has been reported with successful relief of pain in such cases, long-term outcome of cemented THA in these patients has raised concerns about the potential for implant loosening [16]. Recently, the use of cementless implants in patients with PDB has been reported with encouraging results [1, 11, 14, 17, 27].

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The purpose of our study was to report the management and the short- to mid-term outcome of 39 cementless THAs implanted in pagetic bone as well as to evaluate the relationship between disease activity and the risk for uncemented implant loosening, and the benefit of a pre-operative antipagetic medication.

## Materials and methods

We retrospectively reviewed 39 total hip arthroplasties (THA) with acetabular and/or femoral cementless hydroxyapatite fully-coated components. These procedures had been performed at our institution between January 1992 and September 2006 in 32 patients (16 men and 16 women) with PDB affecting the adjacent pelvis, proximal femur or both (Table 1). There were 21 right hips (54%) and 18 left hips (46%). Ten patients (30%) presented with a monostotic form of PDB and 22 (70%) with a polyostotic type. The average age at surgery was 74.2 years (range 55–89) and the average time to diagnosis of PDB was 13.1 years (range 1–36). The average body mass index was 25 kg/m<sup>2</sup> (range 17–32). The 39 procedures included 37 primary replacements and two revision procedures. One revision was performed 41 months after the index surgery for instability (four dislocating events) and consisted of a revision of the acetabular implant to an uncemented cup. The second revision was performed for aseptic loosening of both uncemented acetabular and femoral components 25 years after the index THA. For these two cases, the index surgery was performed in another institution. The revision procedure presented no particular problems and bone stock permitted us to perform cementless revision with standard primary implants. Eight patients (25%) died of unrelated causes at an average of six years (range two to 11 years) after the index surgery. For these patients data were available until death. At the latest follow-up, one patient was lost to follow-up five years after the index operation. All patients were pre-operatively treated by intravenous infusion of pamidronate two to three months before surgery to decrease operative bleeding. Each patient received 180 mg of pamidronate in two intravenous infusions of 90 mg for two days. The mean follow-up for the entire series was 79.4 months (range 24–194).

Charnley score, Postel–Merle d’Aubigné (PMA) score and Harris hip score (HHS) were used to assess patients’ pre- and post-operative status at the latest follow-up. Activity of the PDB was monitored by sAP level before surgery, at the time of the surgery and at latest follow-up (normal value range 40–120 U/l). Blood loss was evaluated using the Mengal formula [22].

Anteroposterior (AP) and lateral radiographs of the pelvis were used for radiographic evaluation. Pre- and post-operative

radiographs at the latest follow-up were analysed by two independent observers. Preoperative radiographs were analysed for: (1) acetabular and femoral bone quality following the three histological stages of PDB (osteolytic or osteosclerotic or mixed features of PDB) [19], (2) coxa vara (neck-shaft angle less than 120°), (3) acetabular morphology (protrusio, expulsive and normal morphology) and femur morphology (normal or bowed), and (4) leg length discrepancy. Acetabular protrusio was determined by the relationship of the femoral head to Köhler’s line.

Postoperative radiographs were analysed for: acetabular cup angle, orientation of the stems (varus/valgus), heterotopic ossifications based on Brooker’s classification, signs of implant loosening, bone graft integration and leg length discrepancy. The acetabular cup angle was measured in reference to the teardrop on AP radiographs. The horizontal reference line was drawn connecting the points at the bottom of both teardrops. Orientation of the femoral component was measured with reference to the femoral shaft axis.

The femoral implant loosening was analysed according to Gruen’s zones. At the femur, loosening and instability of cementless component were assessed according to the criteria described by Engh et al. [5]. Extensive reactive lines around the porous-surfaced portion of the implant were considered to be a major sign for the absence of osseointegration. Reactive lines were defined as extensive if they were present in at least one-half of the Gruen zones where porous coating was present. Others signs of instability include a subsidence of the stem greater than 2 mm, a bone pedestal beneath the stem associated with radiolucent lines surrounding the distal stem, stress shielding and a calcar hypertrophy on successive radiographs. For a cemented femoral implant, loosening was assessed according to the classification of Harris et al. [10]. Definite loosening was defined as subsidence of the femoral component, fracture of the cement mantle or the stem, or the presence of a radiolucent line not seen on early post-operative radiographs at the prosthesis-cement interface. Probable loosening was defined as the presence of a continuous radiolucent line along the entire bone-cement interface. Possible loosening was defined as the presence of a radiolucent line occupying more than 50% but less than 100% of the bone-cement interface on any radiograph, or the presence of a progressive radiolucent line.

The acetabular implant loosening was analysed according to De Lee’s zones to evaluate the location of radiolucent lines and osteolysis. Radiographic loosening of the cementless acetabular component was defined as a progressive circumferential radiolucency greater than 2 mm. Cementless acetabular component migration was defined as a change in the vertical or horizontal position of at least 3 mm or a change of the

**Table 1** Baseline characteristics of patients

Case number	Gender	BMI (kg/m <sup>2</sup> )	PDB location	Side	Age at surgery (years)	Location of cementless components	Primary or revision THA	Follow-up (months)
1	F	26	A	R	76	B	Primary	25
2	M	31	A	R	79	B	Primary	24
3	F	28	A	L	83	B	Primary	43
4	F	27	A	L	75	B	Revision	36
5	M	27	A	L	79	B	Primary	84
6	M	20	A	B: L next R	82	B	Primary	55/67
7	F	25	B	R	81	B	Primary	35
8	M	27	A	L	84	B	Primary	37
9	M	32	A	R	64	B	Primary	50
10	M	18	A	R	55	FM	Primary	49
11	F	28	A	B: L next R	74	B	Primary	43/150
12	F	23	A	L	84	B	Primary	47
13	M	27	FM	R	76	B	Revision	62
14	F	22	A	L	75	B	Primary	63
15	F	18	A	L	70	B	Primary	63
16	M	31	A	B: L next R	64	B	Primary	76/158
17	M	24	B	L	82	FM	Primary	73
18	M	26	B	L	81	B	Primary	25
19	F	28	A	B: L next R	74	B	Primary	26/160
20	M	24	B	B: R next L	78	B	Primary	82/85
21	F	25	A	L	77	B	Primary	109
22	M	20	A	B: R next L	85	B	Primary	105/194
23	F	27	A	R	74	B	Primary	36
24	F	22	A	R	89	B	Primary	94
25	M	24	A	R	75	B	Primary	36
26	F	22	A	R	75	B	Primary	58
27	M	29	A	B: R next L	58	B	Primary	121/193
28	M	22	B	R	68	A	Primary	121
29	M	28	B	R	76	B	Primary	98
30	F	24	A	R	73	B	Primary	85
31	F	28	FM	L	75	B	Primary	97
32	F	22	A	R	72	B	Primary	133

*BMI* body mass index, *PDB* Paget's disease of bone, *THA* total hip arthroplasty, *M* male, *F* female, *FM* femur, *A* acetabulum, *B* both, *R* right, *L* left

cup angle of at least 5° on comparable radiographs, as measured using the criteria of Massin et al. [20]. For cemented acetabular components, definite loosening was defined as migration of the component or the presence of any new fracture in the cement mantle, probable loosening was defined as the presence of a circumferential radiolucent line around the entire component at the bone–cement interface and possible loosening was defined as the presence of a radiolucent line around 50–99% of the component at the bone–cement interface.

Analysis of the results was carried out using the SPSS 16.0 program for Windows (SPSS Inc., Chicago, IL, USA) and Student's *t*-tests were undertaken (level of significance,  $p < 0.05$ ).

## Results

Thirty-nine uncemented hydroxyapatite fully-coated THAs were implanted in patients with PDB of the hip. The follow-up length of the entire series averaged 79.4 months (range 24–194).

### Preoperative data

PDB exclusively involved the acetabulum in 31 hips (80%), exclusively involved the femur in two hips (5%), and both acetabulum and femur were involved in six hips (15%) (Fig. 1). Two patients had undergone a previous hip surgery (revision of a previous THA).

**Fig. 1** Pre-operative anteroposterior (AP) pelvis and plain lower limb radiographs of a 78-year-old woman with full pelvis and right femur involved by Paget's disease. Consequences are right coxa vara, right bowed femur and bilateral hip osteoarthritis



The average Harris hip score was 54 points (range 37–68 points). The average PMA score was 9 points (range 2–13). Seven patients were Charnley A (22%), 11 Charnley B1 (34%), eight Charnley B2 (25%) and six Charnley C (19%).

The average level of sAP was 332.6 U/l (range 156–1408 U/l) before pre-operative pamidronate infusions.

In the 37 pagetic pelvises, 22 (59%) were sclerotic on X-rays, 15 (41%) had a mixed lytic and blastic form, and no patient presented with exclusive osteolytic feature. Acetabular protrusion was observed in 12 cases (32%). In eight femurs with pagetic involvement, four (50%) were bowed and six (75%) were sclerotic. Coxa vara was reported in seven hips (18%). Leg length discrepancy was reported in six cases (15%) with an average shortening of the leg of 1.4 cm (range 1–3).

#### Surgical data

At the time of surgery, the average sAP level was 125.7 U/l (range 28–395 U/l). A 38% decrease of sAP activity was reported after pre-operative intravenous infusions of pamidronate ( $p < 0.001$ ). Thirty-one out of the 39 THA (79%) had been performed in patients with inactive disease (sAP < 120 U/l) at the time of the surgery.

Hip replacements were performed under general anaesthesia in 38 cases and under spinal anaesthesia in one case. An anterolateral approach was performed in 34 cases (87%) and a posterolateral approach in five cases (13%). In 36 cases, both acetabular and femoral uncemented hydroxyapatite fully-coated components were used (Fig. 2). In three cases, the use of a cemented acetabular or femoral

component was required because the fixation without cement was considered likely to fail. Mechanical conditions but not PDB were the reason for the use of cemented components in these three cases. Cemented cups were used in two cases and a cemented stem in one case. Finally, 37 uncemented cups and eight uncemented stems were implanted into pagetic bone. Autologous bone grafting for acetabular defects was necessary in three cases (two major acetabular protrusion and one case of acetabular component revision).

A posterior column and roof fracture was the only intra-operative complication reported in a case with a sclerotic acetabulum, and it was successfully treated with a plate.

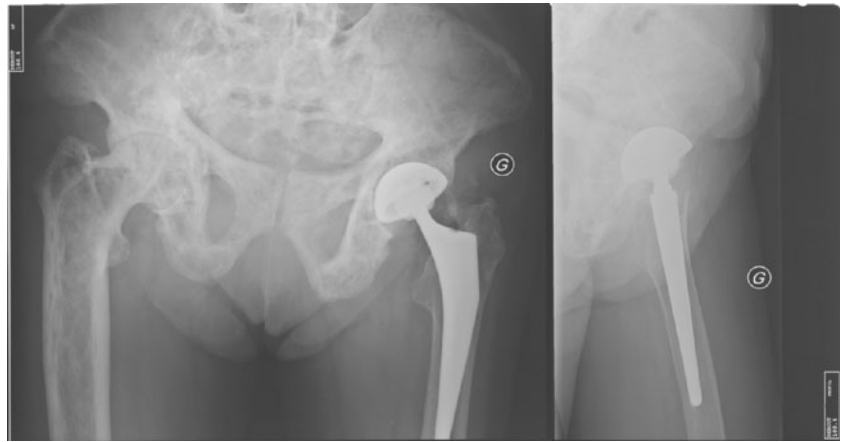
Average intra-operative blood loss was 744 ml (range 250–2,000 ml). Abnormal bleeding (>2,000 ml) was reported in 11 cases (28%) requiring red blood cell transfusions. In these patients, sAP values were found to be higher than in other patients ( $p < 0.001$ ), with average sAP values of 239.4 U/l (range 127–495 U/l) versus 94.3 U/l (range 28–270 U/l), respectively. Normal bleeding in patients without PDB is estimated to be around 200–450 ml at our institution.

#### Post-operative data

##### *Clinical outcome*

All patients except one were allowed immediate full weight-bearing post-operatively. In one case, a nonweight-bearing period of 45 days was necessary because of an intra-operative acetabular fracture. Two patients sustained a

**Fig. 2** Anteroposterior and lateral post-operative radiographs of the same patient 42 months after cementless total hip arthroplasty (THA)



reoperation for peri-prosthetic fractures after a fall, respectively at 38 months and 45 months after the index surgery, and were managed without revision of the implants. Additional post-operative complications included two deep venous thrombosis without pulmonary embolism and one haematoma.

Mean pre-operative Harris hip score improved from 54 points to 89 points (range 74–99 points) at the latest follow-up ( $p < 0.001$ ). The outcome was considered excellent or good (Harris hip score  $\geq 80$  points) in 27 patients (84%) and fair in five patients (18%). Among these five patients, two developed other joint involvement, two sustained a periprosthetic fracture (as mentioned above) and one was developed severe heterotopic ossification (Brooker class III).

Similarly, mean pre-operative PMA score improved from 9 points to 16 points (range 10–18) at the latest follow-up ( $p < 0.001$ ).

At follow-up, the average sAP level was 191.7 U/l (range 77–883 U/l). Twenty-three patients (72%) were treated with oral bisphosphonates with an average sAP level of 123.1 U/l (range 77–208 U/l) and nine patients (28%) did not receive any further treatment with an average sAP level of 220.7 U/l (range 85–883 U/l).

#### Radiographic outcome

The cup abduction was in a satisfactory position ( $40^\circ$ – $55^\circ$ ) in 36 hips (92%). Abduction angle was less than  $40^\circ$  in three hips (8%). The femoral component was in a neutral position in 35 hips (90%), in slight valgus ( $<5^\circ$ ) in three hips (8%) and in slight varus ( $<5^\circ$ ) in one hip (2%). Heterotopic ossification was observed in 22 hips (56%): Brooker I or II in 19 cases (49%) and Brooker III in three cases (7%). No significant correlation was found between sAP level and occurrence of heterotopic ossification ( $p > 0.05$ ) nor between Harris hip score and occurrence of heterotopic ossification ( $p > 0.05$ ).

At the latest follow-up, radiolucent lines around the stem were observed in four of the 38 uncemented stems (10.5%) (Table 2). Among these four stems, one (2.7%) had been implanted into pagetic bone. Radiolucent lines around the cup were observed in three of the 37 uncemented cups (8.1%) (Table 2). A probable loosening of the cup was observed in one case (case number 6). However, no migration of the cup was observed. None of these implants had been revised at the time of this study. All cemented components remained stable; however, none of them had been cemented into pagetic bone.

Implants with longer follow-up were more likely to present signs of loosening such as radiolucent lines. The average follow-up was 133.5 months (range 97–194 months) in THA with signs of loosening versus 65.6 months (range 24–160) in THA without ( $p < 0.001$ ). In addition, signs of loosening were found to be more likely to occur in patients with higher levels of sAP. The average level of sAP in cases of radiolucent lines was 343.3 U/l (range 95–883 U/l) versus 132.2 U/l in patients without (range 77–273 U/l) at the latest follow-up ( $p < 0.001$ ). Twenty-two patients with stable components and one patient with radiolucent lines were still

**Table 2** Signs of implant loosening at latest follow-up)

Case number	Femoral radiolucent lines (Gruen zones)	Acetabular radiolucent lines (De Lee zones)	sAP level at time of surgery (U/l)	sAP level at latest follow-up (U/l)	Follow-up (months)
1	1, 7 <sup>a</sup>	-	102	95	97
2	-	I, II <sup>a</sup>	79	273	98
3	1, 7	-	122	133	121
4	-	I, II <sup>a</sup>	156	376	133
5	1, 2, 7	-	167	300	158
6 <sup>b</sup>	1, 7	I, II, III <sup>a</sup>	395	883	194

<sup>a</sup> Components implanted into pagetic bone

<sup>b</sup> Case with probable loosening of the cup



treated with oral bisphosphonates at latest follow-up. Case number 6 (with probable loosening of the cup) did not receive anymore bisphosphonates at the latest follow-up. In this latest case, sAP level was increased from 395 U/l at the time of surgery to 883 U/l at follow-up.

The three cases where autologous bone grafting was necessary showed satisfactory osseointegration at follow-up. Among the six patients with pre-operative leg length discrepancy, one patient presented with a persistent 1.5-cm post-operative shortening of the leg.

## Discussion

This series reports the management and the short- to mid-term outcome of 39 uncemented hydroxyapatite fully-coated THAs in patients with PDB. To our knowledge, few studies have been reported with the use of cementless components into pagetic bone. At the latest follow-up, we report 84% excellent or good clinical outcome using the Harris hip score. No revision had been performed at our latest follow-up. Of the 38 uncemented femoral components implanted, eight had been implanted into pagetic bone. One (2.7%) of them presented with radiolucent lines without subsidence or loosening at follow-up. Of the 37 uncemented acetabular components implanted into pagetic bone, three (8.1%) presented with radiolucent lines at the latest follow-up. A probable loosening was reported in one of these three (2.7% of the overall uncemented cups). However, this patient (case number 6, Table 2) remained fully asymptomatic and no migration of the cup was observed on successive comparable radiographs. This case also had the longest follow-up (16.2 years) and the highest pre- and post-operative level of sAP (395 U/l and 883 U/l, respectively). Moreover, this patient was no longer receiving bisphosphonates at latest follow-up.

Specific antipagetic therapy is focussed on suppression of the activity of pagetic osteoclasts and is currently achieved by bisphosphonates. Two indications for medical treatment of PDB are the relief of symptoms and prevention of complications [25]. Suppression of the pagetic process has been clearly shown to improve symptoms in most of the patients. Nevertheless, pain related to secondary osteoarthritis may not respond to antipagetic treatment. Therefore, selected orthopedic procedures such as elective joint replacements may be required [18].

Intraoperatively, Paget's disease of the hip may present specific technical challenges [15]. First, hypervascularity significantly increases the risk for blood loss and may compromise cement fixation of the implants. Prior to surgery, the use of potent bisphosphonates has been advocated, although no controlled study is available [13]. The goal of such a recommendation is to reduce bone

hypervascularity and therefore to decrease intraoperative blood loss. Potent intravenous bisphosphonates, such as pamidronate and zoledronic acid, have been most commonly used in this pre-operative period [30]. Another benefit from the use of bisphosphonates is the control of the disease activity and avoidance of rapid post-operative osteolytic bone resorption [26]. Second, the surgeon faces bone deformities such as acetabular protrusion or femoral bowing. The use of an antiprotrusion cage and/or bone graft or femoral osteotomy may be necessary.

Cemented THAs have been reported with short- to mid-term satisfactory outcomes among pagetic patients [16, 21, 23, 24, 31, 32]. Nevertheless, concerns regarding an increased incidence of radiolucent lines around pagetic bone–cement interface and the potential for loosening have been raised at long-term follow-up [12, 21, 23, 24, 33]. Failure rates leading to revision as high as 15% have been reported with cemented components implanted into pagetic bone [21]. The authors reported a 13.5% rate of radiographic cup loosening and a 29% rate of radiographic stem loosening. At latest follow-up, good and excellent results were decreased [21]. Radiolucent lines have been reported in up to 40% of cemented cups implanted into pagetic bone in other series [24]. Inability to achieve a dry bed for cementation may compromise long-term fixation [15].

In recent years, the use of uncemented components in PDB has been proposed and reported with promising results with regard to the mid-term outcome and fixation [1, 11, 14, 17, 27]. However, the benefits from the use of uncemented components remain unclear as active PDB may compromise the fixation and altered quality and morphological features of pagetic bone may influence bone ingrowth into cementless implants. Nevertheless, reports have shown that patients with PDB do not have a compromised ability to heal fractures [16]. Therefore, successful osseointegration of uncemented components can be expected.

Implant loosening was found to be significantly more frequent in patients with higher level of sAP. For this reason, we advocate the use of pre-operative antipagetic medication in order to reduce disease activity. Another benefit of this treatment may be the significant decrease of intra-operative blood loss. As surgical procedures should be performed when the sAP level reaches its nadir, this treatment should be undertaken two to three months before surgery [30].

In agreement with other studies [27], heterotopic ossification was common in our series but remained limited (49% grades I and II versus 7% grade III). Increased risk for development of heterotopic ossification is around 23–52% after THA in PDB and is similar to those patients with a previous history of heterotopic ossification, hypertrophic osteoarthritis or post-traumatic arthritis [6]. No significant correlation was found between sAP level or Harris hip score and heterotopic ossifications.

The main strength of our series is the large number of pagetic hips included with a good short- to mid-term outcome. To our knowledge, the largest previous series included 33 hips with a 6.7-year average follow-up [17]. However, our study has some limitations. The series is retrospective without randomised comparison of cemented and uncemented components implanted in patients with PDB.

Our results are in agreement with previously reported series [1, 11, 14, 17, 27] and close to those described in cementless hydroxyapatite fully coated THAs implanted in patients without PDB [3, 7, 8, 34]. The control of PDB activity in the pre-operative period reduces the risk of implant loosening and the abnormal intra-operative blood loss. However, these results may deteriorate with time. For this reason, longer follow-up is mandatory.

In conclusion, the short- to mid-term outcome of uncemented THAs in patients with hip osteoarthritis secondary to PDB was satisfactory. No revision had been performed at 6.6-year average follow-up. As radiographic signs of loosening were associated with PDB biological activity, adequate control of the disease activity with bisphosphonates therapy is a prerequisite for surgery and PDB activity should be monitored after THA. For this reason, we advocate the use of perioperative antipagetic medication with bisphosphonates in order to reduce disease activity. Another benefit of this treatment is the significant decrease of intra-operative blood loss. Provided the control of disease activity in the pre-operative period with bisphosphonates is achieved, good outcome of cementless THAs can be expected.

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