# Controlled study of the prevalence of radiological osteoarthritis in clinically unrecognised juxta-articular Paget's disease

P S Helliwell, G Porter

**Abstract**

***Background*—Paget's disease of bone is common and often undiagnosed in the population. The association of Paget's disease and osteoarthritis is well described but only in cases ascertained in secondary and tertiary care centres to which they have been referred largely because of pain. This study represents an attempt to confirm the association between Paget's disease and osteoarthritis in a population previously unknown to have Paget's disease.**

***Methods*—Radiographs of people over 55 years that included the entire pelvis, sacrum, femoral heads and lumbar spine (mostly plain abdominal radiographs) were obtained from hospital records for the period 1993–95. Films were screened by a trained observer and the positive films were reviewed by a consultant radiologist who also examined a 1 in 10 sample of the negative films. A subsample of 153 confirmed positive cases were matched for age and sex using cases without Paget's disease and these pairs were assessed by two observers working in tandem. The hip joints were scored 0–5 using a modification of the original descriptive classification of Kellgren and Lawrence and minimum joint space of the hip was also measured.**

***Results*—Not all cases were available for assessment. A total of 248 films were included (137 without Paget's, 89 with unilateral and 22 with bilateral disease). The mean age of the cases and controls was 78.4 years and 77.4 years respectively with 66/45 male/female cases and 78/59 male/female controls. One hundred and twenty nine affected hips were available for comparison with 352 unaffected hips. Median joint space narrowing for the affected hip was 3 mm (range, 0–5 mm) and for the unaffected hip 4 mm (range, 0–6 mm, Mann-Whitney U test, p=0.00001). Median Kellgren and Lawrence grade for both groups was 0, with no statistical difference between the groups (Mann-Whitney U test, p=0.74). In terms of severity of osteoarthritis, there were 19 instances of grades 2+ in the unaffected hips, and only five in the affected hips.**

***Conclusions*—Pagetic coxopathy is characterised by loss of joint space, which may represent a secondary chondropathy. Although joint failure may result from this secondary chondropathy progression may be dependent on non-Pagetic factors. It is also possible that the usual radiological features of osteoarthritis may be modified or obscured by the Paget's disease.**

(*Ann Rheum Dis* 1999;**58**:762–765)

**Rheumatology and Rehabilitation Research Unit, University of Leeds**
P S Helliwell

**Airedale General Hospital, Keighley, West Yorkshire**
G Porter

Correspondence to:
Dr P S Helliwell, Rheumatology and Rehabilitation Research Unit, 36 Clarendon Road, Leeds, LS2 9NZ

Accepted for publication 19 August 1999

Paget's disease of bone is widely thought to cause osteoarthitis (OA) in adjacent joints. There are a number of reasons why this should be so including joint incongruity, altered joint mechanics because of bone deformity, hypervascularity of subchondral bone and increased subchondral bone stiffness. There is no direct experimental evidence examining these factors. The association therefore relies on anecdotes[1] and a recent study that found a higher prevalence of OA in the ipsilateral joint compared with the contralateral joint.[2] However, these studies are subject to selection bias—they are on symptomatic subjects referred to secondary and tertiary care. Paget's disease in the community is largely undiagnosed and there are no studies of OA in this undetected "Pagetic" population. The recent radiological survey conducted by the MRC Epidemiological Unit at the University of Southampton[3] provided an opportunity to look at the association between Paget's disease and OA on a "population" basis.

## Methods

### CASE ASCERTAINMENT

The radiographic survey methodology is given in Cooper *et al*.[3] Survey radiographs were obtained in Carlisle, Lancaster, Preston, Wigan, Warrington and Newcastle upon Tyne. Radiographs of people over 55 years that included the entire pelvis, sacrum, femoral heads and lumbar spine were obtained from records for the period 1993–95. Most of these films were plain abdominal radiographs although other films included barium enemas, intravenous urograms (IVUs) and pelvic radiographs. Paget's disease was indicated as a known diagnosis in only a few of these films (5 of 1000, 0.05%). Samples of about 1000 radiographs were obtained from each centre. Films were screened by a trained observer and the positive films were reviewed by a consultant radiologist who also examined a 1 in 10 sample of the negative films. Paget's disease was diagnosed using the following criteria: (a) increase in bone density, (b) increase in bone size (c) disorganisation of bone architecture (d) cortical thickening (e) enhancement of trabecular pattern (f) thickening of iliopectineal line. A

*Table 1 Modification of Kellgren and Lawrence grades used in this study*

| *Grade* | *Abnormalities* | *Criteria* |
|---|---|---|
| 0 | Normal | |
| 1 | OSP | Minor osteophytes |
| 2 | JSN +OSP | |
| 3 | JSN, OSP, SSC, CYST | JSN + 2 others |
| 4 | JSN, OSP, SSC, CYST | JSN + 3 others |
| 5 | JSN, OSP, SSC, CYST, FD | As grade 4 + FD |

OSP=osteophytes, JSN=joint space narrowing, SSC=sub-chondral sclerosis, CYST=sub-chondral cyst formation, FD=femoral head deformity.

sample of 153 confirmed positive cases were matched for age and sex with non-diseased cases and these pairs were assessed by two observers working in tandem.

### SCORING OF RADIOGRAPHS

Radiographs were read in tandem by two observers (GP, PSH). The site of Paget's disease was noted (pelvic, femoral, sacral, lumbar or combinations) and the film discarded if juxta-articular Paget's disease was not present. The hip joints were scored 0–5 using a modification of the original descriptive classification of Kellgren and Lawrence[4] (see table 1). Additionally, we measured the minimum joint space of the hip (MJS). This was measured with a plastic ruler at the point of greatest narrowing of the hip joint where clear cortical margins could be identified either side of the joint. This meant, in effect, that this distance was usually measured from the superior third of the joint. Measurements were taken to the nearest mm by the same observer (GP). To assess the intraobserver reproducibility of this technique we measured the minimum hip joint space in a selection of 27 radiographs (8 plain abdominal films, 10 intravenous urogram films, 9 barium enemas) using a blinded technique with a 28 day interval between measurements. Test-retest scores were (median and range) 4 mm (0–5 mm) at time 1, and 4 mm (0–5 mm) at time 2. Median of score difference was 0 mm (−1 to +1mm): identical scores were obtained in 45 of 54 hips.

While the radiographs from IVUs, barium examinations and plain abdominal films have the same radiographic geometry, in particular the same centering, this was not the case for films of the pelvis. To assess whether the joint space measurements differed between pelvic and abdominal films 10 randomly selected sets of films, taken from patients who had simultaneous radiographs of abdomen and pelvis, were assessed for joint space. No measurable difference in joint space was detected between the abdomen and the pelvic films. As a consequence the use of these different radiographs was not considered to be a significant source of error in this study although it is accepted that a more sensitive technique may have produced a different result.

## Results

Of the 153 matched pairs (henceforth described as cases and controls) only 111 pairs were available for scoring purposes at the time of the study. However, in total, 259 films were scored. Of these 259 films 11 cases had Paget's disease of the sacrum and/or lumbar spine only and were therefore discarded. In 10 cases Paget's disease could not be confirmed and four controls were thought to have Paget's disease. As a result of these changes we included 87 matched pairs and a total of 248 films (137 without Paget's, 89 with unilateral and 22 with bilateral disease). Of the 248 films 98 were plain abdominal films, 53 pelvic films, 44 IVUs and 17 barium enemas (36 were "unrecorded"). No differences were found in either the occurrence of Paget's disease or the number of severe (grade 3+) cases of OA by film type.

The mean age of the matched cases and controls was 77.9 years and 77.6 years respectively, with 50 men and 37 women in each group. The mean age of all the cases and controls was 78.4 years and 77.4 years respectively with 66/45 male/female cases and 78/59 male/female controls.

Comparisons were made for matched cases and for the whole group. As the overall findings were the same, only the results for the whole group are presented. We compared unilateral disease with the contralateral hip and any disease (that is, any hip joint with juxta-articular Paget's disease) with any unaffected hip. Additionally cases involving the femoral head were analysed separately.

### UNILATERAL DISEASE MATCHED TO CONTRALATERAL SIDE

Unilateral disease was found in 85 cases and 83 contralateral hips were available for comparison. Data were unavailable if the full extent of the hip joint was not visible, or if an arthroplasty had been performed. Comparison showed a significant difference in minimum joint space between the two sides (median MJS for both ipsilateral and contralateral hip, 3 mm: Mann-Whitney U, $Z = -4.88$, $p = 0.00001$). The median Kellgren and Lawrence grade for both sides was 0 with no statistical difference between them (Mann-Whitney U, $p = 0.55$).

### ANY AFFECTED HIP COMPARED WITH ALL UNAFFECTED HIPS

A total of 129 affected hips were available for comparison with 352 unaffected hips. Data were unavailable from 15 hips for reasons given above. Median minimum joint space for the affected hip was 3 mm (range, 0–5 mm) and for the unaffected hip 4 mm (range, 0–6 mm, Mann-Whitney U, $p = 0.00001$; see fig 1). Median Kellgren and Lawrence grade for both groups was 0, with no statistical difference between the groups (Mann-Whitney U, $p=0.74$; see fig 2). In terms of severity of osteoarthritis, there were 19 instances of grades 2+ in the unaffected hips, and only five in the affected hips.

#### *Femoral head involvement*

Cases with femoral head involvement were compared with cases of isolated pelvic Paget's disease. We found 11 cases of femoral head involvement alone, and 21 cases of Paget's disease in both femoral head and pelvis. These cases were compared with 101 cases not

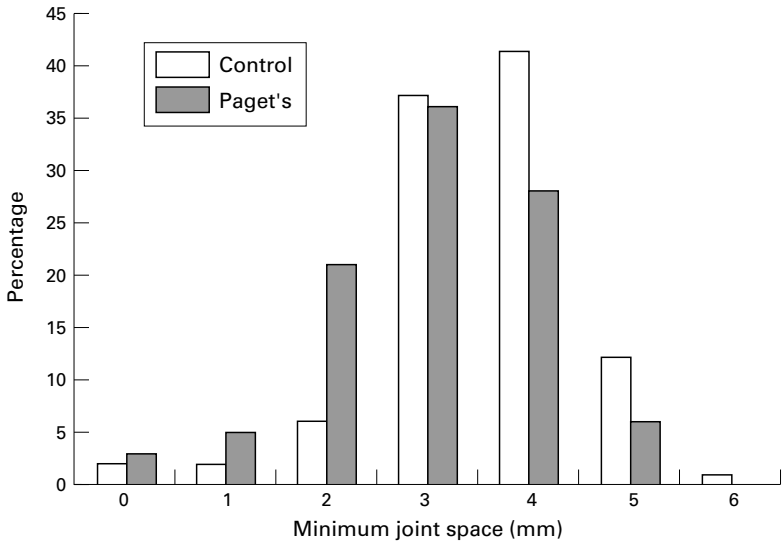


*Figure 1 Histogram of minimum joint space for Pagetic and control joints.*

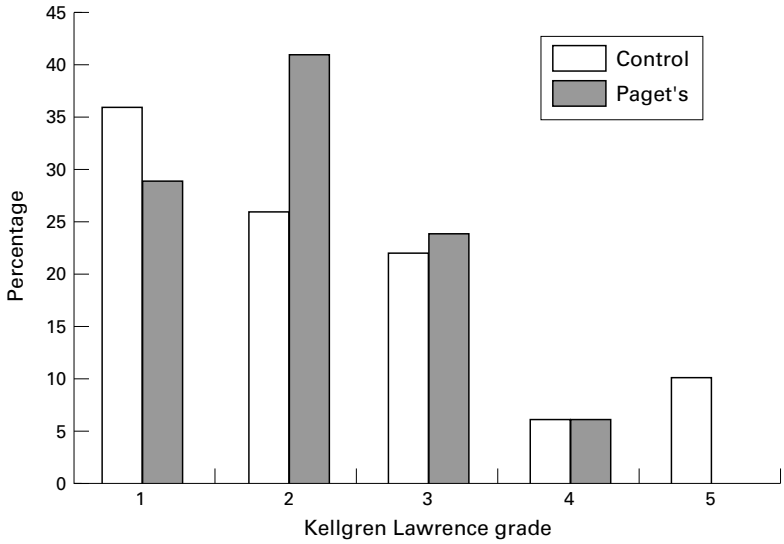


*Figure 2 Histogram of Kellgren and Lawrence scores for Pagetic and control joints. Note joints scoring grade 0 (Pagetic, n = 112; control, n = 304) have been omitted.*

involving the femoral head. No difference in minimum joint space was found between the groups (Mann-Whitney U, p=0.81) but a highly significant difference in Kellgren and Lawrence grades was found (Mann-Whitney U, p=0.0015). However, this difference was based on very small numbers: only one case of isolated pelvic Paget's had a Kellgren and Lawrence grade over 2, whereas four cases of femoral Paget's exceeded this grade.

## Discussion

In contrast with previous studies we have found no increase in the prevalence of radiological osteoarthritis of the hip joint associated with juxta-articular Paget's disease, with the possible exception of cases with femoral head involvement. The major difference between this and previous studies was in the method of case ascertainment. In this study it was assumed that the films were obtained because of reasons other than persistent musculoskeletal pain (the few pelvic radiographs were obtained because of trauma) and that the Paget's disease was largely undiagnosed. Previous studies may have found an increase in the prevalence of osteoarthritis because only symptomatic people were studied.

A further explanation may be given for the observed lack of association between Paget's disease and radiological osteoarthritis. Paget's disease may obscure or inhibit the radiological features of osteoarthritis. Certainly some of the radiological characteristics of Paget's disease such as the increase in bone density, disorganisation of bone architecture and cortical thickening might obscure such features as subchondral sclerosis and cyst formation, and this would lead to a relative under-diagnosis of grades 3–4 osteoarthritis. It is also possible that Paget's disease affects the skeletal response in osteoarthritis such as remodelling and that new bone formation—osteophytosis—is inhibited; this might influence grades 1–4 of the Kellgren and Lawrence classification system. However, this clearly was not the case in previous surveys of largely symptomatic populations where observers had no difficulty identifying the various radiographic features of osteoarthritis.[2 5]

Can we conclude, therefore, that pelvic Paget's disease is not associated with the development of osteoarthritis of the hip? Although osteoarthritis was not associated, we have found a significant reduction in joint space in cases of juxta-articular Paget's disease. The method of measuring joint space in this study was simple and of limited precision. However, as the measurements are comparative rather than absolute, and intraobserver reproducibility was good, this was not considered to be a source of significant error. Our definition of osteoarthritis did not permit us to label joint space narrowing alone as "osteoarthritic": this position was adopted as this isolated feature may be caused by other disorders, including inflammatory arthritides. In this study it was felt that isolated joint space narrowing was abnormal but, in the absence of osteophytes, this could not be called osteoarthritis.

The reduction in joint space may represent a specific Pagetic coxopathy—as suggested originally by Guyer and Dewbury.[6] Is this reduction in joint space a reflection of a secondary chondropathy associated with this disease? The evidence for a Pagetic chondropathy is scanty, although histological studies have shown direct infiltration of the articular cartilage by Pagetic tissue.[7] Alternatively the chondropathy may be secondary to changes in vascularity because of the underlying Paget's disease—a form of steal syndrome—and mechanical aberrations whereby increased stiffness of the subchondral bone and joint incongruity expose the cartilage to undue mechanical stresses. In addition, the succeeding development of osteoarthritis may depend on other individual predisposing factors such as a generalised (and presumably inherited) tendency to develop osteoarthritis. This relation was suggested by our previous survey that identified coexisting osteoarthritis at other anatomical sites as a risk factor for osteoarthritis at the Pagetic joint.[2]

Other theories explaining the association between Paget's disease and osteoarthritis are less relevant to pelvic Paget's disease—notably the altered joint mechanics as a result of

changes in shape of associated long bones. In terms of Pagetic coxopathy mechanical factors might be expected to be more apparent in cases where the femoral head is involved and, although the numbers involved were small, this study supports this theory.

In summary we have found a significant loss of joint space in hips affected by juxta-articular Paget's disease but no increase in associated radiological osteoarthritis. Although joint failure may result from this secondary chondropathy progression may be dependent on non-Pagetic factors. It is also possible that the usual radiological features of osteoarthritis may be modified or obscured by the Paget's disease.

We wish to thank Professor Cyrus Cooper and staff at the MRC Environmental Epidemiology Unit, University of Southampton for providing matched cases and controls from their original survey. We would also like to thank the radiology departments of the NHS Trust hospitals in Carlisle, Lancaster, Newcastle upon Tyne, Preston, Wigan and Warrington for their kind cooperation with this study.

1 Kanis JA. *Pathophysiology and treatment of Paget's disease of bone*. London: Martin Dunitz, 1991.
2 Helliwell PS. Osteoarthritis and Paget's disease. Br J Rheumatol 1995;34:1061–3.
3 Cooper C, Schafheutle K, Dennison E, Kellingray S, Guyer P, Barker D. The epidemiology of Paget's disease in Britain: is the prevalence decreasing? J Bone Miner Res 1999;14: 192–7.
4 Croft, Cooper C, Wickham C, Coggon D. Defining osteoarthritis of the hip for epidemiologic studies. Am J Epidemiol 1990;132:514–22.
5 Graham J, Harris WH. Paget's disease involving the hip joint. J Bone Joint Surg 1971;53B:650–9.
6 Guyer PB, Dewbury KC. The hip joint in Paget's disease (Paget's coxopathy). Br J Radiol 1978;51:574–8.
7 Hadjipavlou A, Lander P, Srolovitz H. Pagetic arthritis. Clin Orthop Rel Res 1986;208:15–19.