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■ CASE REPORT

Resurfacing arthroplasty of the hip in osteopetrosis

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A 34-year-old woman with a benign form of osteopetrosis developed osteoarthritis of the hip. In order to avoid the difficulties associated with inserting the femoral component of a conventional total hip arthroplasty, a hybrid metal-on-metal resurfacing was performed. There were several technical challenges associated with the procedure, including the sizing of the component, press-fit fixation of the acetabular component and femoral head preparation, as well as trying to avoid a fracture. No surgical complication occurred. After more than a year following surgery, the patient showed excellent clinical function and remained satisfied with the outcome. We conclude that the hybrid metal-on-metal resurfacing arthroplasty represents a valuable option for the treatment of patients with osteopetrosis and secondary hip osteoarthritis.

Osteopetrosis is a rare disease, characterised by increased bone mass as a result of osteoclast dysfunction.[1] If we compare the dominant (benign) and recessive (malignant) types, the former is more clinically challenging as these patients have a normal life expectancy, despite their brittle bones.[1-3]

Some authors refer to this disorder as a syndrome because of associated features such as osteomyelitis of the jaw, cranial nerve palsy (optic, trigeminal and auditory), back pain and spondylolisthesis.[2,3] A fracture is the most common orthopaedic complication seen in osteopetrosis. The osteopetrotic bone consolidates with pathological callus without Haversian organisation, even up to one year post-injury.[4] Femoral neck and subtrochanteric fractures have been reported,[3] frequently leading to coxa vara,[5,6] which may contribute to the degenerative process and lead to secondary osteoarthritis.[7-9] The hard nature of the subchondral bone and its susceptibility to microfractures may also play a part in the development of degenerative osteoarthritis.

Few attempts at total hip arthroplasty (THA) in osteopetrosis have been reported,[7,9-11] and most of these cases are secondary to femoral fractures resulting in degenerative osteoarthritis.[7-9] A THA is technically challenging in osteopetrotic bone and the main difficulty is encountered during femoral canal preparation.[8,10] Improvements in metal-on-metal bearing surfaces, such as the modern surface replacement, have shown encouraging early clinical results.[12] The absence of femoral canal preparation at surface replacement makes this procedure an attractive alternative in patients with osteopetrosis. This paper reports the successful use of a hybrid metal-on-metal resurfacing arthroplasty of the hip in osteopetrosis.

Case report

In 2004, a 34-year-old woman with type-II benign osteopetrosis, according to the classification of Bollerslev and Mosekilde,[2] presented with a two-year history of pain and stiffness in the left hip. At the age of 28, she had sustained a right femoral subtrochanteric stress fracture which was treated by a femoral plate. Since the age of 15 years, she had suffered recurrent episodes of septic arthritis of the left sacro-iliac joint. In 2002, sepsis spread to the left hip joint. This was treated by an open surgical lavage and debridement, via a posterior approach and a ten-week course of intravenous antibiotics. The cultures grew *Staphylococcus aureus*, which was sensitive to penicillin. The treatment was successful, with no recurrence of infection.

In 2004, she developed rapidly-progressing pain in the left groin and became confined to a wheelchair or crutches. Difficulty with weight-bearing led her to give up work. Clinical examination revealed a painful left hip with diminished movement. Blood tests revealed a normal C-reactive protein and normal erythrocyte sedimentation rate. Technetium and gallium bone scans showed that there was no persisting

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Fig. 1

Pre-operative anteroposterior radiograph of the left hip, showing the characteristic signs of osteopetrotic bone, with osteoarthritis and coxa valga of the hip.

infection. Radiographs of the left hip demonstrated severe osteoarthritic changes, coxa valga, significant acetabular erosion and proximal migration of the femoral head (Fig. 1). There was concentric thinning of the joint space, without signs of avascular necrosis, the density of the femur was markedly increased and the medullary canal was not visible.

We concluded that the severe groin pain had developed because of degenerative osteoarthritis of the hip, secondary to septic arthritis. Following a discussion with the patient, we decided to resurface the left hip joint.

The operation was performed through a posterolateral approach. There was increased anteversion of the femoral neck. There was no femoral head articular cartilage and no deformation or collapse characteristic of avascular necrosis. The diameter of the femoral head was small (44 mm), while the diameter of the femoral head/neck junction was 42 mm, resulting in a reduced head/neck diameter ratio. The acetabulum was exposed and prepared before inserting the femoral component (Birmingham Hip Resurfacing (BHR), Midland Medical Technologies Ltd, Birmingham, United Kingdom). The acetabulum showed marked erosions and had a larger diameter than the femoral head. It was reamed sequentially, up to 53 mm, with standard reamers. The acetabular bone was sclerotic, although surface bleeding was visible after reaming. The stability of the trial acetabular component was not satisfactory and approximately 20% of its superior area was unsupported by bone when appropriately orientated (20° anteversion, 45° abduction). A 54 mm dysplasia acetabular component (Midland Medical Technologies Ltd) with 1 mm of press-fit and supplemental screw fixation was selected to improve stability. The original screws (BHR dysplasia screw, Midland Medical Technologies Ltd) had small threads with a large core diameter, so could not engage the osteopetrotic acetabular bone. As a result, the bone was tapped and 4.5 mm AO cortical screws (Zimmer, Warsaw, Indiana) were used to obtain good primary fixation.

Because the acetabulum was larger than the femoral head (femoro-acetabular mismatch), minimal reaming of the femoral head was performed (approximately 2 mm). The bone was extremely dense and difficult to ream, similar to the 'marble-top table' described by Ashby.[9] Multiple holes were drilled in the femoral head to increase cement penetration and the area of fixation. No cysts were noted. A 46 mm femoral component was implanted with antibiotic-impregnated bone cement (Antibiotic Simplex, Howmedica, Allendale, New Jersey). The range of movement was assessed, in order to evaluate if the increased anteversion of the femoral neck led to posterior femoro-acetabular impingement. A reasonable arc of movement was established. Peri-operative cultures were negative. Histopathological examination confirmed the diagnosis of osteopetrosis and also revealed a granulomatous reaction in the soft tissues, with abnormal bone remodelling. There was no sign of chronic infection or inflammation post-operatively.

Weight-bearing was not permitted during the first 12 weeks post-operatively and only partial weight-bearing was recommended for the following six weeks. She had returned to her pre-operative functional level and to her work six months after surgery. At the 18-month follow-up, she had a Harris hip score[13] of 96 points. Physical examination revealed a good range of movement without leg-length inequality. There was limited flexion because of scarring from the septic arthritis. She was able to walk for more than one hour and was very satisfied with the clinical outcome. Radiographs of the hip demonstrated a good component position with no fracture or signs of migration (Fig. 2).

Discussion

Of the few reported cases of THA in osteopetrosis, the majority were performed for degenerative osteoarthritis associated with secondary coxa vara, developing after multiple microfractures of the femoral neck or malunion of subtrochanteric fractures. Degenerative arthritis of the hip has also been reported in patients with osteopetrosis, without an anatomical deformity.[7] The pathophysiology of

Fig. 2

Anteroposterior radiograph of the left hip at 18 months post-operatively, showing a stable acetabular component.

degenerative arthritis in these patients remains unclear, but it seems to be caused by a lack of resilience of the subchondral bone. The articular cartilage is compressed between the bone of increased density within the femoral head and the acetabulum.[14,15] Microfractures of the subchondral bone occur, resulting in increased stiffness and decreased shock-absorbing capacity as well as secondary cartilage degeneration.[8,14,16]

Technical difficulties have been reported during THA in patients with osteopetrosis, especially on the femoral side,[9-11] causing surgeons to be more aware of the hard and brittle nature of the bone and the lack of a femoral medullary canal.[7,9,10] There have been studies in which surgeons used a power reamer guided by fluoroscopy in order to cannulate the occluded femoral canal,[7,10,11,17] while some surgeons failed in their attempts and had to shorten the femoral component.[7] Janecki and Nelson[10] reported fractures of the lesser trochanter occurring while reaming the proximal femur. Matsuno and Katayama[18] and Strickland and Berry[11] recommended the use of a cemented femoral component in order to avoid fracture, and Feldman et al[8] reported fissuring of the acetabulum during the introduction of acetabular screws.

With the success of modern metal-on-metal THAs,[12,19] there has been a resurgence of interest in resurfacing arthroplasty of the hip. The new generation of hip resurfacing prostheses appear to match the survivorship of THA in the short term.[20] In this case study, radiographs of the femur showed dense bone without a femoral medullary canal. In addition, the increased valgus angle of the proximal femur offered better biomechanical loading conditions, with greater compressive stresses under the femoral component. This favoured long-term fixation and lower tensile stresses on the lateral femoral neck, decreasing the chance of a fracture.[21] Metal-on-metal resurfacing arthroplasty represented an attractive option to avoid the difficulties associated with the creation of a medullary canal.[8]

We suggest cement fixation in order to fill the gaps between the femoral component and femoral bone and to avoid relying on bone ingrowth for long-term fixation of the component. This was especially important in our patient, as the blood supply to the femoral head was disrupted during surgical lavage and debridement for previous septic arthritis and/or during the resurfacing arthroplasty procedure,[22] thus reducing the potential for bone remodelling of the femoral head. Another advantage of using a cemented femoral component is the presence of antibiotic-impregnated cement. Because cement penetration is impossible in osteopetrotic bone, we recommend that the femoral head is drilled with multiple holes before implantation of the femoral component.

Modern resurfacing arthroplasty of the hip is only available with uncemented acetabular fixation. We believe that bone ingrowth can be expected in osteopetrotic patients with cementless components if primary stability of the acetabular component is achieved.[17] Feldman et al[8] had to convert a cementless acetabular component to a cemented one because a fracture of the acetabulum occurred between two screw holes during implantation. Care should, therefore, be taken when using screws and, as with our case, a different type of screw may be needed.

Resurfacing arthroplasty of the hip is a valuable alternative in young patients with osteopetrosis. The difficulties encountered during THA in similar cases seem to be minimised. However, it is of note that osteopetrotic patients are at risk of femoral neck microfractures with progressive varus deformation. If loosening of the femoral component or fracture of the neck occurs, revision to a conventional stem-type cemented THA with a large femoral head matching the acetabular component is still possible. Resurfacing arthroplasty of the hip is a challenging procedure and attempts at resurfacing in patients with osteopetrosis should only be performed by surgeons who are experienced with the technique.

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