Bilateral hip arthritis in a case of renal osteodystrophy

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SUMMARY

Chronic renal disease is often associated with secondary hyperparathyroidism (HPP) and rarely with tertiary HPP. Hip arthritis with protrusio acetabuli, secondary to tertiary HPP, is a rare case scenario and has not been described well in the literature. We present a rare case of bilateral hip arthritis with protrusio acetabuli secondary to renal osteodystrophy due to tertiary HPP. The diagnosis and aetiology of hip arthritis and its treatment have been discussed along with a detailed review of literature of skeletal lesions due to HPP.

BACKGROUND

This is uncommon presentation of a well-known condition and has not been described in the literature.

CASE PRESENTATION

We present a case of a 30-year-old man (Kenyan) who was born at term normally. He attained his developmental milestones in time and achieved adulthood normally and worked as a full-time cook. He took normal healthy diet and denies ever smoking, taking alcohol or misusing any other form of drug.

In 2001, he started developing kidney problems and hypertension for which he was evaluated and found to have renal failure due to an idiopathic cause. He was put on dialysis for 4 years after which three renal transplants were performed but all failed and the physician concluded that the cause of the kidney disease may be due to lupus nephritis or idiopathic thrombosis of renal vessels. But we could not find any obvious evidence of any specific reason for these transplants rejection. He was put on triple drug immune suppression during and after three renal transplants over 4 years. These drugs included tacrolimus, cyclosporine, azathioprine and steroids. Currently, he undergoes regular dialysis thrice a week and takes 5 mg prednisolone per day.

Four years ago, his parents and peers noticed a mild deformity in upper spine and change in posture which was a gradual stooping. The deformity was not associated with any pain and gradually increased to an extent that he noticed a significant loss in height which decreased from a previous height of 163–147 cm and a severe hump-like deformity of the back ([figure 1](#)) which led to the development of severe respiratory symptoms, and he currently experiences breathlessness even on normal conversation. Three years ago, he started experiencing bony pains in his upper spine which was mild to start with and localised but gradually it spread through the body. The pain was increased with activity and was relieved with rest and

Figure 1 The clinical picture of the patient showing severe dorsal kyphosis.

massage. Since the past 5 months, he developed pain and a limp in his right hip region and he also noted gradual loss of movements in both hips. Currently, he uses crutches or a walking frame for ambulation.

On examination, the patient was of a short stature with a height of 147 cm and had obvious Kyphosis in the upper back ([figure 1](#)). Both the hips had reduced passive and active movements, and mild crepitus were elicited on the right hip. The right lower limb was shorter than the left by 1.5 cm. He had a surgical scar in the anterior neck postsurgical parathyroidectomy.

INVESTIGATIONS

On other evaluation and investigations, he was found to have tertiary HPP secondary to parathyroid hyperplasia. This was confirmed by a parathyroid scan ([figure 2](#)) and very high parathyroid hormone (PTH) levels of 3007 pg/mL (15–65); serum alkaline phosphatase 1379 µ/L (39–117); serum calcium 9.9 mg/dL and vitamin D levels 24.2 (20–32 pg). His T score of the hip on dual emission X-ray absorptiometry (DEXA) scan was –1.9, confirming moderate osteopenia.

He was referred to us because of the hip pains (R>L). A pelvic X-ray ([figure 3](#)) was performed, which showed severe osteoporosis, erosion of the

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Figure 2 Parathyroid scans showing increase in uptake in the region of the left parathyroid consistent with adenoma.

cortical bone with migration of the acetabulum and femoral head medially into the pelvis (protrusio acetabuli) with coxa vara ($R > L$). MRI of the hips (figures 4 and 5) revealed that there was medial displacement of the medial wall of the acetabulum with respect to ischial line on both sides ($R > L$), suggestive of protrusion acetabuli. Irregularity of outline of the femoral heads and the subchondral region was found. The articular cartilage showed irregular and patchy areas of thinning, suggestive of osteoarthritis. Altered heterogeneous marrow signal intensities were seen in femora and pelvic bones suggestive of post-transplant marrow changes. Fatty marrow changes were also noted within the head of the femur and the greater trochanter on both sides. An X-ray of the dorsal spine (figures 6 and 7) confirmed severe kyphosis with multiple wedging of the vertebrae which had also become biconcave. An X-ray of the skull showed classic salt and pepper appearance (figure 8) and the phalanges of hand had subperiosteal resorption (figure 9).

DIFFERENTIAL DIAGNOSIS

- Inflammatory arthritis (eg, rheumatoid arthritis and ankylosing spondylitis).
- Metabolic diseases (eg, Paget's disease).

Figure 3 X-ray of the pelvis anteroposterior showing erosion of acetabuli and sacro iliac joints, with migration of head of the both femur ($R > L$) into the pelvis (protrusio acetabuli).

Figure 4 Multiplanar MRI of both hips obtained using a T1-weighted, T2-weighted and PD-weighted, SE, TSE and FFE sequences. Both femoral heads are in the acetabulum but there is displacement of the medial wall of the acetabulum into the pelvis ($R > L$).

TREATMENT

The patient underwent subtotal left parathyroidectomy.

OUTCOME AND FOLLOW-UP

After parathyroidectomy, the patient reported reduced pain and the serum PTH levels became normal; however, the hip X-rays showed similar changes. Owing to persistent pain in the hips, especially on the right side, the patient was advised to undergo total hip arthroplasty if the problem persisted, but due to financial constraints the surgery could not be performed at present.

DISCUSSION

HPP may be (1) *primary*, where one or more parathyroid gland produces excess PTH, (2) *secondary*, where there is increased

Figure 5 MRI of the sagittal sections showing the irregularities of outline of both femoral heads with subchondral irregularities and conical appearances. There is also widening of both joint spaces. The articular cartilage is irregular with thin patchy areas.

Figure 6 X-ray of the spine lateral and anteroposterior view showing kyphosis with vertebral collapse at the thoracic spine.

secretion of PTH in response to low calcium because of kidney, liver or bowel disease and (3) *tertiary*, when there is autonomous secretion of PTH usually because of chronic renal failure and that parathyroid gland behaves as neoplasm.¹ In our case, the left parathyroid gland became hyperplastic (tertiary HPP)

Figure 7 X-ray of the spine lateral and anteroposterior view showing kyphosis with vertebral collapse at the thoracic spine.

Figure 8 Skull X-ray lateral showing calcium crystal deposit in the frontal parietal bone (salt and pepper appearance) with osteolytic lesions of the frontal bone.

after sustained hyperactivity of the gland due to chronic renal failure (secondary HPP). To the best of our knowledge, no case has yet been reported of bilateral hip arthritis following ROD and tertiary HPP. The incidence of hip arthritis following HPP is quite low² and has been reported once in the USA where it was found to occur in about 5/100 000 patients, hence this was a rare case in our setup also.³

Bone pains are a common manifestation of ROD. It usually presents with pain in the bones, joints and the spinal column weakness⁴ as the condition progresses, as seen in this case, with the development of kyphosis. The loss of calcium from bones also causes pain in the back, wrists and legs. There is also 'osteogenic synovitis' with features of bone collapse and secondary osteoarthritis. Hip arthritis is one of the rarer presentations of

Figure 9 X-ray of the hands showing subperiosteal resorption and subchondral bone erosions.

patients with HPP. Protrusio acetabuli hip deformity⁴ occurs where the medial wall of the acetabulum invades the pelvic cavity with associated displacement of the femoral head, as seen in this case; bilateral protrusio acetabuli is a rare form of presentation of hip arthritis. It may involve one of the hip joints but it is mainly bilateral. It was first described in 1824 by a German pathologist (Adolph William Otto) in a woman's skeleton in a natural museum,⁵ where the right acetabula protruded in the pelvis like half an orange. The main characteristic was the insertion of the femoral head deeply in the acetabulum with imperfect ankylosis. Protrusio acetabuli can be primary⁶ where the acetabulum is congenitally too deep resulting in a thin acetabular floor. This floor does not withstand the pressure exerted on it by the femoral head leading to the protrusio acetabuli effect. This form of protrusio acetabuli is rare and is mainly bilateral and is seen in women.

Secondary protrusio acetabuli is also rare, and was first described in 1854 in a 40-year-old woman who had metastatic carcinoma of the breast.⁶ Although still rare, the condition has been described in various conditions similar to our case of tertiary HPP. Other causes of this condition (box 1) are primary idiopathic, secondary neoplastic, infective, metabolic, inflammatory, traumatic and genetic disorders.⁷ Among these, those due to metabolic cause, such as osteogenesis imperfecta, osteoporosis, HPP and Paget's disease, are also reported (box 1). Infections in which protrusio acetabuli has been described are: tuberculosis (first described in 1903), gonococcus arthritis,⁸ echinococcus cysts⁹ and *Staphylococcus* and *Streptococcus* infection.

Box 1 Causes of protrusio acetabuli

- A. Primary:
 1. Idiopathic
 2. Familial
- B. Secondary:
 1. Inflammatory
 - Rheumatoid arthritis
 - Ankylosing spondylitis
 - Psoriatic arthropathy
 2. Metabolic
 - Vitamin D deficiency (Osteomalacia)
 - Paget's disease
 3. Traumatic
 - Central fracture dislocation of hip
 4. Infective
 - Tuberculosis
 - Septic arthritis
 5. Degenerative
 - Osteoarthritis
 6. Bone tumours
 - Fibrous dysplasia
 - Malignant tumours
 7. Iatrogenic
 - Hip replacement
 8. Miscellaneous
 - Ehlers Danlos Syndrome
 - Marfan's Syndrome
 - Sickle Cell Disease
 - Osteogenesis imperfecta

Box 2 Bony changes in tertiary hyperparathyroidism

- ▶ Subperiosteal bone resorption (especially in phalanges)
- ▶ Osteoporosis
- ▶ Osteosclerosis (eg, rugger-jersey spine)
- ▶ Soft tissue calcification (eg, salt and pepper appearance of skull)
- ▶ Generalised increased uptake on Tc-99m pertechnetate bone scan (focal uptake with adenoma)
- ▶ Superior and inferior rib notching
- ▶ Pseudo (stress) fractures

The condition has been described in rheumatoid arthritis where steroid therapy has been shown to cause exacerbation of the condition.⁷ Also to add on the lists are genetic disorders such as trisomy 18, Ehlers-Danlos syndrome and sickle cell disease.¹⁰ Bilateral hip protrusio is relatively uncommon and one of the most common causes of it is metabolic (osteomalacia), apart from inflammatory arthritides.

X-ray findings (box 2) in secondary (and tertiary) HPP are often associated with the osteosclerosis of renal osteodystrophy and the osteomalacia of vitamin D deficiency.¹¹ The biochemical abnormalities that may precede radiological change are also well recognised. In this case, the classical X-ray changes of tertiary HPP were present (figures 3, 6–9). Here, calcification occurs in soft tissues and it helps to differentiate this condition from rheumatoid arthritis.^{6 8 11}

MRI had been used to evaluate early musculoskeletal pain in patients who have had kidney transplant¹² and it has been found that avascular necrosis of the head of the femur was the commonest cause of the hip pain.¹³ In our case, the cause of the hip pain was due to osteoarthritis, and associated protrusio acetabuli (figures 7 and 8), in which there was irregularity of outline of the femoral heads and the subchondral region. However, there was no evidence of avascular necrosis. Although MRI has been used to detect osteonecrosis at an early stage in patients who have had kidney transplant,¹⁴ other causes of hip pain such as osteoarthritis and protrusio acetabuli should be considered as in this case and proper treatment be given.

Nishi and Gejvo¹⁵ observed that after renal transplantation three major problems commonly occur, that is, (1) osteoporosis, (2) renal osteodystrophy and (3) osteoarthritis derived from haemodialysis-associated amyloidosis. Osteoporosis is usually inevitable due to prolonged steroid administration in these cases, similar to ours. They further remarked that, in patients with tertiary HPP, renal osteodystrophy progresses even though kidney grafts have good renal function, and therefore one must be concerned about the bone health of the grafted patients after kidney transplantation.

The guidelines for the treatment of hip arthritis in a HPP have not been discussed in the literature adequately. In a case of hip arthritis with protrusio acetabuli secondary to Marfan's syndrome, total hip arthroplasty (THA) was found to be beneficial to relieve the intractable pain.⁸ We believe that the hip pain reduces when the hyperplastic parathyroid gland is removed as seen in this case.¹⁶ However, if the hip arthritis is severe and if associated with significant protrusio, as seen in our case, should justify THA. During THA, in acetabular protrusion, reconstruction of the contained cavitory defect is required, using morsellised bone grafts. Ries¹⁷ and McBride

*et al*¹⁸ remarked that the central acetabular defect can be effectively treated with a lateralised cementless porous-coated component supported on the peripheral acetabular bony rim. Ries concluded that the results of THA with medial acetabular bone grafting for protrusio acetabuli have been favourable, with success rates similar to conventional THA¹⁷ and a predictable long-term solution.¹⁸

According to Tiponez *et al*,¹⁶ the indications for parathyroidectomy in patients with HPP after kidney transplantation (3HPT) include persistent severe hypercalcaemia (defined as a serum calcium level greater than 11.5 mg/dL), unexplained renal function deterioration or progressive bone mineral density loss. They concluded that the best surgical approach for patients with 3HPT is subtotal or total parathyroidectomy with autotransplantation, as is in this case.

Learning points

- ▶ Patients with chronic renal disease may develop tertiary hyperparathyroidism (HPP) and renal osteodystrophy, so if they have bony pains then they need regular screening by assaying for serum levels of calcium and parathyroid hormone (PTH).
- ▶ Subtotal parathyroidectomy helps to reduce musculoskeletal pain associated with tertiary HPP.
- ▶ Musculoskeletal pain, especially of the hip joint pain, can be caused by secondary osteoarthritis and protrusio acetabuli, other than avascular necrosis, which is the most common cause of hip arthritis in patients with HPP.
- ▶ MRI is a useful tool in the evaluation of musculoskeletal pain associated with HPP, especially in early disease.
- ▶ Total hip arthroplasty is the treatment of choice in the management of end-stage secondary osteoarthritis with protrusio acetabuli in patients with tertiary HPP.

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