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CASE REPORT

Pathological hangman's fracture after successful renal transplantation

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Renal osteodystrophy and post renal transplantation bone disease are increasingly common causes of pathological fracture. The authors present the first case of a pathological hangman's fracture in a patient after successful renal transplantation.
An anterior instrumented C2-C4 fusion was performed.

Keywords : vertebra ; C2 ; axis ; hangman's fracture ; renal osteodystrophy ; renal transplant.

INTRODUCTION

Renal transplantation is an increasingly common treatment modality in End Stage Renal Failure (ESRF) and is associated with post-transplantation bone disease on top of the already existing pre-transplantation bone disease, which is principally Renal Osteodystrophy (ROD) (*2*). ROD is a broad term encompassing all osseous disorders that arise due to renal insufficiency and is multifactorial in aetiology. The incidence of ROD and post transplantation bone disease is on the increase, mirroring the increased life expectancy of patients with long standing ESRF, due to advances in both haemodialysis and renal transplantation (*8*). Renal transplantation corrects many of the metabolic disturbances that cause ROD, but is still associated with ongoing bone disease (*5*). The authors describe a pathological hangman's fracture due to renal bone disease in a patient after successful renal transplantation.

CASE REPORT

A 55-year-old male was referred to the authors' spine clinic with a two-week history of neck pain. There was no trauma or systemic upset. Examination revealed point tenderness over C2 without neurological disturbance. The medical history was complicated. There was a chronic renal failure of unknown aetiology, treated with haemodialysis for a period of 26 years, prior to receiving a renal transplant. The renal transplant had been functioning well for over 10 years and the patient remained on cyclophosphamide. He had sustained osteoporotic fractures to his ribs and left femoral neck, 29 years previously. Osteoporosis had been diagnosed by bone mineral density testing, and treatment had been commenced with good effect. The patient was also known to have dialysis related amyloidosis (DRA) ; a serum amyloid protein (SAP) scan performed during the previous work up for the pathological fractures revealed deposits in the distal radius bilaterally. Plain cervical spine

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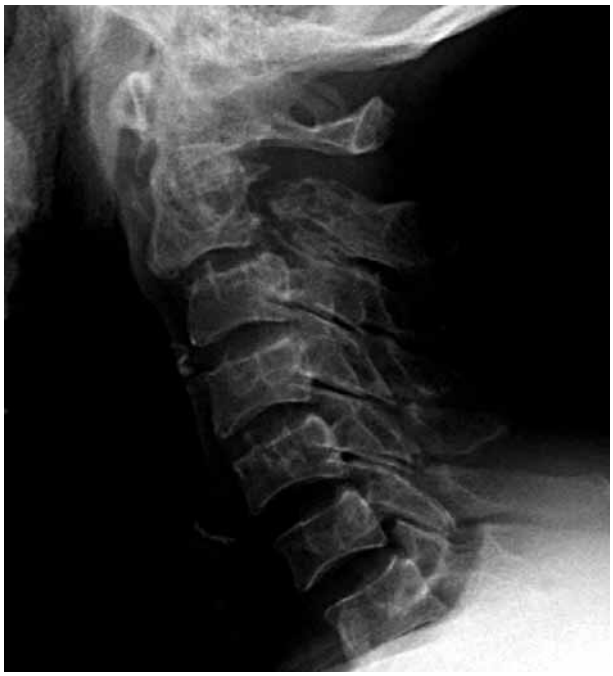

Fig. 1. — Lateral radiograph of the cervical spine, clearly showing the hangman's fracture.

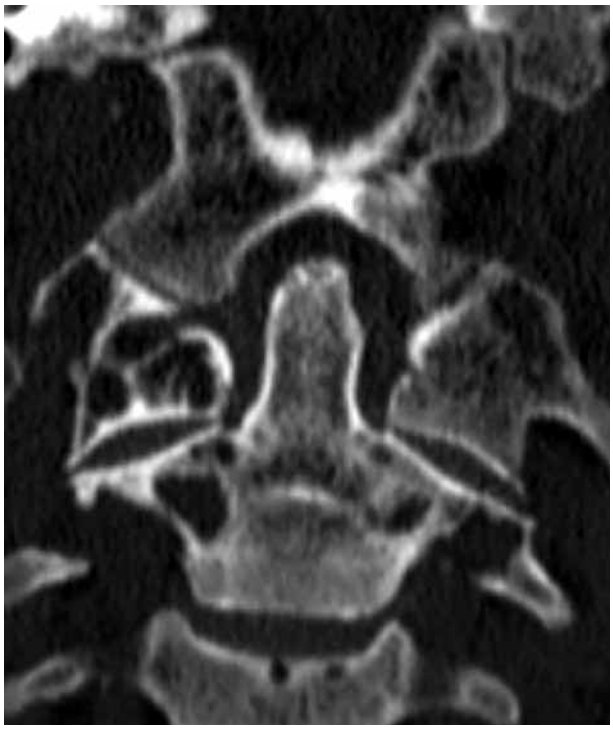

Fig. 2. — Coronal CT-scan revealing multiple C1 and C2 subarticular cystic lesions.

radiographs, requested by the referring physician at the time of this admission, revealed a hangman's fracture (fig 1).

The cervical spine was immobilised with a Miami-J collar. A CT-scan of the cervical spine revealed multiple lytic lesions (fig 2). The differential diagnosis for this fracture at that time included spondylodiscitis, myeloma, metastases, osteoporosis, osteomalacia, dialysis related amyloidosis and secondary hyperparathyroidism with multiple brown tumours. The biochemistry was normal (see below). The MRI was non-diagnostic and the bone scan was normal. The patient's renal function remained normal throughout.

Preoperatively, without histopathological confirmation, DRA was felt to be the most likely diagnosis. Spondylodiscitis was excluded as the patient was systemically well and had normal inflammatory markers. Myeloma was excluded by negative plasma electrophoresis, normal immunoglobulin levels, negative Bence Jones proteins and a negative iliac bone marrow biopsy. Metastases were felt unlikely as the patient had no red flag symptoms, while CT-scans of abdomen, pelvis and chest were normal. Osteoporosis and osteomalacia were not thought to be responsible as multiple cysts were not in keeping with either diagnosis, the patient's recent BMD scans had been normal and he had normal vitamin D3 levels. Secondary hyperparathyroidism was thought unlikely as the serum calcium, phosphate and parathyroid hormone levels were also within their normal range, while radiographs of the hands and feet which characteristically show subperiosteal bone resorption affecting the phalangeal tufts, radial aspect of the proximal and middle phalanges of the fingers and metacarpals, were normal.

The patient underwent anterior cervical arthrodesis and plating of C2-C4 (fig 3) ; he made an uncomplicated recovery. Bone samples taken during surgery were limited in order to conserve bone stock, and were non-diagnostic. It was still felt that systemic amyloidosis was the most likely diagnosis and the patient was subsequently referred

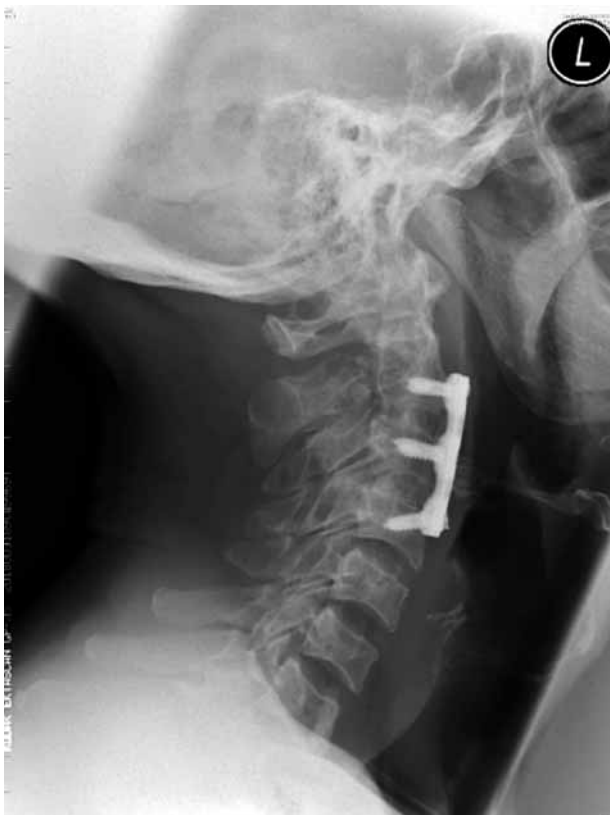

Fig. 3. — Lateral radiograph showing anterior cervical arthrodesis C2-C4.

to the National Amyloid Centre where a Serum Amyloid Protein scan was performed. This did not show any evidence of spinal amyloidosis. Review at six months showed the patient to be asymptomatic. Imaging showed no further progression of the cervical lesions. After discussion with the radiologists and the nephrology team it was felt that this patient's pathology was secondary to combined residual bone disease from his pre-transplantation renal osteodystrophy and ongoing post-transplantation bone disease.

DISCUSSION

The pathophysiology of ROD is complex. In patients on haemodialysis it is believed to arise due to a combination of decreased 1,25 vitamin D3 production by the kidneys, retention of substances excreted by the normal kidney (aluminium, amyloid, inorganic phosphates) and increased production plus decreased excretion of parathyroid hormone (*4, 4*). The changes induced range from those typical of osteomalacia to those of secondary hyperparathyroidism and spondyloarthropathy (ankylosing spondylitis, psoriatic arthritis and others). Spondyloarthropathy is associated with excess iron, aluminium and, most commonly, amyloid. Most patients, however, exhibit mixed patterns of disease. Successful renal transplantation largely restores the endocrine and exocrine renal function of patients with ESRF. This was previously thought to lead to a gradual improvement of the established renal osteodystrophy, but it is becoming increasingly recognised that even patients who have had successful renal transplants remain at risk for many of the problems seen under haemodialysis (*3, 6*). Furthermore, immunosuppressive drugs used after renal transplantation perpetuate the situation by promoting increased rates of bone turnover and/or promoting bone loss. Indeed, increase of bone loss and fracture incidence following renal transplantation is well documented (*1, 7*).

CONCLUSION

Reaching a definitive diagnosis for the cause of pathological vertebral fractures after successful renal transplantation can be difficult. Haemodialysis and renal transplantation are increasingly common treatment modalities and the patients receiving them are surviving longer than ever before. Each modality is associated with its own specific side effects. It is imperative that the attending physician and spinal surgeon make themselves familiar with these. Our case emphasises the importance of a multi-specialty approach to the investigation of these individuals and of the difficulties that can arise in making a definitive diagnosis.

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