

Simultaneous, Non-traumatic, Bilateral Neck Femur Fractures in Uremic Renal Osteodystrophy: A Report of Three Cases and Literature Review

Rakesh John¹, Prasoon Kumar¹, Sameer Aggarwal¹, Rajesh Kumar Rajnish¹, Saurabh Agarwal¹,
Kuldeep Vatsyan¹

Learning Point for this Article:

Chronic renal failure could lead to spontaneous non traumatic fractures of the femoral neck due to the associated metabolic abnormalities, and this requires high vigilance on the part of the healthcare providers, for prompt diagnosis and management.

Abstract

Introduction: Bilateral, simultaneous, non-traumatic, pathological femur neck fractures due to renal osteodystrophy (RO) in chronic renal failure cases are uncommon but are increasingly being reported in the literature. Seizure episodes due to uremic encephalopathy could lead to such fractures.

Case Reports: We describe three cases of patients with end-stage renal disease and RO, who presented with bilateral, non-traumatic femur neck fractures after episodes of grand mal con-vulsions. We also review the literature for all such similar cases and briefly discuss the pathophysiology of RO and the management of these pathological fractures.

Conclusion: Spontaneous, pathological, bilateral neck femur fractures are increasingly being reported in chronic renal failure patients with high morbidity and mortality. These fractures tend to be missed as the patients are generally immobilized and bed ridden due to the renal disease. Early diagnosis with high index of suspicion in patients with hip pain followed by supervised treatment with the involvement of a nephrologist is of uttermost importance.

Keywords: Bilateral neck femur fractures, pathological fracture, autosomal dominant poly-cystic kidney disease, renal osteodystrophy, grand mal convulsions.

Introduction:

Bilateral femur neck fractures are relatively uncommon and are usually secondary to seizure disorders, trauma, metabolic diseases, electric shock injuries, and osteoporosis.[1] Renal osteodystrophy (RO) secondary to chronic renal failure (CRF) is an established cause for pathological femur neck fractures [1]. However, bilateral pathological neck femur fractures in CRF are extremely uncommon [1]. We present three cases of simultaneous bilateral neck of femur (NOF) fractures with no history of trauma. The first case was an autosomal dominant

polycystic kidney disease (ADPKD) with end-stage renal disease (ESRD) and RO who presented with bilateral, simultaneous, non-traumatic, pathological femoral neck fractures after two episodes of seizures probably secondary to uremic encephalopathy/electrolyte imbalance. Till date, this is the first case reported in patients with ADPKD. The second case was a 15-year-old girl with congenital dysplastic kidney causing RO of 3 years duration, who also presented similarly with bilateral NOF fractures after an episode of seizure. The third case was a 64-year-old male with chronic kidney disease (CKD) due to

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Dr. Rakesh John



Dr. Prasoon Kumar



Dr. Sameer Aggarwal



Dr. Rajesh Kumar Rajnish



Dr. Saurabh Agarwal



Dr. Kuldeep Vatsyan

¹Department of Orthopaedics, Post Graduate Institute of Medical Education and Research, Chandigarh, India.

Address of Correspondence:

Dr. Prasoon Kumar,

Department of Orthopaedics, Post Graduate Institute of Medical Education and Research, Chandigarh – 160 012, India.

E-mail: drprasoonksingh@gmail.com

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Table 1: Summary of all studies in the English literature reporting bilateral pathological neck femur fractures in chronic renal disease

Sl no.	Authors	Year	No. of cases	(Age of patients) in years	Primary renal	Treatment disease	Remarks
1	Zingraf et al ⁶	1974	1	45	Not known	Arthroplasty	First case reported in the literature
2	Gerster et al ⁷	1983	1 out of 2 cases reported had CRF	Case 1: 69 Case 2: 78	Case 1: chronic pyelonephritis Case 2: none	Case 1: bilateral THR Case 2: Conservative for right hip and Osteosynthesis for left inter-trochanteric hip fracture.	Excessive fluoride retention; fluorides given for spinal osteoporosis
3	Ogun et al ⁸	2001	2	Case 1:45 Case 2:35	Case 1: Renal amyloidosis Case 2: Chronic nephrolithiasis	Osteosynthesis with multiple cannulated screws in both cases	Bilateral neck femur fractures significantly deteriorate the general condition of patient.
4	Karapinar et al ¹	2003	1	23	Paralytic bladder leading to obstructive uropathy.	Bilateral cemented THA	Emphasised high morbidity and mortality in such patients; need for early surgical fixation and mobilisation
5	Hung et al ⁹	2009	1	39	Unknown etiology	Bilateral hemiarthroplasty	Look for renal disease in patients presenting with bilateral neck femur fractures.
6	Devkota et al ¹⁰	2013	1	47	Nephropathy secondary to diabetes mellitus and hypertension.	Non-operative management	Patient did not consent for surgery.
7	Garcia et al ¹¹	2013	1	43	CKD secondary to unknown etiology	Osteosynthesis	High degree of suspicion is necessary among renal osteodystrophy patients. Osteosynthesis may be superior to arthroplasty in these cases.
8	Satyanarayana et al ¹²	2015	1	23	Reflux nephropathy	Bilateral, uncemented, modular bipolar hemiarthroplasty	-
9	This study (John et al)	2018	3	1:44 2:15 3:64	ADPKD Renal dysplasia Pyelonephritis	Osteosynthesis Osteosynthesis Conservative(infected)	High mortality in this subset of patients, after bilateral NOF fractures.

chronic pyelonephritis, presented with RO causing two episodes of seizures and bilateral simultaneous atraumatic NOF fractures.

Case Report:

Case 1

A 44-year-old male patient was referred to our institute with a history of sudden onset of pain in both hips and inability to stand for 2 days. On clinical examination, both his lower limbs were lying in positions of excessive external rotation and he was unable to do an active straight leg raise. He was a known case of ADPKD, diagnosed with ESRD 4 years back; the patient was on hemodialysis albeit irregularly due to his non-compliance with

the treatment regimen. He had two episodes of generalized tonic-clonic seizures (GTCS) which preceded his symptoms. The patient had no previous history of seizures/epilepsy before this attack. The seizures were mostly precipitated due to electrolyte abnormalities (most probably hypocalcemia) or as a result of uremic encephalopathy due to the inadequately treated CKD. Plain radiographs revealed bilateral, displaced, and intracapsular, neck femur fractures (Fig. 1). On routine blood workup, he had moderate anemia (hemoglobin [Hb] = 7.2 g/dl) with de-ranged renal function tests; the serum creatinine level was elevated with a value of 6.4 mg/dl (N: 0.5–1.6 mg/dl), and urea level was 110 mg/dl (N: 7–21 mg/dl). His serum calcium, phosphorus, Vitamin D, and parathormone levels were 5.79 mg/dl (N: 8.5–10.5 mg/dl), 6.8 mg/dl (N: 2.5–5 mg/dl),



Figure 1: Plain radiograph of bilateral hips with pelvis in anteroposterior (AP) view showing bilateral, displaced, intracapsular neck femur fractures (Garden's type 4). Also note the significant osteopenia in both neck femur regions.



Figure 2: Post-operative x-ray in antero-posterior view after bilateral osteosynthesis with multiple cannulated screws.



Figure 3: X-rays of the 15 year old female, showing bilateral displaced neck of femur fractures.

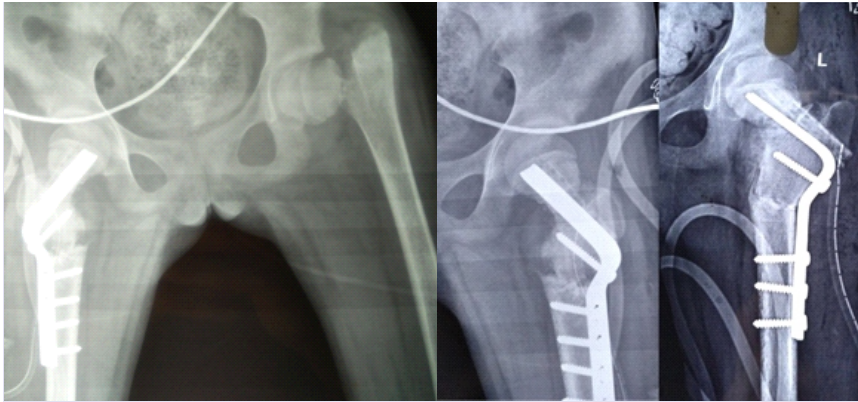


Figure 4 & 5: Post-operative radiographs showing open reduction & internal fixation with valgus osteotomy, angled blade plates & Fibula grafting.



Figure 6: Plain Radiographs Rays of the 64 year old male, showing bilateral neck femur fractures with gross osteopenia.

7.2 ng/ml (N: 9.2–34 ng/ml), and 137 pg/dl (N: 9–60 pg/dl), respectively, revealing hypocalcemia, hyperphosphatemia, osteomalacia, and secondary hyperparathyroidism. Patient's metabolic parameters were optimized under the guidance of the nephrologist by hemodialysis and multiple packed erythrocyte suspension transfusions. Calcium and Vitamin D supplementation were started simultaneously. 1 week after admission, osteosynthesis through closed reduction on a radiolucent fracture table under C-arm guidance and internal fixation with multiple cannulated screws was performed for both the fractures in two separate sittings (Fig. 2). The patient had no complications during or after surgery. The patient was discharged 2 weeks post-surgery after removal of sutures with no pain. Crutch walking with tolerable weight bearing was allowed 3 weeks after surgery. However, the patient expired 3 months after the surgeries due to renal and cardiopulmonary complications.

Case 2

A 15-year-old female patient presented to the outpatient department of our institute, with a history of sudden onset of pain in both hips and inability to stand for 6 weeks. On inspection, in supine position, both her lower limbs were lying in positions of excessive external rotation. She was still in considerable pain and would not allow further examination. She was a known case of RO secondary to renal dysplasia, diagnosed with ESRD 3 years back; the patient was on regular hemodialysis. She also had an episode of GTCS like the first case before the onset of pain. She had no previous history of seizures episodes. Plain radiographs revealed bilateral, displaced, and intracapsular neck femur fractures (Fig. 3). On routine blood workup, she had anemia (Hb = 8.2 g/dl) with deranged renal function tests; the serum creatinine level was elevated with a value of 12.6 mg/dl, and urea level was 40 mg/dl. Her serum calcium, phosphate, Vitamin D, and parathormone levels were 7.9 mg/dl, 5.5 mg/dl, 5.9 ng/ml,

and 469 pg/dl, respectively, revealing hypocalcemia, osteomalacia, and secondary hyperparathyroidism, akin to the first patient. Before the fixation, we optimized her metabolic status under the guidance of the nephrologist and proceeded with fixation of the fractures one by one, with angle blade plate (ABP), valgus osteotomy, and fibula grafting on 2 separate days (Fig. 4a and b).

Case 3

A 64-year-male was referred to our department by the nephrologist, with inability to walk for last 1 month. On clinical examination, both his lower limbs were again lying in positions of excessive external rotation and he was unable to do an active straight leg raise. He was a known case of CKD diagnosed 2 years back. Hemodialysis was done four times before admission. He had two episodes of generalized tonic-clonic seizures (GTCS) which preceded his symptoms. The patient had no previous history of seizures/epilepsy before the two episodic attacks. His plain radiographs again revealed bilateral, displaced, and intracapsular neck femur fractures (Fig. 5). His metabolic parameters were also found deranged and he had mild anemia (Hb = 10.1 g/dl) with deranged renal function tests; the serum creatinine level was elevated with a value of 5.0 mg/dl, and urea level was 148 mg/dl. His serum calcium, phosphorus, Vitamin D, and parathormone levels were 7.1 mg/dl, 6.6 mg/dl, 16.34 ng/ml, and 252 pg/dl, respectively, revealing hypocalcemia, hyperphosphatemia, osteomalacia, and secondary hyperparathyroidism akin to the other two cases. The patient had severe chest infection with culture positive for *Pseudomonas aeruginosa*. He was given antibiotics, steam, and nebulization for the same, as advised by the pulmonologist. Bipolar hemiarthroplasty was planned for NOF fractures, but intraoperatively fracture site was found to be infected, so only debridement was done. The patient was discharged on antibiotics. He suffered a cardiopulmonary arrest during a session of dialysis and expired 4 weeks later.

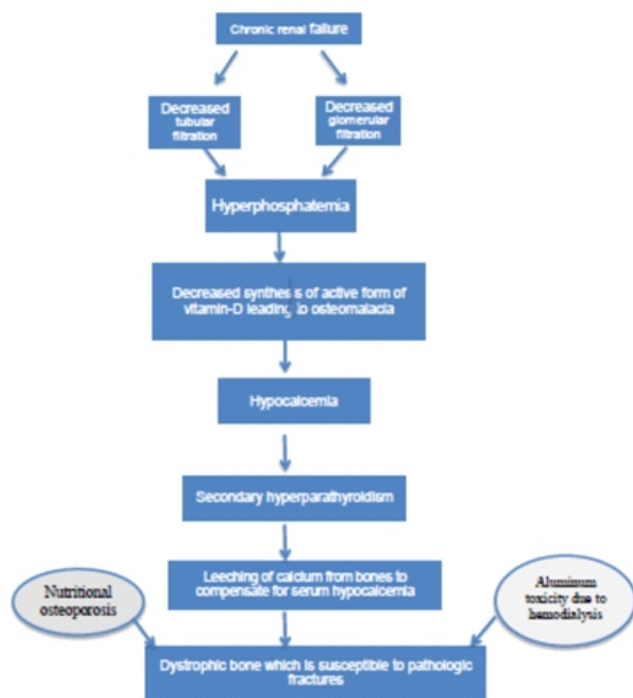


Figure 7: Flowchart depicting the vicious cycle of renal osteodystrophy leading to pathologic fractures.

Discussion:

RO is a fairly common complication observed in chronic renal failure [2]. It encompasses a wide gamut of skeletal abnormalities such as osteitis fibrosa, osteomalacia, hyperosteoidosis, osteosclerosis, and several types of developmental anomalies in children [3]. The pathophysiology of RO is complex and multifactorial (Fig. 6) [1, 4, 5]. Chronic renal failure is associated with hyperphosphatemia; this is due to decreased glomerular filtration leading to undue phosphate retention. Hyperphosphatemia along with decreased tubular filtration leads to a decrease in 1, 25 di-hydroxy vitamin-D synthesis which in turn causes hypocalcemia. Decreased absorption of calcium from gastrointestinal tract because of higher phosphate levels in its cell cytoplasm matrix adds to the insult and further exacerbates the hypocalcemia. This hypocalcemia in turn stimulates overactivity of the parathyroid glands. There is a resultant clear cell hyperplasia of all the 4 parathyroid glands leading to secondary hyperparathyroidism which tends to compensate for the low serum calcium levels by leaching calcium off the bones. The overall result of this vicious cycle is that the bone structure, especially in vertebra, pelvis, and neck femur regions, is significantly weakened and is, hence, rendered susceptible to pathological fractures [1, 4, 5]. In addition, associated nutritional osteoporosis and aluminum toxicity induced osteomalacia in patients undergoing long-term hemodialysis further weaken the bony architecture [1, 4]. In our patients, the fractures occurred after episodes of grand mal seizures (most probably secondary to hypocalcemia or uremic

encephalopathy) in pathologic bone already weakened by RO. Simultaneous, bilateral neck femur fractures in CRF, as observed in these cases, are extremely uncommon with only a handful being reported in the literature (Table 1) [1, 6, 7, 8, 9, 10, 11, 12]. Seizure-induced bilateral neck fractures in RO patients are even rarer with only two cases being reported so far [2, 8]. Our first case is also the first and only case of ADPKD in which this complication has been reported in the literature. Operative management and aggressive mobilization are necessary in CRF patients with pathological neck femur fractures [2, 13]. Schaab et al. in their case series of 11 pathological femur neck fractures in patients with chronic renal failure (including one case with bilateral fractures after GTCS) reported superior functional outcome, reduced complications and mortality rates in the seven patients who were treated operatively compared to the four patients who were treated non-operatively [2]. In the first case, as both the fractures were displaced, we decided to fix these fractures with closed reduction and internal fixation through multiple cannulated screws to prevent re-displacement. This allowed us to avoid the more extensive and complex hip arthroplasty surgery which is associated with a high complication rate and mortality in patients with chronic renal failure. In our second case, since the patient was of only 15 years, we went ahead and fixed the fractures with osteotomy, ABP, and fibula graft. Among the nine other cases of bilateral pathological neck femur fracture cases reported in the English literature (in patients with CKD), only Garcia et al. and Ogun et al. have opted for osteosynthesis, instead of hip arthroplasty [8, 11]. However, Devkota et al. treated their case non-operatively as the patient did not consent for surgery [9]. We were forced to manage one of the cases conservatively in whom bipolar hemiarthroplasty was planned, as done in the majority of reported cases; but due to the infection it was deferred and conservative management was initiated. A high degree of morbidity and mortality is associated with neck femur fractures in chronic renal failure. Klein et al. reported 1st-year mortality of 38% after surgical management of pathological neck femur fractures in chronic renal failure patients [13]. They also reported that optimization of the metabolic status of the patient before surgery under the guidance of nephrologists decreases the complications. The high mortality in this set of patients is reflected in two of our cases, as both the patient expired within 3 months after the fractures.

Conclusion:

Spontaneous, pathological, bilateral neck femur fractures are increasingly being reported in CRF patients. These fractures are associated with a high morbidity and mortality. These fractures can be prevented to a large extent by an early diagnosis of CKD

and proper supervised treatment. Treating physicians should always keep a high index of suspicion to rule out these fractures in such patients as these injuries can easily be missed in bed-ridden patients. Hip radiographs should be ordered in patients of CRF complaining of persistent hip pain as spontaneous non-traumatic fractures are known to occur in this subset of patients. Prior optimization of metabolic parameters under the care of an experienced nephrologist followed by early operative treatment and aggressive rehabilitation is the treatment of choice in these

cases.

Clinical Message

Atraumatic bilateral NOF fractures in patients of RO are rare entities, that are associated with significant morbidity and mortality and require early and efficient diagnosis and management.

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