

CONCOMITANT PYOMYOSITIS, OSTEOMYELITIS AND PATHOLOGIC FRACTURE OF THE DISTAL FEMUR: A CASE REPORT

Niko Augustine R. Abella, MD
Ruperto O. Estrada Jr., MD, MHA, FPOA, FPCS

INTRODUCTION

Pyomyositis is a purulent skeletal muscle infection that arises from hematogenous dissemination and is usually accompanied by abscess formation. [53] The definition of osteomyelitis is generally accepted as an inflammatory process of bone and bone marrow caused by an infectious organism(s) which results in local bone destruction, necrosis and apposition of new bone. The concomitant presence of pyomyositis (PYM), osteomyelitis (OM) and a septic pathologic fracture of the associated femur is rare. This situation presents challenges because the treatment may be staged and customized. Management of PYM heavily depends on the stage it is first diagnosed.

The Cierny-Mader classification is a clinical system that categorizes osteomyelitis based on its anatomical, clinical, and radiological characteristics. It divides osteomyelitis into four stages. In stage 1, known as medullary, the infection is limited to the medullary cavity of the bone. Stage 2, referred to as superficial, affects only the cortical bone and typically stems from direct inoculation or contiguous focus infection. Stage 3, termed localized, typically affects both cortical and medullary bone but doesn't compromise the entire bone diameter, with the bone generally remaining stable. Stage 4, called diffuse, involves the complete thickness of the bone and results in loss of stability, resembling infected nonunion. [59]

Some patients are diagnosed well beyond the first stage despite extensive tests. Targeted therapy for OM and the pathologic fracture by an antibiotic cement-coated nail (ACCN) using a cheaper core Kuntscher nail (K-nail) is historically associated with differing antibiotic combinations, timing of removal of the ACCN and up to 40% persistence of infection. [3,15,17,28,37,38,41] Here we present a case report of concomitant pyomyositis, osteomyelitis and closed pathologic femoral fracture managed with an ACCN using a core K-nail.

CASE REPORT

Our patient is a 34-year-old male laborer who consulted the Emergency Department (ED) because of pain and deformity on the right thigh. One day earlier he was lifting heavy equipment when he felt a crack over his right thigh followed by severe pain. Three months

earlier he experienced a gradual onset of right distal thigh and knee pains aggravated by prolonged standing and working. He never consulted a physician. Fourteen years previously he suffered a saddle injury wherein allegedly he sustained a urethral injury for which he was permanently placed on an indwelling urinary catheter. At the ED he denied recent episodes of chills, sweats, rashes, anorexia and weight loss. He was of thin frame, with tender and swollen right distal thigh. The latter was shortened and rotated. No wound, neurologic deficit, erythema, fever, warmth nor draining sinus were observed. Vital signs were stable. There was no osteolytic or -blastic lesion, periosteal reaction, cortical erosion or loss of cortico-medullary differentiation visualized on the knee and thigh radiographs leading to admission, application of a Steinmann Pin over the proximal tibia followed by placement on a balanced skeletal traction to prevent shortening. Chest x-ray was unremarkable. The diagnosis was a closed minimally comminuted fracture of the right distal femur. (Fig.1)



Figure 1. Radiographs at the ED revealed an oblique fracture of the Distal third of the right femur

Hemoglobin (HGB) was 131 g/L, hematocrit (HCT) 0.37, elevated white blood cell count (WBC) at $14.56 \times 10^9/L$, with neutrophilic predominance at (PMN) 0.86, lymphocytes 0.08, monocytes 0.06, platelet count (PC) $223 \times 10^9/L$, C-reactive protein (CRP) 6.30, erythrocyte sedimentation rate (ESR) 57 mm/hour. The initial urinalysis was unremarkable. From the day of admission, the patient was maintained on balanced skeletal traction to prevent shortening and maintain the femoral length. By the 17th hospital day (HD) the ESR elevated to 82 mm/hr and the CRP > 13.0 . Urinalysis on the HD 19 revealed red cells 2-5/hpf, pus cells 10-25/hpf, squamous epithelial cells 5-10/hpf, leukocytes +2 and slightly turbid transparency. The urinary tract infection (UTI) was treated with oral cefuroxime 500 mg twice daily for seven days. On HD 24 there was palpable fluid accumulation on the fracture site, warmth and erythema. (Figure 2.) The patient became febrile and

complained of pain while on balanced skeletal traction. WBC was $16.55 \times 10^9/L$, PMN 0.82, CRP 8.69, and ESR 82 mm/hour.



Figure 2. Gross Photo of the Lateral aspect of the Right Thigh

Ultrasound imaging is shown in Figure 3. A suspected inflammatory exudate was visualized (blue arrows). This abnormal fluid collection adjacent or around a bone without intervening soft tissue was suggestive of osteomyelitis according to the sonographer.



Figure 3. Ultrasonography of the Right Thigh

The blue arrows point to fluid within the fracture and adjacent to bone and quadriceps muscle. A sonographic diagnosis of osteomyelitis was made based on fluid in direct contact with bone without intervening soft tissue.

At the Operating Room the knee joint aspiration failed to demonstrate any purulent aspirate. The normal saline solution injected and aspirated later revealed no bacterial growth. Incision and drainage over the lateral aspect of the thigh was able to express approximately 700 milliliters of sero-purulent abscess fluid. During debridement the fascia were intact and the quadriceps appeared viable and bleeding. The femoral cortex surface was irregular and eroded; the operative site was thoroughly irrigated. The clinical diagnosis was revised to pathologic fracture secondary to chronic osteomyelitis, Cierny-Mader 3-A.

He was started empirically on intravenous (IV) piperacillin-tazobactam 4.5 gm every eight hours and IV clindamycin 600 mg every eight hours. *Klebsiella pneumoniae* was the organism isolated. As he was recuperating we planned to insert a (donated) interlocking antibiotic cement-coated Kuntscher nail 11 mm x 36 cm. This nail construct was intentioned to address the infection and fracture. We created one locking hole seven centimeters from the proximal end and two locking holes three and five cm from the distal end based on his templated radiographs and clinical measurements of his femur. (Fig. 3-A)



Fig. 3 A B C D

On HD 34 he underwent debridement, excision of a postero-medial sequestrum and reaming of the medullary canal up to 2 mm over the ACCN diameter. (Fig. 3-B) No purulence or abscess was observed. Two packs of bone cement each containing 20.4 gm polymethylmethacrylate (PMMA) powder and 7.2 gm liquid monomer (C-ment®, KRT Healthcare, Haryana, India) were hand-mixed with four grams of vancomycin powder. The doughy mixture was then applied into the whole length of the slot of the K-nail except for the proximal eye. No cement was applied on the external surface. Approximately 40 per cent of the antibiotic-cement mixture was in excess thus leaving about 2.4 grams of vancomycin in the antibiotic K-nail. Holes were drilled through the three locking holes for passage of the screws. The ACCN was inserted under firm pressure. The two peripheral holes were interlocked free-hand under fluoroscopy. (Fig. 3-C and 3-D).

The distal locking screw head can be seen on the distal femur. (Fig 3-C) On rotation of the thigh the fracture was still unstable and radiographically the fracture stability was precarious. We placed the lower extremity on a hip spica splint post-operative. Bacteriology culture was negative so that the current empirical antibiotics were continued. The post-operative recovery was without incident and his symptoms improved. His white cell count was $7.04 \times 10^9/L$, neutrophils 0.58, CRP 0.56 and ESR 52 mm/hour before his hospital discharge. Overall he was on parenteral empirical antibiotics for four weeks with which he responded well. The infectious disease service decided no oral antibiotic coverage on hospital discharge.



Fig. 2. A



B

C

Three months post-nailing there was bridging callus at the posterior and medial cortices without a change in the position of the implant. (Fig. 4-A) Knee flexion was up to 40° and there was extension lag of 10°. (Fig. 4-B) Heating modalities and therapeutic ultrasound were avoided during his physiotherapy. At seven months post-nailing he was fully ambulatory at home with a limp and a slow gait. (Fig. 4-C). Note the IFC bag in his left hand. We advised him to have the implant removed as soon as the fracture is fully consolidated and that he should exercise prudence regarding his activity restrictions.

DISCUSSION

Pyomyositis (PYM) is a purulent or suppurative infection of skeletal muscle mainly caused by *Staphylococcus aureus* in tropical regions. It is gaining incidence in temperate climes especially encountered among the immunocompromised and healthy children. Its incidence is unknown. The quadriceps muscle group is one of the most commonly affected. [8,29,34,45] Most cases are secondary to transient bacteremia. Urogenital sepsis associated with urine retention and medical intervention has been reported as a hematogenous etiology for PYM. [21,40,47] In-vitro studies on healthy rabbits have shown that the striated muscles are resistant to the development of abscesses when the animals are injected with *Staphylococcus aureus*. [49] Myoglobin binds strongly to iron which the microorganism needs to survive. A traumatized muscle undergoes sequestration of elemental iron which will lead to hematogenous invasion of bacteria. Ashken reported an unusual high incidence of PYM in military recruits who sustained repetitive minor trauma. [2] It has been postulated, without significant evidence, that malnutrition and multiple parasitism may play a role in causation. Immunological testing on PYM populations is generally unremarkable. This infection has three stages: invasive, purulent and late. [9] The invasive stage occurs during the first 10 days with gradual dull pain usually of a large muscle group, followed by edema, low-grade fever and progression of pain. The purulent stage presents 10-21 days after with worsening edema, tenderness and pyrexia. There are elevated inflammatory markers and findings of fluid using ultrasound and MRI. The late stage is distinguished by fluctuance, high fever, septicemia, possibly coma and end-organ failure. Generally stage 1 is managed by parenteral antibiotics, stage 2 by incision and drainage and stage 3 by debridement. The differential diagnoses include fever of unknown origin (FUO), muscle contusion, septic arthritis, osteomyelitis, cellulitis, muscle hematoma, deep vein thrombosis (DVT), muscle rupture or strain, muscle sarcoma, trichinosis and leptospirosis. [10] *K. pneumoniae* usually resides in the human intestines, yet it is one of

the most common Gram-negative bacteria that cause hospital-acquired UTI. It is cited as causative of PYM in diabetic individuals. [51,52]

Our patient's OM started as sub-acute as evidenced by pain/tenderness, absence of systemic symptoms, and the late diagnosis. This case possibly started as a Brodie's abscess similar to the report of Patel et al [36]; a magnetic resonance imaging (MRI) scan could have demonstrated early edema around the femur. Also an abnormal fluid collection adjacent or around a bone without intervening soft tissue as seen on ultrasound could have suggested osteomyelitis. [1,33] The vertebrae, symphysis pubis, sternoclavicular and sacro-iliac joints are mainly affected by adult OM while the long bones are impacted in children. UTIs, renal transplantations and urogenital interventions have been linked to hematogenous OM of multiple bones. The bacterial isolate was *Escherichia coli* in three case reports. [23, 47, 50] We do not know whether the PYM and OM were caused by a single organism; hematogenous OM is classically monomicrobial. Antibiotic therapy was recommended by our infectious disease service. It is unclear whether short intravenous monotherapy with or without an oral antibiotic regimen would be therapeutic for our patient's OM considering there was no organism isolated from medullary tissue. Hsieh and co-workers reported that in 13 studies there was no significant difference in outcomes between short- and long-term antibiotic therapy. In vertebral OM, outcomes between short and long term treatments were comparable. [19] Limited high-quality studies cite that oral therapy is non-inferior to parenteral treatment and that shorter antibiotic therapy may be suitable for low-risk individuals. Post-operatively our patient was hemodynamically stable, free of draining sinuses, was capable of taking in oral medications, had access to a regimen that will cover likely pathogens and had no psychosocial reason that precluded the safe use of oral therapy, all requirements that he could be transitioned to oral antibiotics according to Spellberg and co-workers. [43] No oral antibiotics were prescribed on account of potential bacterial resistance.

Slotted ACCNs are occasionally utilized for septic non-unions and infected fractures. Tandon and Thomas described two patients with persistent femoral osteomyelitis in which gentamicin beads were implanted inside the slotted nails. No IV gentamicin was given. The femora eventually consolidated, the nails removed and after a year the infections were clinically arrested. [46] Mantri and associates studied a cohort of 25 culture-positive cases with infected long bone fractures with bone defect less than 2 cm. K-nails were used for both femur and tibia of diameter 1 mm less than last reamer size. 1.0 gm vancomycin powder and 1.5 gm cefuroxime Na were mixed with 20 gm PMMA-

monomer. The doughy mix was inserted in the nail slot only. Twenty per cent patients showed excellent results with fracture union averaging 4.9 months, 60% had good results with union averaging 7.8 months and 16% had fair results with union averaging 10.4 months. [29] Craveiro-Lopez reported on a prospective controlled cohort of 30 patients with open fractures of the tibia. Sixteen patients were treated with an interlocking IM nail with a core of PMMA cement in which vancomycin 2 gm and flucloxacillin 2 gm were added to the cement compared to a group of 14 patients managed with standard locking non-ACC nails and IV antibiotics. Healing time averaged 4.5 months in the ACCN group vs. 7.5 months standard group, $p = 0.02$. The complication rate including infection was 25% in the ACCN group vs. 64% in the standard group, $p = 0.03$. [13] We do not possess in-vitro data regarding local elution of vancomycin from inside a slotted nail. Whether our patient can avoid recurrence is uncertain.

A reason for avoiding implanting AB-cement on the K-nail surface was the possibility of debonding and cement retention. The incidence of cement debonding is up to 40%. [11,17,26,30,39] Avoidance of hammering while inserting the nail [3], pushing the nail to the end of the mantle then pulling it back 1-2 mm [4], avoiding drilling through the cement-laden locking holes [11], ensuring uniform thickness of the cement coating throughout [30], allowing the cement to set for 15 minutes [15,39] and simply using a threaded metal core [24] are some of the recommendations to minimize debonding phenomenon. Nallamilli et al recommended winding a 22-G wire on a tibial nail from one end to the other end and then return in reverse direction to the starting end. [32] The recommendations for removing retained cement after ACCN removal include using hooks under fluoroscopy [12], utilizing a pituitary rongeur through a cortical window [25] and using long K-wires and S-pins and instrumentation for revision THA. [30] It may be challenging to convince a patient to have his/her ACCN removed electively since in recent years the ACCN has become a form of definitive treatment.

The local K-nail is an alternative for Winquist grades 1 and 2 isthmic and para-isthmic femoral fractures [5] and tibial mid-third diaphyseal fractures [16] at much lower cost and lesser radiation exposure. The reasons for seldom using it in infected femora are 1) the absence of a local commercial interlocking model and 2) the absence of a reliable targeting jig to use for customized interlocking holes. In 1968 Kuntscher introduced the "detensor" nail, soon to be called the interlocking nail, with one crossing screw proximal and the other distal, to keep the length and rotation of the limb. [14] We customize at POC by creating one to two holes proximally and two holes distally to simulate a contemporary interlocking design. One challenge is to create the right-sized hole diameter

to accommodate the locking bolt and to minimize weakening that junction. It is not clear on what is the minimum diameter for a K-nail to be used as a ACCN core. Small femora of many Filipinos preclude using wider-diameter nails. Alternative implant constructs to stabilize the pathologic fracture in our case include 1) an antibiotic-cement-coated (ACC) plate, 2) an ACC rod augmented with plate fixation or uniplanar external fixation, 3) a ring fixator plus antibiotic beads/spacer and 4) a retrograde nail. Antibiotic cement-coated plates (ACCP) have recently emerged as a prophylactic instrument for patients with a high risk for infection and with an infected bone. [7,12,22,36] Reports on this technique are few, study populations are small and for our thin patient the plate may become prominent and painful. The plate may fail due to a non-union. [44] Kumar et al evaluated 10 cases of infected femoral non-unions, seven of whom had additional uni-cortical plate fixation. Nine cases had stable union and complete arrest of the infections. [26] Procurement of this extra implant will further delay his surgery. A uniplanar external fixator supplementing an ACCN [25] will potentially develop pin-tract infection and loosening because of the poor-quality bone. A ring fixator, while providing better stability, is labor-intensive for the patient, will create problems during physiotherapy and can produce pin-tract infections, peroneal nerve palsy and unexplained femoral pains. A coated retrograde nail was not considered for fear of contaminating the knee joint. We considered a low-profile plate device and vancomycin cement beads if resources were available. The customization and off-label use of an orthopedic device is a practice in a low-resource setting. For now the present implant enabled the treatment of two contiguous infections and a fracture with one definitive surgical procedure. The antibiotic cement-coated K-nail for selected patients may not disappear any time soon.

SUMMARY

A 34-year-old male sustained a closed fracture of the distal third of the right femur. He has a past history of permanent indwelling urinary catheterization due to a urogenital injury 14 years previously as well as prodromal pains over the distal thigh and knee three months before. He developed 1) clinical subacute osteomyelitis months before the injury and 2) pyomyositis of the quadriceps muscles during hospital confinement due to presumptive bacteremia from recurrent UTI. *Klebsiella pneumoniae* was isolated from the drained pyomyositic abscess. On the second surgery the infections and fracture were simultaneously addressed by the application of an interlocking antibiotic-cement coated Kuntscher nail. The cement was applied inside the slot throughout the nail's length except for the eye extraction site. Despite the less than ideal stability afforded by the nail construct on a distal shaft fracture the infection was arrested and the fracture united

at seven months post-nail fixation. A high index of suspicion is warranted for pyomyositis or osteomyelitis in a patient with unexplained osteo-articular symptoms who suffers from recurrent urogenital infections.

DISCLOSURE

The authors declare that no competing interests exist.

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The POC Ethics Review Board does not review single-patient case reports.

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