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OPINION REVIEW

Delirium after emergency hip surgery – common and serious, but rarely consented for

Louis J Koizia, Faye Wilson, Peter Reilly, Michael B Fertleman

ORCID number: Louis J Koizia (0000-0003-4074-6926); Faye Wilson (0000-0002-4438-0370); Peter Reilly (0000-0002-3872-910X); Michael B Fertleman (0000-0003-4023-1156).

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Louis J Koizia, Michael B Fertleman, Geriatric Medicine, Imperial College NHS Trust, London W2 1NY, United Kingdom

Faye Wilson, Geriatric Medicine, City Hospitals Sunderland, Sunderland SR4 7TP, United Kingdom

Peter Reilly, Department of Trauma and Orthopaedics, Imperial College Healthcare NHS Trust, London W2 1NY, United Kingdom

Corresponding author: Louis J Koizia, MRCP, Doctor, Geriatric Medicine, Imperial College NHS Trust, Praed Street, Paddington, London W2 1NY, United Kingdom. l.koizia@nhs.net
Telephone: +44-20-33121077
Fax: +44-20-33121541

Abstract

A quarter of patients admitted with a proximal femoral fracture suffer from an acute episode of delirium during their hospital stay. Yet it is often unrecognised, poorly managed, and rarely discussed by doctors. Delirium is important not only to the affected individuals and their families, but also socioeconomically to the broader community. Delirium increases mortality and morbidity, leads to lasting cognitive and functional decline, and increases both length of stay and dependence on discharge. Delirium should be routinely and openly discussed by all members of the clinical team, including surgeons when gaining consent. Failing to do so may expose surgeons to claims of negligence. Here we present a concise review of the literature and discuss the epidemiology, causative factors, potential consequences and preventative strategies in the perioperative period.

Key words: Delirium; Proximal femoral fractures; Consent; Hip surgery; Montgomery; Medicolegal

Core tip: Delirium is defined as a clinical syndrome characterised by disturbed consciousness, cognitive function or perception, which has an acute onset and fluctuating course. Delirium is common amongst patients admitted following proximal femoral fracture but is frequently unrecognised and mismanaged. Delirium can dramatically affect an individual's stay in hospital. Delirium increases mortality and morbidity, leads to lasting cognitive and functional decline, and increases both length of stay and dependence on discharge. Delirium should be routinely and openly discussed by all members of the clinical team, including surgeons, physicians, anaesthetists, nurses and

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therapists. Simple strategies can be adopted to try and reduce the risk of delirium occurring and trying to manage delirium once it occurs.

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INTRODUCTION

The care and management of patients with femoral neck fractures have been the focus of growing interest over the past twenty years[1]. Early intensive geriatric input following hip fractures has been shown to reduce mortality and medical complications[2]. Similarly, multidisciplinary, collaborative models of care have been shown to decrease the incidence of postoperative complications and readmission rates[3]. Multidisciplinary care involving orthopaedic surgeons, geriatricians, therapists and discharge co-ordinators, allow regular discussion of patients and planning for discharge. Agreed standards of care now exist jointly written by geriatricians, orthopaedic surgeons and anaesthetists, a nationwide data collection system keeping track of management and outcomes (the National Hip Fracture Database) and a payment system that rewards trusts adhering to best practice care[4]. There is an evidence base to guide practice and also scores that may predict poor outcomes[1,5].

One predictor of poor outcome is post-operative delirium, which can affect a large proportion of patients post operatively, yet is rarely mentioned to patients or families when the treatment and recovery from a proximal femoral fracture is discussed[6]. Delirium is defined as a "clinical syndrome characterised by disturbed consciousness, cognitive function or perception, which has an acute onset and fluctuating course"[7].

INCIDENCE AMONGST OLDER ADULTS

Many studies have looked at delirium rates amongst older adult inpatients, and estimates of prevalence vary from 29% to 64% amongst medical wards, with patients being admitted for a number of reasons and not primarily delirium[8-10]. The variance is thought to be due to heterogeneous or poorly defined diagnostic criteria used in different clinical trials, often leading to under diagnosis. Surgical patients are no different, with studies of post-operative cardiac bypass patients detecting it in up to 52% of subjects[11]. A meta-analysis looking at incidence of delirium post hip fracture in elderly patients found an accumulated incidence of 24%[12].

There are many triggers for developing delirium, including infection, myocardial and cerebral ischaemia, electrolyte abnormalities, commencement of new medications, withdrawal from alcohol or drugs (such as benzodiazepines), pain, constipation and urinary retention[13]. Certain patient characteristics also predispose to developing delirium; these include advanced age, pre-existing cognitive impairment, sensory impairment (from visual and hearing loss), poly-pharmacy, and medical co-morbidities[14]. Specifically related to hip fractures, the mechanism of injury, the fracture, the operation and anaesthetic can all increase the risk of developing delirium[12,15,16].

DIAGNOSING DELIRIUM – CRITERIA AND PATHOPHYSIOLOGY

In clinical trials the incidence of diagnosed delirium varies widely, even when the diagnosis is being actively sought as an outcome measure, implying that in the everyday ward-based setting diagnosis may be even more elusive[17]. Most healthcare professionals would recognise that the agitated, aggressive patient keeping the ward awake at night is unwell, but how many accurately diagnose hyperactive delirium? The sleepy patient, "just not herself", refusing therapy and staying in bed may have hypoactive delirium, but this is less likely to be diagnosed and treated appro-

priately[18].

The Confusion Assessment Method (CAM) is a quick, accurate and frequently used tool to recognise and diagnose delirium[19]. It can be incorporated into routine assessment of patients and has been shown to have high sensitivity, specificity and interrater reliability[20]. The CAM algorithm is based on: (1) Acute onset and fluctuating course; (2) Inattention; (3) Disorganised thinking; and (4) Altered consciousness[21]. A diagnosis of delirium requires the presence of features 1, 2 and either 3 or 4 (Table 1).

Hyperactive and hypoactive subtypes are broadly defined according to psychomotor activity, with clinical features varying accordingly, but mixed delirium, with features of both, also exist[22]. The pathophysiology of delirium is not fully understood, and of the many theories none explain the variation in presentation and severity. Current theories centre on deranged neurotransmission and signalling pathways, as well as neuroinflammation and glial activation, which appear to be pivotal in the post-operative period[22].

CONSEQUENCES OF DELIRIUM ARE AS SERIOUS AS INFECTION OR THROMBOEMBOLIC EVENTS

Does confusion matter in the context of breaking a hip and having a major operation, and is it important enough to mention during the consent process? Are the consequences of deep wound infection, thromboembolic events and bleeding more severe, necessitating their inclusion on consent forms over delirium?

Whilst it is true that a large pulmonary embolus (PE) can be fatal, and revision surgery to replace an infected implant can be catastrophic for functional recovery and is associate with poorer outcomes, there is evidence that delirium also has a major and long-lasting effect[23]. Suffering from an episode of delirium has an impact on length of stay, post discharge institutionalisation and mortality[24]. Patients require greater input from nursing staff on the ward, both to provide personal care and to ensure safety. They are less able to work with physiotherapists leading to longer periods of therapy being required and cost of admission[25]. A substantial proportion of those who eventually return home will require extra assistance, both formally from social services and also informally from family and friends[26]. Delirium is an independent marker for increased 12 month mortality following a hospital admission, with the effect more marked for patients without pre-existing dementia[27].

As well as a change in dependency and social circumstances, quality of life can be directly affected by the psychological impact of delirium itself, both during the actual episode and also in the longer term[28]. A study looking at cancer patients suffering delirium identified that patients recalled their episodes of delirium, and expressed distress as a result[29]. Patients commonly describe concerns with day-night orientation, communication difficulties and delusional thoughts[30]. The longer lasting psychological impact of delirium, is not only experienced by patients but members of staff and patient relatives; caregiver distress as a result of delirium has been identified as being even higher than patient distress[28].

MANAGEMENT STRATEGIES FOR DELIRIUM

Once delirium has developed there is limited evidence that medications such as benzodiazepines or neuroleptics are beneficial and they are associated with severe side effects, particularly in the elderly population[31]. The main stay of pharmacological measures is to treat causative factors, such as using antibiotics to treat an underlying infection[22]. Proactive geriatric consultation can reduce episodes of delirium by 40% in patients with proximal femoral fractures[6]. The Hospital Elder Life Program reduced the number and duration of episodes by using delirium prevention strategies (such as maintaining orientation, reducing sensory deficits, ensuring adequate nutrition and hydration) to improve the overall hospital experience[32].

The best management strategy for delirium is prevention in the first place. Early orthogeriatric input as part of a multidisciplinary team has been found to prevent delirium[33]. The comprehensive geriatric assessment focuses on eliminated unnecessary medication, early identification and treatment of complications and mobilisation.

Specifically, in patients with femoral neck fractures, adequate pain management and analgesics have effect on delirium. Yang *et al*[12] identified patients receiving morphine were three times more at risk of developing delirium after hip fracture surgery than nonusers. Despite it being important to rationalise and ensure safe

Table 1 Confusion assessment method – adapted from Inouye *et al*[20]

Feature 1 Acute onset and fluctuating course
Is there evidence of an acute change in mental status from the patient's baseline? Does the abnormal behaviour fluctuate during the day?
Feature 2 Inattention
Does the patient have difficulty focusing attention, *e.g.*, easily distractible, difficulty keeping track of what was being said?
Feature 3 Disorganised thinking
Is the patient's thinking disorganised or incoherent, such as rambling or irrelevant conversation?
Feature 4 Altered level of consciousness
Is the patient's level of consciousness reduced?
Features 1 and 2 and either 3 and 4 are required for a diagnosis of delirium

prescribing in our population group to prevent delirium, undertreated pain and inadequate analgesia appear to be risk factors for developing delirium in frail older adults[34,35]. The analgesic, pain and delirium association remains inconsistent. Therefore, a fine balance with regular review of patients is imperative.

Prophylactic pharmacological therapies have been trialled, but showed no efficacy in reducing the incidence of delirium[36]. One important element for the prevention of delirium, is education of the medical staff. An education program can lead to an increase in detection of delirium as more cases are recognised, yet a decrease in point prevalence (19.5% to 9.8%) due to a more proactive approach[37]. Multidisciplinary teaching programs are also essential, as other members of the team who spend a proportionally large amount of time with patients, play an important role in implementing management and prevention strategies.

CONSENT PROCESS

Patient autonomy is one of the four pillars of modern medical ethics and stemming from this is the right of the patient to determine whether they wish to undergo any medical intervention. In order to give consent for surgery, patients must be presented with sufficient information about the proposed procedure and the treatment options, to be able to determine the pros and cons of either giving or refusing their permission[38]. The Courts have long-held that the potential complications which should be mentioned are both the common and the rare but serious[39]. In hip fracture the emphasis is slightly different, as the surgery is fundamentally essential, and outcomes worsen as surgery is delayed[40]. However, the premise that both common and serious complications should be mentioned still holds true.

Traditionally patients giving their consent sign a consent form as part of this process, but the varying standards of the quality of information provided to the patient are well documented[41]. A review of 100 consent forms of patients undergoing surgery for hip fracture, looked at the quality of documentation of patient and procedure details, as well as which potential complications were mentioned[42]. The number of complications mentioned ranged from four to eleven, with infection, thromboembolic complications, and bleeding most commonly included. In total 30 different complications were stated, with not a single mention of post-operative confusion or delirium. Rates of PE and deep wound infection are far lower than delirium, at under 7% and 3.6% respectively, but these complications are almost always mentioned[43-45].

Since 2008, the British Orthopaedic Association has endorsed the use of standardised, pre-printed consent forms for common operations, in order to improve the information given to patients prior to consent[46]. This was initially designed to reduce litigation and induce a standard amongst surgeons[46]. However, the consent form for fixation of a fractured hip does not mention delirium as a complication.

The 2015 supreme court case of Montgomery changed the way surgeons should approach consent. This was an obstetric case where a woman with diabetes had a complicated vaginal delivery that resulted in complications of shoulder dystocia and cerebral palsy. The mother sued for negligence stating that if she had known of the increased risk, in her case she would have requested a caesarean section. The supreme court held that doctors are required to take "care to ensure that the patient is aware of any material risks involved in any recommended treatment, and of any reasonable alternatives or varied treatments"[47]. The ruling established a duty to warn of material risks, with a material risk being defined as something that a patient would consider relevant. Effectively, Montgomery changed consent in the United Kingdom from

what the reasonable doctor would tell a patient, to what a reasonable patient would want to know. All interventions must be based on individualized discussions and be patient centred, allowing shared decision-making.

PARADOX – COULD SURGERY BE A PREVENTER TO DEVELOPING DELIRIUM?

Today, the vast majority of patients are managed following a hip fracture with an operation; this stems from the greater mortality in those patients not operated[48]. In addition, the operation provides fixation of the underlying fracture and reduces pain overall. Therefore, a hip operation following a hip fracture reduces mortality, pain, length of stay and increases mobility[49-51]. The operation aids recovery and therefore possibly reduces the risk of delirium from increased analgesic use, medical complications and prolonged hospitalisation. So therefore, should we be consenting for delirium preoperatively if it could be seen to reduce delirium occurring? The simple answer remains yes. The perioperative period brings its own risks for developing delirium, anaesthetic, theatre environment, stress and thus patients should be informed. In addition, a similar conundrum could be considered for VTE. As previously mentioned, VTE is nearly always discussed during the consent process. However, the risk of VTE following a hip fracture without an operation remains present due to prolonged immobility.

RECOMMENDATIONS

As highlighted, delirium is a common complication with consequences for the patient. The General Medical Council view on consent is that all possible adverse outcomes should be discussed with patients. It is for this reason that all patients should be consented for delirium in advance of surgery to repair a proximal femoral fracture.

As Montgomery emphasised the duty to inform a patient of a particular risk lies with the operating surgeon. Given the surgery is being performed by the orthopaedic team, it is their duty to inform and consent patients for possible delirium. That beings said, it is the job of the multidisciplinary team including physicians, anaesthetists, therapists and nurses to reduce risks of developing delirium and manage as best as possible.

CONCLUSION

In summary, delirium is a common complication of emergency hip surgery, with serious and long-lasting effects that have a significant and deleterious impact on patients' lives. It occurs as frequently as DVT, infection, haemorrhage and PE combined, currently the most commonly mentioned potential complications of emergency hip surgery. However, at present delirium is rarely, if ever, mentioned by surgeons to patients or their families during the consent process, despite being a good predictor of poor outcome and being very distressing to the patient, their relatives, and carers.

The United Kingdom Supreme Court and General Medical Council state that patients should be properly informed about risks of operations prior to surgery. A failure to mention delirium exposes the operating surgeon to risks of allegations of negligence and could be interpreted as unethical behaviour.

A further benefit of delirium being mentioned routinely by senior surgeons during the consent process would be to increase awareness of delirium and its consequences amongst junior doctors and allied health care professionals. In many centres they provide the routine day-to-day care of frail elderly patients following surgery for a proximal femoral fracture, yet receive little training on how to recognise or manage delirious patients. Whilst geriatric consultation is beneficial and improves outcomes, this service is not available out of hours in the majority of hospitals, and the initial investigations and management are often simple measures such as treating infection, anaemia, alcohol withdrawal, dehydration, pain and constipation.

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ORIGINAL ARTICLE

Retrospective Study

Functional physiotherapy method results for the treatment of idiopathic clubfoot

Noriela Carmen García-González, Jorge Hodgson-Ravina, Armando Aguirre-Jaime

ORCID number: Noriela Carmen García-González (0000-0001-8274-9653); Jorge Hodgson-Ravina (0000-0001-6351-5827); Armando Aguirre-Jaime (0000-0002-0749-4993).

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Noriela Carmen García-González, Jorge Hodgson-Ravina, Armando Aguirre-Jaime, Servicio de Rehabilitación, Servicio de Ortopedia y Traumatología, Unidad de Investigación Clínica y Experimental, Hospital Universitario Nuestra Señora de Candelaria, Santa Cruz de Tenerife 38010, Spain

Armando Aguirre-Jaime, Colegio de Enfermería, Laureate International Universities, Santa Cruz de Tenerife 38001, Spain

Corresponding author: Noriela Carmen García-González, BSc, MSc, Physiotherapist, Servicio de Rehabilitación, Hospital Universitario Nuestra Señora de Candelaria, Carretera del Rosario 145, Santa Cruz de Tenerife 38010, Spain. ngargon@gobiernodecanarias.org
Telephone: 34-92-2-602310
Fax: +34-92-2-600597

Abstract

BACKGROUND

Idiopathic clubfoot is a congenital deformity of multifactorial etiology. The initial treatment is eminently conservative; one of the methods applied is the Functional physiotherapy method (FPM), which includes different approaches: Robert Debré (RD) and Saint-Vincent-de-Paul (SVP) among them. This method is based on manipulations of the foot, bandages, splints and exercises adapted to the motor development of the child aimed to achieve a plantigrade and functional foot. Our hypothesis was that the SVP method could be more efficient than the RD method in correcting deformities, and would decrease the rate of surgeries.

AIM

To compare the RD and SVP methods, specifically regarding the improvement accomplished and the frequency of surgery needed to achieve a plantigrade foot.

METHODS

Retrospective study of 71 idiopathic clubfeet of 46 children born between February 2004 and January 2012, who were evaluated and classified in our hospital according to severity by the Dimeglio-Bensahel scale. We included moderate, severe and very severe feet. Thirty-four feet were treated with the RD method and 37 feet with the SVP method. The outcomes at a minimum of two years were considered as very good (by physiotherapy), good (by percutaneous heel-cord tenotomy), fair (by limited surgery), and poor (by complete surgery).

RESULTS

Complete release was not required in any case; limited posterior release was

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done in 23 cases (74%) with the RD method and 9 (25%) with the SVP method ($P < 0.001$). The percutaneous heel-cord tenotomy was done in 2 feet treated with the RD method (7%) and 6 feet (17%) treated with the SVP method ($P < 0.001$). Six feet in the RD group (19%) and twenty-one feet (58%) in the SVP group did not require any surgery ($P < 0.001$).

CONCLUSION
Our study provides evidence of the superiority of the SVP method over the RD method, as a variation of the FPM, for the treatment of idiopathic clubfoot.

Key words: Congenital clubfoot; Clubfeet; Talipes equinovarus; Conservative treatment; Physical therapy; Physiotherapy techniques; Conservative methods

Core tip: We have compared the clinical results of the treatment of idiopathic clubfoot in the context of the improvement accomplished and the frequency of surgery needed to achieve a plantigrade foot with two Functional physiotherapy methods: Robert Debré (RD) and Saint-Vincent-de-Paul (SVP). Both approaches managed to avoid complete surgery, which shows that the physiotherapies achieve a more flexible foot, allowing a more conservative surgery. Our data indicate that the SVP method achieves prolonged correction of deformities more efficiently than the RD method; the best advantage of the SVP method over the RD method was the greater number of cases without any surgery.

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INTRODUCTION

Idiopathic clubfoot is a common birth defect that occurs in one per 1,000 births. The etiopathogenesis has been linked to several genes and environmental factors, such as consanguinity of the parents, smoking during pregnancy, maternal age, alcohol consumption, oligohydramnios, among others. Approximately 20% of clubfeet are associated with distal arthrogryposis, myelomeningocele, amniotic bands, or other genetic syndromes, and in some cases with talar vertical. In 80% of cases, the etiology is unknown and are referred to as idiopathic clubfoot, of which almost 25% have family history[1-5]. Children with idiopathic clubfeet may have problems with balance, coordination, gross motor function, strength and agility. Neurological developmental difficulties should also be taken into account at the time of assessment, since knowledge of these conditions could facilitate the management of treatment, and the support needed for the patient and their families. The perception of difficulties in mobility, day-to-day activities, pain and discomfort negatively affect the quality of life. The diagnosis of clubfoot has a negative psychological impact for the parents; therefore, it is important that they receive emotional support, information and education about the pathology[6-10]. Currently, the initial treatment of clubfoot is eminently conservative. Among the best-known conservative methods, we highlight the Ponseti method (PM) and the Functional physiotherapy method (FPM), also called the French method. The PM includes manipulation, serial casting, Achilles tendon tenotomy and foot abduction brace. Several studies have reported that PM achieved the initial correction in a shorter time (3 to 13 castings), and 79%-96% of cases are subjected to tenotomy. Some problems have been reported, which include plaster, the above-knee cast making the perineal hygiene more difficult, the removal of cast making it stressful for the child and parents, skin wounds that can be produced by the cast knife or saw, and even skin burns caused by exothermic reaction[11-14]. The FPM is based on manipulations of the foot, bandages, splints and exercises adapted to the motor development of the child aimed to achieve a plantigrade and functional foot. The treatment is extended after the correction phase (around 3 mo) until the child reaches independent walking. The thermoplastic splints are light and of variable

rigidity, easy to place by the parents, have good acceptance by the family and child, allow adequate perineal hygiene, and adapt to the phases of motor development. The FPM provides comprehensive care; it deals with very important aspects such as propioception, coordination, balance, flexibility, muscular reinforcement, resistance, facilitates the acquisition of motor skills, in addition to educating and training parents for the management of the pathology. The different approaches of the FPM[15] have enriched the working context of the different multidisciplinary teams, including Bensahel *et al*[16] [Robert Debré (RD) method], Seringe *et al*[17,18] [Saint Vincent de Paul (SVP) method] and Diméglio *et al*[19] (Montpellier method). However, there is a lack of comparative studies between them. Our hypothesis was that the SVP method could achieve prolonged correction of deformities more efficiently than the RD method and decrease the rate of surgeries. The goal of this study was to compare the clinical results of the treatment of idiopathic clubfoot regarding both the improvement accomplished and the frequency of surgery needed to achieve a plantigrade foot with two FPM: the RD and SVP.

MATERIALS AND METHODS

Patients

This is a retrospective study of a series of cases of clubfeet (n = 71). The review of the therapeutic outcome was carried out on 46 children born between February 2004 and January 2012 with idiopathic clubfoot. The feet were treated in a public hospital with the RD method (n^1 = 34, before 2009) and the SVP method (n^2 =37, from 2009). Data were taken from the medical records. The children were between 1 and 45 d old when they began treatment, and had a minimum follow-up of two years. Before starting the treatment, feet were photographed (to observe the conditions of the feet in detail and the sequential progress during the treatment, serving as support of the information obtained with the scale), evaluated and then classified according to the severity based on the Dimeglio-Bensahel scale[20] by physicians or physiotherapists experienced with this rating system. This scale ranges from 0 to 20 points (0, 1-5, 6-10, 11-15 and 16-20, corresponding to normal, benign, moderate, severe, and very severe foot, respectively). This scale is widely used, and has proven to be reliable and reproducible in preceding intra-observer and interobserver studies[21,22]. We included children that were treated in our hospital with moderate, severe and very severe idiopathic clubfoot; those who attended the treatment sessions and complied with good observance of the protocol (these data was reflected in the clinical records through the care control sheet carried out by the physiotherapist responsible for each case), and we excluded those classified as benign or non-idiopathic, those previously treated with another method in other hospitals, and those who did not perform the sessions or did not properly comply with the protocol. The Ethics Committee of the Nuestra Señora de Candelaria University Hospital approved this study.

Interventions

Trained physiotherapists with over ten years of experience working with the RD method performed the treatment. Our experience with the SVP method started in 2009 after the team received training in Paris.

The RD group was treated according to the approach proposed by Bensahel *et al*[16], and also followed by Souchet *el al*[23]. We manipulated the foot daily and sequentially, and then applied elastic and nonelastic taping (closing the taping with a cohesive bandage to give greater consistency) between the sessions to maintain the correction obtained, until the pre-standing stage. When the child began to walk, a straight shoe was worn during the day and the Denis-Browne bar at nighttime. After correction of the foot was obtained, the parents continued the stretching, the splinting and exercises daily. The SVP group was treated according to the approach proposed by Seringe *et al*[17,18] with an additional manipulation for the correction of the cavus, as previously described[11]. This additional manipulation consisted in slightly supinate position with the forefoot moving into its proper alignment with the hindfoot. We manipulated the foot daily and globally, applied an inextensible taping on a rigid plantar plate between the sessions, and then a splint was placed full-time to keep the foot aligned with respect to the leg. The physiotherapist shaped the splint according to the correction achieved and the growth of the child. When the child began to walk, a straight shoe was worn during the day and the short splint during naps, with an above-knee splint overnight. After correction of the foot was achieved, the parents continued the stretching, placing the taping, splinting and exercises daily.

Both interventions were similarly performed. Before starting the treatment, the team explained to the parents the procedure and the care needs. We also stressed that

the adhesion of parents to the treatment is a prerequisite for success. Therefore, they were given clear instructions about the use of the splint and the importance of rigorously complying with the protocol. During the sessions, the parents were asked if they complied with the guidelines given and if they experienced any difficulty. The treatment was divided by stages, always adapted to each case: (1) Stage of deformities reduction: the first 3 mo, with physiotherapy daily; (2) Stage of maintenance: from 4th month until pre-standing, with physiotherapy two or three times a week. The parents were trained by the physiotherapist to perform daily stretching, taping, splinting and exercises; and performed these tasks in some sessions in order to check the training; (3) Stage of standing and walking: passive mobilizations, with active physiotherapy one or two times a week, adapted to the motor development of the child (some manipulations and active physiotherapy are shown in Figures 1 and 2, respectively). In those cases in which we observed a slight adduct of the forefoot when the child walked, a flexible bandage was applied to use with the footwear in order to improve the support and realign the foot with the leg.

The manipulations were performed daily with gentle joint tractions with the child stress-free, each session lasted 30 min per foot and was done by the same physiotherapist. In each bimonthly consultation, the feet were rated again using the Dimeglio-Bensahel scale[20] in order to objectify the improvement achieved (also, a series of photographs of the feet were taken); a score ≤ 5 was considered good enough, and > 5 not good enough. If at 8 mo of age the treatment was no longer effective, the evolution was considered stabilized and two surgeons evaluated the need for surgery and the optimal time to perform it. In our hospital, it was generally between 10-11 mo old; the surgeon considered that upon re-initiation of physiotherapy post-surgery, the child would be prepared to stand up, and this contributed to maintaining the correction of the equine of the calcaneus. According to the clinical assessment, we estimated that for the feet that did not exceed 90 degrees ankle dorsiflexion, a percutaneous heel-cord tenotomy was scheduled. When the calcaneus remained elevated with contracture of the posterior soft tissues without reaching 90 degrees of dorsiflexion of the ankle, a limited release was scheduled (Achilles tendon lengthening, with subtalar and tibiotalar capsulotomy). In specific cases, limited surgery could be supplemented with release of the adductor hallucis, and/or plantar fascia through a mini-incision; these were noted as nonrelease surgeries). When the foot was not corrected, and kept triple deformation and stiffness, a complete release (extensive posteromedial release) would be indicated. The surgery was a complementary intervention and was tailored to the specific needs of each case, with an intent to be as conservative as possible. The feet were not X-rayed at the time of revision.

The immobilization was performed with long plaster in knee flexion at 90 degrees for 4-6 wk. At 3 wk, the cast was changed in the operating room under anesthesia to check the correctness achieved, the skin and the scar. The physiotherapy post-surgery was immediately provided to stabilize the correction achieved, including in cases of surgery for recurrence. When the child walked properly, the treatment was considered complete. Then the child was discharged and was controlled each month, then eventually every 3-6 mo, and throughout the growth to detect any functional impairment. If there was any deterioration, it was again referred to physiotherapy. We recommended using the splints up to 4-5 years old, according to severity and evolution.

We could not complete the data for three patients (four feet) because they did not follow the treatment properly for various reasons: three feet in the RD group (two of which developed an allergy to the taping and had to stop the treatment, and one of a child who was changed to another hospital) and one foot in the SVP group also due to a change of residence. Therefore, we did not get considered for the results.

Assessment of outcome

The primary outcome measure was the rate of the severity of deformity by the Dimeglio-Bensahel scale[20]. To get this scoring, the degrees of reducibility of the internal rotation of the calcaneo-forefoot block, the adduction of forefoot relative to hindfoot, the equinus and the varus of the hindfoot were measured using a small goniometer and the charts. These four components can add a maximum of 16 points. It was also taken into account whether the foot presented medial and posterior creases, cavus, and the poor muscle condition (hypertonic, contracture, amyotrophic). Each of these conditions adds one more point. A second outcome measure was the need of complementary surgery to achieve a plantigrade foot. Other data recorded were the affected laterality, gender, and date of birth. To achieve a plantigrade foot, patient outcome were defined as: (1) Very good, when obtained only by physiotherapy; (2) Good, complemented by percutaneous heel-cord tenotomy; (3) Fair, complemented by limited release; and (4) Poor, complemented by complete release.

Figure 1 Manipulations of the clubfoot. A: Reduction of the talo-navicular joint subluxation; B: Derotation of the calcaneo-forefoot block; C: Achilles tendon stretching; D: Achilles tendon stretching with midtarsal protection; E: Stretch the median groove and plantar fascia; F: Passive mobilizations in plantar flexion and dorsiflexion.

Statistical analysis

A biomedical statistician performed a statistical review of the study. The sample characteristics were summarized as relative frequencies of their component categories for nominal variables and as median (range) for numerical variables due to its non-normal distribution. Comparisons between treatments were performed with the χ^2 tests for nominal variables and the *U* test for numerical ones. The odds-ratio (OR) analysis was used to determine the relapse rate of the approaches. We observed a 25% success rate (good and very good results) with the RD method; so in order to detect a clinically relevant difference of at least 40% more success with the SVP method, a sample of 31 feet in each treatment group would be required to achieve a study power of 90% in bilateral hypothesis testing with a level of statistical significance $P \leq 0.05$. All calculations were carried out using the statistical software package IBM SPSS 24.0® for Windows NT®.

RESULTS

Due to the lack of data from 3 patients (4 feet), the final sample consisted of 67 idiopathic clubfeet from 43 children (29 males and 14 females). Thirty-one feet (46%) were treated with the RD method and thirty-six feet (54%) were treated with the SVP method. The comparison of both groups at baseline is shown in Table 1. Both groups showed homogeneity at baseline for all the considered factors; although the average age of the children in the SVP group was lower, the differences were not statistically significant. The comparison of the improvement achieved by category with RD and SVP groups at 8 mo with respect to baseline is shown in Table 2. It has been found that the percentage of moderate feet that reached normality was lower in the group treated with the RD method (40%) compared to those who were treated with the SVP method (80%); the percentage of severe feet that managed to reclassify as benign was lower in the group treated with the RD method (16.6%) compared to those who were treated with the SVP method (100%); the percentage of very severe feet that managed to reclassify as benign was null in the group treated with the RD method (0%) compared to those who were treated with the SVP method (44.4%). A statistically significant difference was reached $P = 0.001$. The comparison of the results obtained by category at two years of age for the RD and SVP groups to achieve a plantigrade foot according to the procedure that was necessary is shown in Table 3. It has been

Figure 2 Physiotherapy adapted to motor development of the child. A: Strengthening of the fibularis muscles; B: Squat for active stretching of the Achilles tendon; C: Incline support to stretch the triceps surae; D: Support by four points to stretch the entire posterior muscle chain; E: Propiocepción on an air cushion; F: Sitting down in a toy car to stimulate support and propulsion; G: Going up and down stairs; H: Squat at four points to stretch the Achilles tendon with feet abduction.

seen that only 60% of moderate feet treated with the RD method were corrected with physiotherapy *versus* 100% of those treated with the SVP method. The 100% of severe feet treated with RD needed limited surgery; while of those treated with SVP, 65% were corrected with physiotherapy and 35% with hell-cord tenotomy. With both methods, all very severe feet required limited surgery. The outcomes were very good for 19% (only physiotherapy), good for 7% (with heel-cord tenotomy), fair for 74% (limited posterior release), and poor for 0% (complete release); and very good for 58% (only physiotherapy), good for 17% (with heel-cord tenotomy), fair for 25% (limited posterior release), and poor for 0% (complete release) for the feet treated with RD and SVP methods, respectively ($P < 0.001$). In the RD group, surgery was supplemented with release of the adductor hallucis and/or plantar fascia in 11 feet, and in the SVP group it was supplemented only in two feet. Examples of the progression of the clubfeet with the RD and SVP methods are shown in Figures 3 and 4, respectively.

In the RD group, the relapses occurred between two and three years old in five very severe feet that initially had fair outcomes; these were treated again with physiotherapy, but required another limited surgery. In the SVP group, the relapses occurred at 3 years old in five feet. Four of these feet were initially very severe and one foot was severe. Three of these feet initially had a fair outcome; two feet required another limited surgery, and one foot at the time of review was undergoing physical therapy; and the remaining two feet that initially had good outcomes were rescued with physiotherapy. An OR = 1.20 indicates that the probability of relapse in the RD group is 1.2 times more than in the SVP group. It is shown in Table 4.

DISCUSSION

We compared two approaches of the FPM regarding the improvement accomplished and the frequency of surgery needed to achieve a plantigrade foot. Our data indicate that the SVP method achieved prolonged correction of deformities more efficiently than the RD method, and substantially decreased the rate of surgeries. In this study, we revealed that both approaches managed to avoid complete surgery, which points to the overall effectiveness of the FPM. This shows that the physiotherapy achieved a more flexible foot, allowing a more conservative surgery. This is particularly significant because it has been shown that extensive surgery results in long-term overcorrection, stiffness, pain and osteoarthritis of the foot and ankle; a lesser correction is well-tolerated and easier to treat in adulthood than a hypercorrection[24]. Our major difficulty with the RD method was the failure to satisfactorily correct the

Table 1 Comparisons at baseline of Robert Debré and Saint Vincent de Paul groups

	Robert Debré group	Saint Vincent de Paul group	*P* value[3]
Number of feet	31	36	0.747
Number of feet by gender, boys/girls	23/8	25/11	0.667
Number of laterality affected feet, R/L	16/15	20/16	0.809
Number of moderate feet[1], %	10 (32)	10 (28)	0.782
Number of severe feet[1], %	12 (39)	17 (47)	0.782
Number of very severe feet[1], %	9 (29)	9 (25)	0.782
Age at start of treatment in d[2]	14 (2-30)	10 (1-45)	0.050

[1]According to the classification of Dimeglio;
[2]Median (range);
[3]Estimated with χ^2 test. R: Right; L: Left.

equinus of the calcaneus; most children did not keep the feet inside the shoes with the Denis-Browne bar, despite the adjustments made inside the shoes. In these cases, we had to use only a simple taping. The taping was not enough to maintain the correction achieved in the sessions, and this therefore led to a high frequency of surgeries. In the RD group, two moderate feet improved and became benign, but required a limited release at 15 mo of age due to the persistence of the equine ankle; perhaps in this case, an early hell-cord tenotomy could have prevented it, but we thought that it was due to the lack of efficacy of the containment available. With the SVP method, we more effectively maintain the corrections, and this aspect defines its success. Our result was not due to a reflection of experience with the RD method, however the success rate of the SVP was a consequence of the SVP protocol itself. In the first place, the bandage used on the rigid plate has a greater consistency than just a simple bandage; the strips of tape are more effective because we can achieve a better traction over the calcaneus and fix them to the plate. Second, the rigid plate has the advantage of being angled to 20 degrees on the back in order to continue the descent of the calcaneus. Third, the splints we used were very effective to maintain the correction, adapting completely to each foot. Despite frequent checks and the education received by parents, who assured us that they had followed the instructions received, some families reported that they were unable to achieve compliance post-treatment of the splinting, and these were the cases that relapsed. The relapse rates had a negative association with the applied approach. We want to show when the first relapse appeared in each group, and to determine which approach was able to maintain the correction for a longer period. In the recurrent cases that required surgery, the limited surgery was considered the best option at this stage of growth, and also because the physical therapy was able to maintain the foot with degrees of flexibility, allowing us to consider a more conservative surgery. The improvement achieved with the RD method by category was similar to that described by Souchet *et al*[23] in the evaluation of the results at the end of the conservative treatment. They compared the outcomes of conservative treatment to the at-birth classification, and found that 50% of moderate feet had a reclassification to benign; 100% of severe feet improved and became moderate; the very severe feet improved and 60% became moderate. Our results with the RD group also correlated with those obtained by Van Campenhout *et al*[25]. In this study, 75% of the cases require an operation in order to achieve a plantigrade foot. Rampal *et al*[26] report that more than half of the cases treated with the SVP method do not require surgery; we obtained similar results with our SVP group (58% without any surgery). Different teams had compared the results of the FPM with the PM. Richards *et al*[27] define their good results as plantigrade foot with or without heel-cord tenotomy; they reported good results for 72% of the feet treated with the PM. Although they obtained better results with the PM than with the FPM, the differences did not reach statistical significance. In our SVP group, we found good results in 75% of the feet. The results obtained by Chotel *et al*[28] with the FPM showed 17% of hell cord tenotomy and 21% of surgeries (correlates with our SVP group, 17% and 21% respectively). However, these authors found that 94% of the feet treated with PM were subjected to hell-cord tenotomy and 16% required surgery. In this study, the difference between FPM and PM was the type of surgery applied, and this was also due to the criterion of surgical indication in each team. They showed that the increase in the ratio of hell-cord tenotomy in the FPM (24%) managed to decrease the ratio of need for surgery (10%). Two studies of magnetic resonance imaging (MRI) showed similar corrections achieved with the PM and the FPM, except for the persistence of

Table 2 Comparison by category at 8 mo with respect to baseline[1]

Group	Severity at baseline, *n* (%)	Severity 8 mo after, *n* (%)		
		Normal	**Benign**	**Moderate**
Robert Debré	Moderate 10 (32)	4 (13)	6 (19)	0 (0)
	Severe 12 (39)	0 (0)	2 (7)	10 (32)
	Very severe 9 (29)	0 (0)	0 (0)	9 (29)
Saint Vincent de Paul	Moderate 10 (28)	8 (22)	2 (6)	0 (0)
	Severe 17 (47)	0 (0)	17 (47)	0 (0)
	Very severe 9 (25)	0 (0)	4 (11)	5 (14)

[1]$P = 0.001$ estimated with χ^2 test (Extended method).

the equine of the calcaneus on the feet treated with the FPM[29,30]. We must bear in mind that these feet had not suffered any hell-cord tenotomy at the time of the last MRI study, contrary to the feet treated with the PM, and all feet had been subjected to the hell-cord tenotomy before the last MRI study. There is still controversy as to which method is better, but it has been demonstrated that both methods achieve similar results; the FPM is as effective as the PM, and the differences between them do not reach statistical significance[8,31-33]. Regarding the learning curve of FPM approaches, we want to point out that learning and mastering the necessary skills to successfully apply the method takes time and requires knowledge. Thanks to its repetitive and extensive process, learning and experience are facilitated until reaching the stability that allows maintaining an adequate rhythm of work.

This study has limitations; the change from one treatment method to another resulted in an inherent bias in the techniques, since the onset of the SVP approach concurred with the learning curve of the latter by the physiotherapists. However, this did not appear to affect the comparability of groups, nor to the SVP group. In any case, the bias would have been produced against the SVP method, meaning that if our experience with the SVP protocol had matched the experience gained with the RD method, and if we had applied randomization, this would have generated even better results for the SVP group. Moreover, the rehabilitation department organization did not allow us to perform both treatment protocols simultaneously; for that reason, we carried out a retrospective study. Due to the high rate of surgery required for complete correction of idiopathic clubfoot using the RD method, we discarded the RD method in favor of the SVP method. In conclusion, the SVP method achieves prolonged correction of deformities more efficiently than the RD method; the best advantage of the SVP method is the greater number of cases without any surgeries. The SVP method should be regarded as a clearly beneficial option for the treatment of idiopathic clubfoot. Further studies are needed to corroborate or refute our results.

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Table 3 Comparison by category at two years of age to achieve a plantigrade foot

Group	Severity at baseline, *n* (%)	Very good[2]	Good[3]	Fair[4]	Poor[5]
Robert Debré	Moderate 10 (32)	6 (20)	2 (6)	2 (6)	0 (0)
	Severe 12 (39)	0 (0)	0 (0)	12 (39)	0 (0)
	Very severe 9 (29)	0 (0)	0 (0)	9 (29)	0 (0)
Saint Vincent de Paul	Moderate 10 (28)	10 (28)	0 (0)	0 (0)	0 (0)
	Severe 17 (47)	11 (30)	6 (17)	0 (0)	0 (0)
	Very severe 9 (25)	0 (0)	0 (0)	9 (25)	0 (0)

[1]$P < 0.001$ compared with χ^2 test (Extended method);
[2]Only physiotherapy;
[3]Heel-cord tenotomy;
[4]Limited posterior release;
[5]Complete release.

Table 4 Relapse rates

Group	Relapses Yes	Relapses No	Total
Saint Vincent de Paul	5	31	36
Robert Debré	5	26	31
Total	10	31	67

OR = 1.20. OR estimated with OR test. OR: Odds ratio.

Figure 3 Progression of clubfeet treated with the Robert Debré method. A, B: Derotation of the calcaneo-forefoot block; C: Simple taping closed with an cohesive bandage; D: Denis-Browne bar; E: Severe foot at the start of treatment; F: At 3 mo of age; G: The feet required posterior release; H: Plantigrade feet.

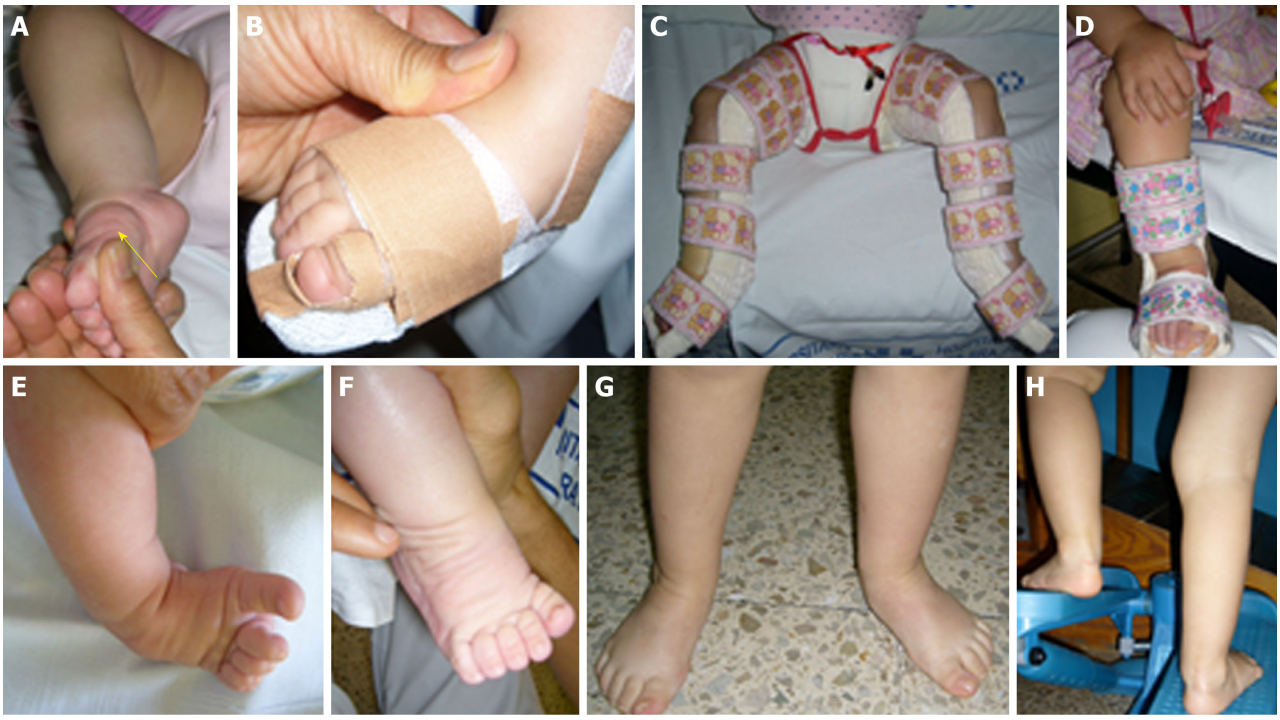

Figure 4 Progression of clubfeet treated with the Saint Vincent de Paul method. A: Reduction of the cavus (the forefoot is slightly supinated to align with the hindfoot); B: Performing the taping on plantar sole; C: Above-knee splints; D: Below-knee splint; E: Severe foot at the start of treatment; F: At 3 mo of age; G: Foot front view; H: Foot back view (plantigrade foot without surgery).

ARTICLE HIGHLIGHTS

Research background

Idiopathic clubfoot is a common birth defect that affects the musculoskeletal system. The initial treatment is conservative. The Functional physiotherapy method (FPM) is based on manipulations of the foot, bandages, splints, and exercises adapted to the motor development of the child to achieve a plantigrade and functional foot with the smallest surgical gesture possible. There are different approaches to the same method, but there is a lack of comparative studies between them. This study describes the results obtained with two approaches of this method [Robert Debré (RD) and Saint-Vincent-de-Paul (SVP)] revealing a significant difference in the ratio of surgeries before and after implementing the SVP method

Research motivation

The motivation behind this study was to detect the most effective FPM approach for maintaining corrections and reducing the rate of surgeries. This is very important because it would translate into saving resources, and would determine whether our institution should continue supporting the application of this method. The results of this study can encourage the implementation of FPM for us by other professionals who are seeking to both improve their interventions in clubfoot and reduce the ratio of surgeries.

Research objectives

The objective of this study was to compare two approaches of the FPM (RD and SVP) with regard to the improvement achieved and the frequency of surgery necessary to achieve a plantigrade foot, and to determine if the choice of one method or another would generate a substantial decrease in the rate of surgeries of clubfoot.

Research methods

A retrospective review of the therapeutic outcome was carried out for a series of 71 idiopathic clubfeet on 46 children born between February 2004 and January 2012. Data were taken from the medical records. The clubfeet were evaluated and classified according to severity by the Dimeglio-Bensahel scale; we included moderate, severe and very severe feet. Thirty-four feet were treated with the RD method, and 37 feet with the SVP method. The outcomes at a minimum of two years were considered as very good (by physiotherapy), good (by percutaneous hell-cord tenotomy), fair (by limited surgery), and poor (by complete surgery). Comparisons between treatments were performed with the χ^2 tests for nominal variables, and U test for numerical ones. The OR test was used for relapse rates. A two-tailed P-value ≤ 0.05 was considered statistically significant.

Research results

Complete release was not required in any case; limited posterior release was done in 23 cases (74%) with the RD method and 9 (25%) with the SVP method ($P < 0.001$). The percutaneous heel-cord tenotomy was done in 2 feet treated with the RD method (7%) and 6 feet (17%) treated with the SVP method ($P < 0.001$). Six feet in the RD group (19%) and twenty-one feet (58%) in the SVP group did not require any surgery ($P < 0.001$). The Dimeglio-Bensahel scale is useful for reflecting the severity of the deformity, and for analyzing the results category by category.

Research conclusions

Our hypothesis that the SVP method could achieve prolonged correction of deformities more efficiently than the RD method, as well as decrease the rate of surgeries, was confirmed in this study. The best advantage of the SVP method was the greater number of cases without any surgeries. No new methods were proposed in this study, but we would like to highlight that the SVP method is a clearly beneficial option for the treatment of idiopathic clubfoot.

Research perspectives

This study helped emphasize the importance controlling the equine of the calcaneus to avoid the need for surgery, and showed the efficacy of the FPM (the physiotherapy achieves a flexible, functional and painless clubfoot, and substantially reduces the need for surgery). The results obtained correlate with the initial severity of the deformity and with the protocol applied. The success of the treatment is based on two basic pillars: the adherence of parents to treatment and team training. It is essential to inform, educate and train the family, accompanied by a follow-up throughout the growth. We believe that it is necessary to carry out a future prospective investigation applying the SVP method with long-term follow-up. It is important to note that the current results obtained by different teams with the FPM correlate with those reported in the literature of the Ponseti method, and their differences do not reach statistical significance.

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CASE REPORT

Bilateral sequential femoral neck stress fractures in young adult with HIV infection on antiretroviral therapy: A case report

Saisunder Shashank Chaganty, Deeptiman James

ORCID number: Saisunder Shashank Chaganty (0000-0001-7447-1591); Deeptiman James (0000-0003-4712-613X).

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Saisunder Shashank Chaganty, Faculty of Medicine and Dentistry, University of Plymouth, Plymouth PL6 8BU, United Kingdom

Deeptiman James, Assistant Professor, Department of Orthopedics, Christian Medical College, Vellore, Vellore 632004, Tamil Nadu, India

Corresponding author: Saisunder Shashank Chaganty, MBBS, Medical Student, Faculty of Medicine and Dentistry, University of Plymouth, John Bull Building, 16 Research Way, Plymouth Science Park, Plymouth PL6 8BU, United Kingdom.
saisunder.chaganty@students.plymouth.ac.uk
Telephone: +0044-7952831648

Abstract

BACKGROUND

Femoral neck stress fractures are rarely encountered among young adults and are often associated with either repetitive excessive loading or underlying bone pathology. Preliminary research has indicated human immunodeficiency virus (HIV)/antiretroviral therapy (ART) as predisposing agents to osteopenia and osteoporosis related complications. We report a case of HIV/ART induced insufficiency fracture in a resource limited setting in Central India. Our aim is to increase awareness and promote screening of HIV/ART related osteopenia and osteoporosis in order to prevent catastrophic orthopaedic complications.

CASE SUMMARY

A 35-year-old HIV positive male presented with a stress fracture of left femoral neck. The patient was on ART and reported no comorbidities. He went on to be successfully managed surgically. However, during work-up osteopenia of the contralateral proximal femur was recognised using Singh's Index. Six months post-op the patient presented with right-sided femoral - neck stress fracture. At this stage the patient was nonconcordant with ART and denied surgical fixation.

CONCLUSION

In the absence of co-morbidities, several mechanisms of HIV/antiretroviral therapy may have played a role in predisposing our patient towards such a presentation. We recommend routine screening all HIV-infected patients for osteopenia, especially in younger individuals. In low resource settings and district hospitals, pelvis radiograph & Singh's index can be used for screening.

Key words: Bilateral femoral neck stress fracture; Human immunodeficiency virus-antiretroviral therapy related osteopenia; Osteoporosis in resource limited setting; Human

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immunodeficiency virus related fragility fractures

Core tip: Stress fractures of the femoral neck among young adults are extremely rare. They usually result from either fatigue or predisposing conditions that give rise to weakened bones. We present a case of bilateral sequential femoral neck stress fractures in a young adult with human immunodeficiency virus (HIV) infection on antiretroviral therapy (ART) in the absence of comorbidities. Several mechanisms of HIV/ART induced osteopenia and osteoporosis are indicated to have played a role in predisposing our case towards such a presentation. Our case study adds to the paucity of evidence exploring the association between HIV/ART and reduced bone mineral density. Our case highlights the need for screening and prophylactic treatment for osteopenia and osteoporosis coexisting with HIV infection and ART.

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INTRODUCTION

Evolution of antiretroviral therapy (ART), wide coverage of national acquired immunodeficiency syndrome (AIDS) policies and advances in management of opportunistic infections has enabled human immunodeficiency virus (HIV) infected individuals to live longer, stay healthier and enjoy better quality of life. However, HIV infection and ART have both been implicated in osteopenia, which can lead to catastrophic orthopaedic complications[1-3]. Limited access to healthcare and higher disease prevalence noted in resource limited areas in low-middle income countries (LMICs) are reported compounding factors[4]. Despite the huge disease burden of HIV infection as well as osteoporosis across the globe, minimal evidence exists regarding these two coexisting conditions, associated complications, screening methods and prevention. Limited awareness and lack of screening for HIV and ART related orthopaedic complications further add to the conundrum[5,6].

We present a rare report of bilateral sequential femoral neck stress fracture nonunions, evaluated and managed in a resource-limited secondary level rural mission hospital located in Central India. Our aim is to enhance awareness and highlight the need for screening, prophylactic and definitive treatment for HIV and ART related osteopenia and associated catastrophic orthopaedic complications, which can be implemented even in district hospital level healthcare setup.

CASE PRESENTATION

Chief complaint

A 35-year-old male patient presented to the orthopaedic outpatient clinic with a three months history of limp on left side.

History of present illness

Patient was unable to bear weight on the left side. He was able to walk with a walking stick and had to put full weight in the right leg. There was no antecedent history of significant trauma. Patient was unemployed and was not involved in any heavy or strenuous activity. There was no history of substance abuse or steroid intake. There was no pain on the right side.

History of past illness

Patient had been diagnosed with HIV infection more than two years ago. He has been getting the first line National AIDS Control Organization regime of AZT (Zidovudine) and 3TC (Lamivudine) for the last two years from the nearest ART centre, located more than 100 kilometres away[6]. History revealed poor ART compliance. Upon

enquiry about the delay of three months in seeking treatment, patient alleged stigma related refusal of treatment elsewhere due to his seropositive status. He did not have any medical comorbidity.

Physical examination upon admission

Upon examination, the patient was comfortable and alert at rest, Patient's weight was 74.6 kg (BMI 26.4 kg/m^2). General physical examination was normal.

He had an externally rotated left lower limb, telescopy and positive Trendelenburg sign with antalgic, assisted gait pattern. No abnormalities were noted on the right lower limb.

Laboratory examinations

Serum 25(OH) vitamin D level (24.6 ng/mL), phosphorus, alkaline phosphatase (113 IU/L), renal function and liver function tests and erythrocyte sedimentation rates were within normal limit. Screening for hepatitis B and hepatitis C was negative.

Provision for bone densitometry-FRAX tool was not available locally and could not be administered due to the patient's financial constraints. CD4 count done within the last two months was 350/mm^3.

Imaging examinations

Displaced fracture neck of left femur with break in Shenton's line, characteristic opening out on the tension side, varus settling of distal fragment and established non union was noted on the radiographs (Figure 1). Significant osteopenia (break in primary tensile trabeculae; Singh's grade III) was noted on the right proximal femur. (Figure 1). LS spine x-rays were normal

Computed tomography and magnetic resonance imaging scan facilities were not performed due to local inaccessibility and patient's financial constraints.

FINAL DIAGNOSIS

Based on clinical presentation, physical examination, relevant radiographs and laboratory work-up, most alternative causes of missed fracture or secondary osteoporosis were excluded. Non-union of left Femoral neck stress fracture was deemed the final diagnosis.

TREATMENT

Operative technique

Patient underwent open reduction of left femur neck fracture, valgus osteotomy of the left proximal femur and fixation with dynamic hip screw and additional derotation screw (Figure 2). Patient was positioned supine on a fracture table. Open reduction was done - through a lateral Hardinge approach and confirmed under image intensifier. Fracture was fixed with a 130° dynamic hip screw (Richard's screw) applied at 110° of neck shaft angle and augmented single partially threaded 6.5 mm cancellous screw.

A twenty degree lateral closing wedge osteotomy performed at the level of lesser trochanter and the distal femur fragment was valgised and attached to the dynamic hip screw barrel plate to gain a valgus neck shaft angle. .

Postoperative management

Partial weight bearing with axillary crutches was initiated on second postoperative day progressing to full weight bearing after two weeks. Postoperative convalescence was augmented with dietary and pharmaceutical calcium and calcitriol granules. Bisphosphonate was delayed as they are contraindicated in view of osteotomy and fracture. Radiological confirmation of union and return to prefracture functional status was confirmed at the three months review (Figure 3).

OUTCOME AND FOLLOW-UP

At six months follow-up, patient complained of limp and inability to weight bear on the nonoperated (right) limb for one-month duration. There was no history of significant trauma or heavy, strenuous activity. He was able to walk independently in the intervening period. Patient had noted slight pain and 'pop' over the right hip area following a mild twisting injury.

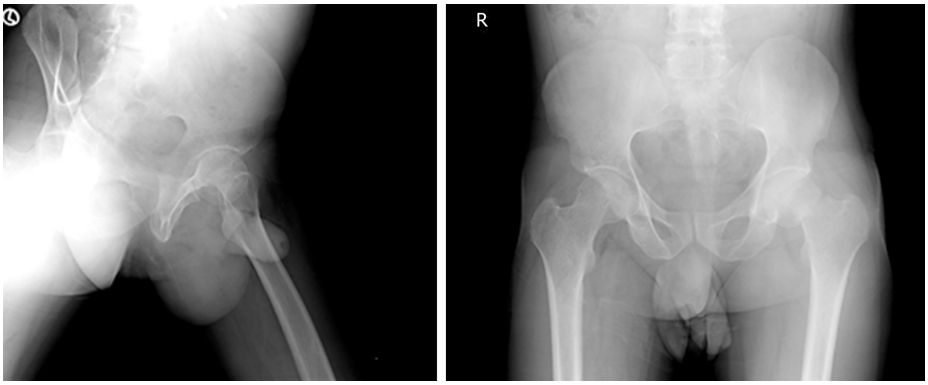

Figure 1 Preoperative anteroposterior and lateral X-rays demonstrating transcervical stress fracture of left femoral neck, with Singh's grade III osteopenia noted on the right proximal femur.

Clinical examination revealed external rotation deformity and telescopy of the right proximal femur. Characteristic features of stress fracture non-union of the right neck of femur were confirmed on radiograph (Figure 4). His CD4 count was 180/mm^3. Patient was offered surgical fixation of the right femoral neck but declined treatment.

DISCUSSION

Stress fractures of the femoral neck among young adults are extremely rare. They can result from fatigue or secondary to repetitive excessive force exerted over a prolonged duration on normal bone architecture. In young people, such mechanical overloading can lead to excessive bone resorption and temporary ischemic disruption with resultant impaired bone remodelling. This leads to weakening of the bone structure and microfractures. Relentless stress leads to complete fracture[7,8].

Diagnoses of stress fractures are often missed for several weeks due to lack of symptoms and absence of history of traumatic event. Besides, incomplete femoral neck stress fractures are difficult to diagnose in plain radiographs. Though complete fractures are easily identified, their rarity as well as physician's low index of suspicion often leads to delayed diagnosis and treatment[1,7].

Sometimes, coexisting metastatic disease or metabolic conditions that contribute to weakened bones can aid in screening and diagnosing stress fractures among young adults. Associated infective diseases such as hepatitis B and hepatitis C infection, severe renal impairment, inflammatory arthritis, adrenal insufficiency, corticosteroid excess, alcohol abuse and vitamin D deficiency are often cited as contributory factors[2,9]. In our case, laboratory investigations and performance status of the patient ruled out the aforementioned conditions.

Another avenue of enquiry would be to consider stress fractures as noted in osteoporotic bones (as in postmenopausal osteoporosis or senile osteoporosis) that result from normal stress exerted on weakened bone. Yet, osteoporosis is primarily a disease of the elderly and correlates inversely with the peak bone mass noted in young. Recent evidence has postulated the role of HIV infection and ART, especially Tenofovir, Zidovudine and almost all protease inhibitors as a probable cause of osteopenia, osteoporosis and occasionally, osteonecrosis of the femoral head[2,5,10,11].

Femoral neck stress fractures associated with HIV infection or ART are rare. Even fewer reports of femoral neck stress fracture exist in current literature. Using radiographs with Singh's Index we were able to classify osteopenia in this case, though ideally access to bone mineral density measurement and application of FRAX tools would be preferred. We hereby review the aetio-pathophysiology for HIV/ART induced secondary osteoporosis.

HIV and osteoporosis

Since the widespread availability of ART in 1996, several studies have reported an increased prevalence of osteopenia in HIV-infected patients. A cross-sectional analysis by Knobel *et al*[12] revealed osteopenia was present in 25% of healthy adults and in 67.5% of HIV-infected patients; whilst osteoporosis was present in 5% of healthy adults and in 21.2% of HIV-infected patients. Some studies have blamed this increased prevalence as statistically insignificant and biased by confounding covariables such as alcohol abuse, heroin use, glucocorticoid excess and hypogonadism[13]. A meta-analysis from Triant *et al*[2] reported a three times higher risk of

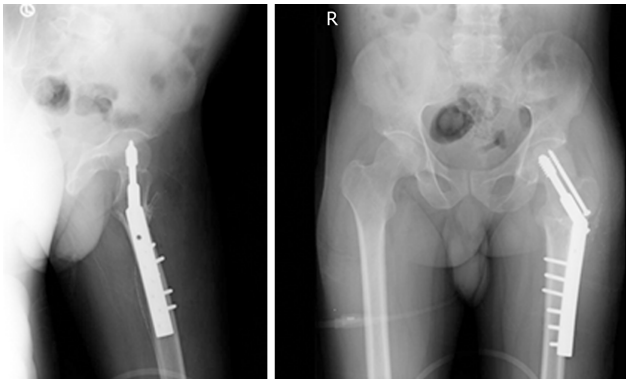

Figure 2 Postoperative AP and lateral radiographs of left proximal femur valgus Osteotomy demonstrating sliding hip screw fixation.

osteoporosis among HIV-infected individuals compared to the general population and an even higher risk of osteoporosis associated with ART.

Viral induced systemic inflammation and direct viral mechanisms are alluded to as mediators of HIV-mediated osteoporosis. Specifically, elevated inflammatory cytokines such as tumor necrosis factor alpha in conjunction with HIV proteins Vpr/gp120 (pro-osteoclast) and p55-gag (anti-osteoblast) contribute to a state of enhanced bone resorption without a commensurate rise in bone formation[5,14]. T-cell activation leading to osteoprotegerin ligand mediated osteoclastogenesis, direct inhibition of osteoblasts and activation of osteoclast leading to disturbance of bone resorption/formation coupling are other proposed theories (Figure 5A).

ART and osteoporosis

More convincing evidence is appreciated when considering the role of ART in predisposing for osteoporosis. Multiple longitudinal studies have generally shown a 2%–6% loss of bone mineral density after 48–96 wk of therapy, regardless of the type of ART initiated[12,15]. This is presumably due to a "catabolic window" phenomenon wherein the ratio of serum markers of bone resorption to bone formation is elevated during the first six months after ART initiation. Following this, a longer period of bone mineral density stability, particularly as the patient regains weight with commencement of ART, has been observed[11,16].

Mechanisms of ART-induced osteoporosis remains largely speculative (Figure 5B). For Tenofovir, a mechanism has been suggested wherein alterations in renal phosphate handling results in hypophosphatemia and osteomalacia. These effects are enhanced during concomitant vitamin D deficiency[14,17]. Protease inhibitors lead to reduction in RANKL: Osteoprotegerin ratio leading to recruitment and activation of osteoclasts and inhibition of osteoblasts. Rate of bone resorption associated with initiation of ART has been found comparable to that in the postmenopausal period. While the rate of resorption is believed to decrease after one to two years of ART initiation, return of bone mineral density to pre-ART status has not been found[11,17]. Nucleoside reverse transcriptase inhibitors such as Zidovudine and protease inhibitors are associated with vitamin D deficiency as well as altered vitamin D metabolism[11,18].

Recommendations

Our patient with long standing HIV infection with suspected ART noncompliance and with a calcium deficient diet was susceptible to osteopenia, which lead to this catastrophic event. No screening program for osteopenia exists under the National AIDS Control Organization guidelines. Lack of awareness of osteopenia and osteoporosis coexisting with HIV infection and ART could have contributed to the delay in treatment. Review of literature to evaluate the incidence and cause of bilateral femoral neck stress fractures revealed four to nine times higher risk of fractures among young adults with HIV infection and on ART.

Current AIDS control policy and ART policy does not advocate routine screening for osteopenia in HIV-infected individuals regardless if they are being treated with ART[6]. Limited evidence exists regarding prevalence of HIV/ART related osteopenia and associated catastrophic orthopaedic complications from low-middle income countries, even though more than two-thirds of the HIV-infected population hail from these geographic areas. We conducted a pilot study to assess awareness of HIV/ART related orthopaedic complications among healthcare providers. Twenty-five

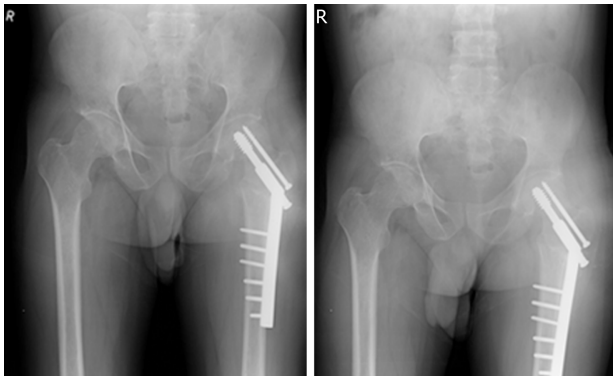

Figure 3 Three months postop follow up with sound union

orthopaedic surgeons and ten infectious disease specialists from three primary, nine secondary and one tertiary level healthcare centre revealed very poor awareness about HIV/ART related osteopenia and associated catastrophic orthopaedic complications.

In the given scenario, this case report highlights the need for increasing awareness among healthcare providers as well as establishes routine screening programs and prompt intervention to avoid osteopenia related catastrophic orthopaedic complications in HIV-infected patients. Novel methods are necessary to overcome resource limitation such as lack of bone density measurement and FRAX tools as they continue to remain inaccessible to the majority of rural and even urban populations. In this case, we used radiological evaluation with Singh's index for assessing osteopenia and osteoporosis to overcome the resource constraint at the healthcare centre. It may be substituted as an immediate need for screening of the susceptible population including HIV-infected patients on ART until a better accessible and affordable solution is developed[9].

CONCLUSION

In the absence of any additional discernible cause, HIV infection and ART must be considered as independent risk factors for incapacitating complications of osteopenia and osteoporosis. We recommend routine screening all HIV-infected patients for osteopenia, especially in younger individuals[4]. Radiological evaluation using Singh's index can be used as an alternative to DEXA screening in fiscally stressed settings. Indeed, a major concerted effort between government agencies, public health and district hospitals is essential for implementation. Yet only then, further research on cost benefit analysis can be performed.

Figure 4 Six months post operative follow up with stress fracture non union of the Right femur neck

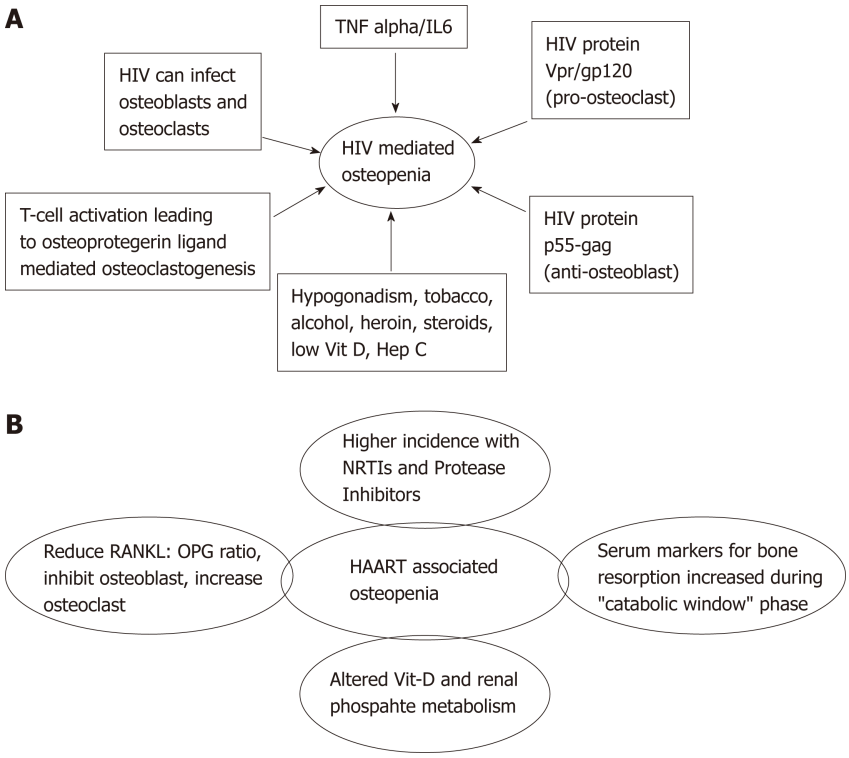

Figure 5 Suspected mechanisms. A: Human immunodeficiency virus and mediated osteopenia; B: Highly active antiretroviral therapy associated osteopenia. HIV: Human immunodeficiency virus; TNF alpha: Tumor necrosis factor alpha; HAART: Highly active antiretroviral therapy; Vit D: Vitamin D; Hep C: Hepatitis C; NRTIs: Nucleoside reverse transcriptase inhibitors ;OPG: Osteoprotegerin.

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CASE REPORT

Recurrent surgical site infection after anterior cruciate ligament reconstruction: A case report

Don Koh, Shi Ming Tan, Andrew Hwee Chye Tan

ORCID number: Don Koh (0000-0002-4777-675X); Shi Ming Tan (0000-0001-9572-6370); Andrew Hwee Chye Tan (0000-0003-4395-3322).

Author contributions: Koh D, Tan SM and Tan AHC designed, performed the research; analyzed the data; Koh D wrote the paper.

Informed consent statement: All study participants, or their legal guardian, provided informed written consent prior to study enrollment.

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Don Koh, Shi Ming Tan, Andrew Tan, Department of Orthopaedic Surgery, Singapore General Hospital, Singapore 169856, Singapore

Corresponding author: Don Koh, BSc, MBBS, Doctor, Department of Orthopaedic Surgery, Singapore General Hospital, 20 College Road, Academia Building Level 4, Singapore 169865, Singapore. don.koh@mohh.com.sg
Telephone: +65-62223322
Fax: +65-62248100

Abstract

BACKGROUND

Surgical site infections following anterior cruciate ligament (ACL) reconstruction are an uncommon but potentially devastating complication. In this study, we present an unusual case of recurrent infection of the knee after an ACL reconstruction, and discuss the importance of accurate diagnosis and appropriate management, including the issue of graft preservation versus removal.

CASE SUMMARY

A 33-year-old gentleman underwent ACL reconstruction using a hamstring tendon autograft with suspensory Endobutton fixation to the distal femur and an interference screw fixation to the proximal tibia. Four years after ACL reconstruction, he developed an abscess over the proximal tibia and underwent incision and drainage. Remnant suture material was found at the base of the abscess and was removed. Five years later, he re-presented with a lateral distal thigh abscess that encroached the femoral tunnel. He underwent incision and drainage of the abscess which was later complicated by a chronic discharging sinus. Repeated magnetic resonance imaging revealed a fistulous communication between the lateral thigh wound extending toward the femoral tunnel with suggestion of osteomyelitis. Decision was made for a second surgery and the patient was counselled about the need for graft removal should there be intra-articular involvement. Knee arthroscopy revealed the graft to be intact with no evidence of intra-articular involvement. As such, the decision was made to retain the ACL graft. Re-debridement, excision of the sinus tract and removal of Endobutton was also performed in the same setting. Joint fluid cultures did not grow bacteria. However, tissue cultures from the femoral tunnel abscess grew Enterobacter cloacae complex, similar to what grew in tissue cultures from the tibial abscess five years earlier. In view of the recurrent and indolent nature of the infection, antibiotic therapy was escalated from Clindamycin to Ertapenem. He completed a six-week course of intravenous antibiotics and has been well for six months since surgery, with excellent knee function and no evidence of any

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further infection.

CONCLUSION
Prompt and accurate diagnosis of surgical site infection following ACL reconstruction, including the exclusion of intra-articular involvement, is important for timely and appropriate treatment. Arthroscopic debridement and removal of implant with graft preservation, together with a course of antibiotics, is a suitable treatment option for extra-articular knee infections following ACL reconstruction.

Key words: Chronic surgical site infection; Anterior cruciate ligament reconstruction; Surgical site infection; Graft preservation; Enterobacter cloacae complex; Septic arthritis; Case report

Core tip: Chronic surgical site infections following anterior cruciate ligament (ACL) reconstructions are rare. Most infections occur in the acute or subacute post-operative periods. Astute clinic judgement and patient involvement are key when managing recurrent infections post-ACL reconstruction. Early intervention involving joint washout and debridement as well as commencing culture-directed antibiotic therapy are key. The decision for graft-sparing versus graft-sacrificing surgery remains controversial.

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INTRODUCTION

Surgical site infections following arthroscopic anterior cruciate ligament (ACL) reconstruction are uncommon. Current literature suggests the incidence rate to be around 1.7%[1-3] with almost all cases presenting in the acute or subacute post-operative periods[1,2,4-6]. Prompt diagnosis and treatment are necessary to avoid devastating consequences such as septic arthritis and its sequelae[7].

Various management protocols have been described in the literature with limited consensus on the best practice. Controversy remains with regards to the decision for graft-sparing versus graft-sacrificing surgery, especially for recurrent infections. Most authors, however, agree on the need for early irrigation and debridement as well as culture-directed antibiotic therapy[8].

While Gram-positive cocci are the most frequently encountered pathogens in surgical site infections, members of the Enterobacteriaceae family, namely Enterobacter cloacae complex (ECC), have also been reported[6,9]. ECC is a major cause of nosocomial and opportunistic infections, but its involvement in joint and graft infections is rare[6,9]. Its ability to form biofilms, produce cytotoxins and its intrinsic resistance to multiple beta-lactam antibiotics make this pathogen difficult to eradicate and treat effectively[10]. To date, there is a paucity of literature describing chronic or delayed surgical site infections after ACL reconstruction. We describe the first case of a delayed, recurrent infection involving both the tibial and femoral surgical sites which occurred at four and nine years respectively following an ACL reconstruction.

CASE PRESENTATION

Chief complaints

A 33-year-old gentleman who presents with a history of recurring ACL surgical site infections.

History of present illness

This patient underwent left knee ACL reconstruction, using a hamstring tendon autograft with Endobutton fixation to the distal femur and interference screw fixation

to the proximal tibia. He had an uneventful recovery and was able to return to his pre-injury activity level. Four years after his index surgery, he developed a left proximal tibial abscess over the interference screw site. He underwent incision and drainage of the tibial abscess. Intraoperatively, a remnant non-absorbable Ethibond suture was found at the base of the abscess. This remnant suture was removed and there was no infective extension into the tibia tunnel or graft. Tissue cultures grew ECC. He completed a two-week course of culture-directed oral Ciprofloxacin and was subsequently noted to be well during outpatient follow-up.

Nine years after the index surgery, this patient spontaneously developed left lateral thigh swelling (Figure 1). Magnetic resonance imaging (MRI) revealed a large rim-enhancing fluid collection form the femoral tunnel extending to the subcutaneous tissue with marrow enhancement adjacent to the femoral tunnel. An incision and drainage were performed and initial wound cultures yielded no bacterial growth. This was complicated by persistent discharge and the formation of a sinus tract over the lateral thigh wound. Patient subsequently presented to our clinic seeking a second opinion.

Physical examination

Clinical examination revealed a discharging sinus over the left lateral thigh wound. He was afebrile, ambulated well, demonstrated good range of motion in the left knee and anterior drawer test was negative. There was no clinical evidence suggesting septic arthritis.

Laboratory examination

Laboratory investigations revealed elevated C-reactive protein (CRP) of 17.8 mg/L but normal erythrocyte sedimentation rate (ESR) of 1.0 mm/hr as well as total white cell count (TW) of 5.70 × 10^9/L.

Imaging examination

Repeat MRI showed a fistulous communication between the lateral thigh wound extending toward the femoral tunnel with early features of osteomyelitis with no evidence of graft infection (Figure 2).

FINAL DIAGNOSIS

Recurrent surgical site infection after ACL reconstruction.

TREATMENT

The patient was counselled extensively for further debridement and understood the potential need for graft removal should there be evidence of intra-articular involvement. Arthroscopic exploration and washout as well as lateral thigh wound re-debridement, wound exploration, excision of sinus tract and removal of Endobutton was performed. Intra-operatively, the graft was noted to be in tact with no evidence of infection within the knee (Figure 3). In addition, the sinus tract led to the Endobutton site. The tract was excised in its entirety and the Endobutton removed. Underlying bone was curetted and noted to be healthy.

OUTCOME AND FOLLOW-UP

Tissue cultures from the sinus tract grew ECC, demonstrating similar antibiotic sensitivities to the tibial abscess cultures done in 2012. Joint fluid cultures, however, did not grow any organisms. Antibiotic therapy was escalated to Ertapenem in view of the recurrent nature of the infection. He completed a six-week course of antibiotics. Six months after surgery, his wound had healed well (Figure 1) and the inflammatory markers had normalised (CRP 4.6 mg/L, ESR 2 mm/h and TW 5.51 × 10^9/L). He remains asymptomatic with excellent knee function and has returned to his normal activities.

DISCUSSION

Majority of post-ACL reconstruction surgical site infections occur acutely (< 2 wk after surgery) and sub-acutely (between 2 wk and 2 mos after surgery)[4,6,11]. Pathogens

Figure 1 Wound site throughout recovery. A: Showing pre-operative discharging sinus; B: Post-operative wound showing wound dehiscence and discharge; C: Wound at six weeks after commencing intra-venous Ertapenem; D: Well healed wound.

commonly identified are skin commensals associated with peri-operative inoculation. Graft contamination can occur at any point during its harvest, preparation and re-introduction. Prior to a graft's re-incorporation, its lack of blood supply makes it a potential nidus for infection[4,12]. Another source of infection includes the surgical incisions or arthroscopic portals. The use of long instruments is also a potential source of intra-articular inoculation[4,12,13].

Mouzopoulos *et al*[4] postulated that late infections (> 2 mo) are often extra-articular in origin. The tibia and femoral tunnels are potential tracks for infections – bridging extra-articular infections with the intra-articular environment. The relatively superficial position of the interference screws and metallic endobutton, coupled with local tissue injury, predisposes to wound complications and infections. Judd *et al*[13] reported similar findings after noting eight out of eleven intra-articular infections having concomitant extra-articular wound complications - both of which having similar causative organisms on culture. McAllister *et al*[14] recommended the use of non-cannulated interference screws suggesting cannulated screws are a potential conduit for extra-articular pathogens.

In patients with extra-articular infection after ACL graft reconstruction, the risk of intra-articular involvement is ever present. Communication of the extra-articular with the intra-articular space via both tibial and femoral tunnels is a constant risk factor. The presence of foreign material such as sutures promote bacterial seeding and limits the effect of antibiotic clearance. In a recurrent case of infection, we suggest the removal of hardware with a view of graft removal should its integrity be compromised or if there is evidence of intra-articular involvement.

In our patient, the first surgical site infection involved the tibial interference screw site and occurred four years after the index surgery, with a subsequent infection five years later on the femoral site, centred around the Endobutton. Reaction to the implants and suture material and seeding of bacteria around these implants may be the cause of the infection on both occasions. Some studies have demonstrated increased risk of infection with hamstring autograft use[2,5,13]. Kim *et al*[2] hypothesised that the relative short length of the harvested hamstring autograft results in additional suture and hardware within the bone tunnels[5]. The increased foreign body load – both graft and hardware, increases both infection risks and foreign body reactions[2,5,15]. Even when sterile, foreign body reaction can stimulate inflammatory mediators, activating multinucleated giant cells. This produces a reaction of varying severity, ranging from oedema, sinus development to destruction to local structures[13,16].

In view of the chronic and indolent nature of the infection in our patient, incomplete eradication of the infection remained a concern. Our patient was counselled extensively pre-operatively and was agreeable for graft removal should

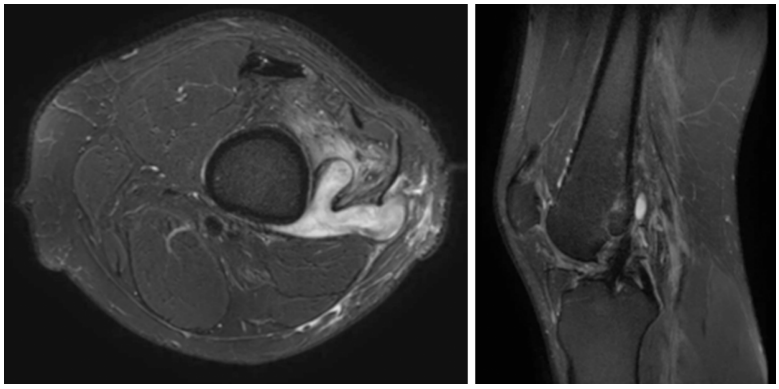

Figure 2 Magnetic resonance imaging knee showing a fistulous communication of a complex Y-shaped abscess in the lateral aspect of the distal thigh extending towards the femoral tunnel of the anterior cruciate ligament reconstruction.

there be a clinical indication.

The management of septic arthritis after ACL reconstruction remains controversial. Multiple management protocols have been proposed with limited consensus amongst authors. The main debate involves graft preservation versus graft removal in recurrent infections. Most authors to date opt for graft preservation treatment protocols involving long-term culture-directed antibiotics, arthroscopic irrigation and debridement as the first line of action. Should recurrent infections occur, this process is repeated until the complete eradication of infection. Proponents for graft preservation recommend such a protocol, boasting success rates of 85%[17]. This pathway is most agreeable amongst most authors and patients as it avoids morbidity from graft removal and the need for a staged reconstruction.

Opponents of graft preservation argue that the graft, if left in place, will remain a source of infection[1]. Kim *et al*[2] reported that an average of 1.9 procedures were needed for complete eradication of infection. Failure to eradicate the pathogen completely would result in repeated infections thereby requiring additional procedures. In patients with recurrent infections, risk of graft failure, loss of hyaline cartilage, damage to menisci and arthrofibrosis are complications that these group of authors strive to avoid[4,17,18]. Prompt removal of the infected graft and early reconstruction have also demonstrated good outcomes.

Intra-articular involvement can be ascertained through thorough history taking; detailed clinical examination and obtaining pertinent laboratory tests (*e.g.*, CRP, ESR and TW). Arthroscopy can be performed to visualise the intra-articular structures and study the ACL graft. Joint fluid can also be collected for culture and to ascertain bacterial antibiotic sensitivities. In our patient, it was only after establishing that this was an extra-articular infection that the decision made intra-operatively to proceed with debridement, graft preservation and removal of implant.

Interestingly, ECC was cultured from affected tissue on both occasions. While *Staphylococcus aureus* and coagulase-negative *Staphylococcus* are the commonest skin flora accounting for up to 88% of post-arthroscopic infections, there are reports involving gram-negative bacteria as well[6]. However, the literature detailing gram-negative pathogens such as ECC is scarce, limiting our understanding of its true incidence and pathogenicity[10]. ECC comprises of six species showing genetic relatedness to *E. cloacae* – namely *E. ludwigii, E. nimipressuralis, E. kobei, E. asburiae, E. cloacae* and *E. hormaechei*[19]. ECC are commensals of the gut, but is known to cause up to 5% of hospital-acquired sepsis[5,6]. The gastrointestinal tract as well as our skin are common sites whereby ECC is contracted[1,19]. Other sources include medical devices as well as intravenous products. It is a pathogen of increasing interest, due to its ability to form biofilms, secrete various cytotoxins as well as its innate resistance to beta-lactams due to AmpC beta-lactamase production[10,20]. These traits make it a potentially difficult pathogen to treat, especially after it has colonised an implant. This may explain the insidious onset of symptoms and chronic nature of this case.

The attributes of *E. cloacae* make complete eradication challenging, especially in setting of implant involvement. Unlike common pathogens (*e.g.*, *Staphylococcus aureus* and coagulase-negative *Staphylococcus*), *E. cloacae* presents as an indolent chronic infection presenting years after initial inoculation. The cause remains unclear. However, in the setting of prior infection, incomplete eradication is the most likely cause for recurrent surgical site infection. In addition, foreign body reaction played an important role in the initial swelling and effusion over the interference screw and

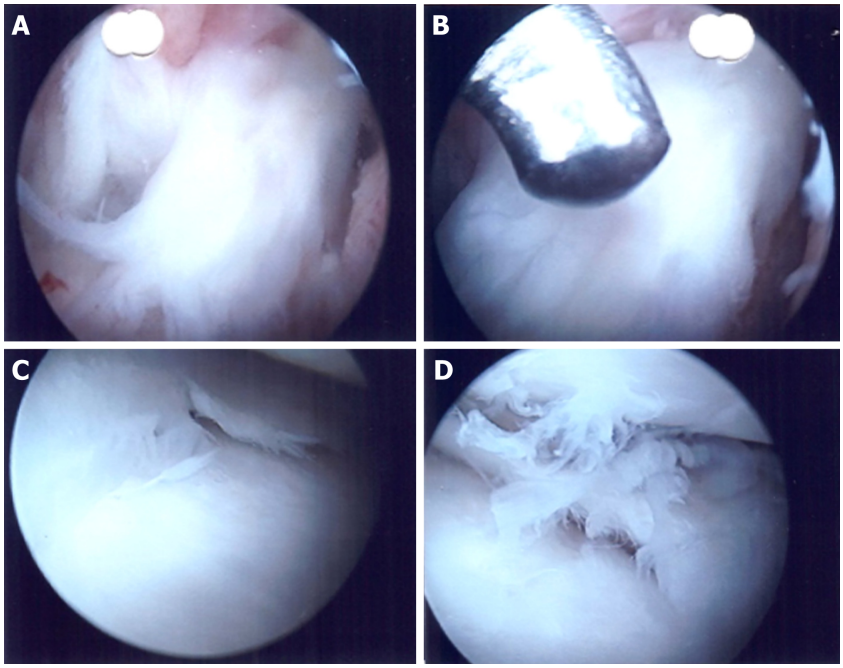

Figure 3 Arthroscopic images of the left knee joint. No evidence of purulent joint fluid. A, B: Anterior cruciate ligament graft was noted to be intact and healthy; C, D: Partial oblique tear of the medial meniscus as well as degenerative fraying of the lateral meniscus were also noted.

Endobutton sites. Non-involvement of the graft, noted during arthroscopy, suggests that the graft did not serve as a conduit for infection. The authors have demonstrated that favourable outcomes can be achieved with graft preservation when the graft is not compromised, even with chronic recurrent surgical site infections.

CONCLUSION

This report illustrates an unusual case of recurrent infections involving both the tibial and femoral surgical sites years after an ACL reconstruction. It emphasizes the importance of prompt and accurate diagnosis – especially the exclusion of intra-articular infection. The authors have demonstrated that in cases without graft involvement, debridement with graft preservation, removal of implants and a course of antibiotics may be a suitable treatment option.

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