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World Journal of Orthopedics (*World J Orthop*, *WJO*, online ISSN 2218-5836, DOI: 10.5312) is a peer-reviewed open access academic journal that aims to guide clinical practice and improve diagnostic and therapeutic skills of clinicians.

WJO covers topics concerning arthroscopy, evidence-based medicine, epidemiology, nursing, sports medicine, therapy of bone and spinal diseases, bone trauma, osteoarthritis, bone tumors and osteoporosis, minimally invasive therapy, diagnostic imaging. Priority publication will be given to articles concerning diagnosis and treatment of orthopedic diseases. The following aspects are covered: Clinical diagnosis, laboratory diagnosis, differential diagnosis, imaging tests, pathological diagnosis, molecular biological diagnosis, immunological diagnosis, genetic diagnosis, functional diagnostics, and physical diagnosis; and comprehensive therapy, drug therapy, surgical therapy, interventional treatment, minimally invasive therapy, and robot-assisted therapy.

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Evaluation and treatment of internal impingement of the shoulder in overhead athletes

Keith T Corpus, Christopher L Camp, David M Dines, David W Altchek, Joshua S Dines

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Abstract

One of the most common pathologic processes seen in overhead throwing athletes is posterior shoulder

pain resulting from internal impingement. "Internal impingement" is a term used to describe a constellation of symptoms which result from the greater tuberosity of the humerus and the articular surface of the rotator cuff abutting the posterosuperior glenoid when the shoulder is in an abducted and externally rotated position. The pathophysiology in symptomatic internal impingement is multifactorial, involving physiologic shoulder remodeling, posterior capsular contracture, and scapular dyskinesis. Throwers with internal impingement may complain of shoulder stiffness or the need for a prolonged warm-up, decline in performance, or posterior shoulder pain. On physical examination, patients will demonstrate limited internal rotation and posterior shoulder pain with a posterior impingement test. Common imaging findings include the classic "Bennett lesion" on radiographs, as well as articular-sided partial rotator cuff tears and concomitant SLAP lesions. Mainstays of treatment include intense non-operative management focusing on rest and stretching protocols focusing on the posterior capsule. Operative management is variable depending on the exact pathology, but largely consists of rotator cuff debridement. Outcomes of operative treatment have been mixed, therefore intense non-operative treatment should remain the focus of treatment.

Key words: Internal impingement; Overhead athlete; Partial rotator cuff tear; Scapular dyskinesis; Posterior capsular contracture; SLAP tear

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Core tip: "Internal impingement" is a term used to describe a constellation of symptoms which result from the greater tuberosity of the humerus and the articular surface of the rotator cuff abutting the posterosuperior glenoid when the shoulder is in an abducted and externally rotated position. The pathophysiology in symptomatic internal impingement is multifactorial, involving physiologic shoulder remodeling, posterior

capsular contracture, and scapular dyskinesis. Mainstays of treatment include intense non-operative management focusing on rest and stretching protocols focusing on the posterior capsule.

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INTRODUCTION

Overhead throwing athletes, in particular baseball players, often place unique and significant repetitive stresses across the shoulder at the extremes of the functional arc of motion^[1]. Over the course of a career, repeated loading can lead to osseous and soft-tissue changes and ultimately pathology of the shoulder girdle^[2-6]. The complex biomechanics of the elite thrower predispose them to multiple types of shoulder dysfunction, and these patients often serve as a clinical challenge for the sports medicine physician.

One of the most common pathologic processes seen in this patient population is shoulder pain resulting from internal impingement. "Internal impingement" is a term used to describe a constellation of symptoms which result from the greater tuberosity of the humerus and the articular surface of the rotator cuff abutting the posterosuperior glenoid when the shoulder is in an abducted and externally rotated position^[7].

The purpose of this article is to review the patho-anatomic features of symptomatic internal impingement, as well as review the current concepts involved with diagnosis and treatment of this condition.

HISTORICAL PERSPECTIVE

Posterior shoulder pain has been a debated topic dating back to the 1950s, as described by Bennett^[8]. At that time, he proposed that pain in the posterior shoulder was secondary to inflammation in the posterior capsule and inferior glenohumeral ligament due to triceps traction. This traction injury resulted in exostosis of the posteroinferior glenoid rim, which became known as the "Bennett lesion"^[8]. Bennett also described the presence of articular-sided posterosuperior rotator cuff tears in these same athletes^[9]. In 1977, Lombardo *et al.*^[10] further described ossification of the posterior glenoid rim in their description of an open approach for treatment of posterior shoulder pain in overhead throwing athletes.

In 1985, Andrews *et al.*^[11] described a series of overhead athletes with posterior shoulder pain that developed articular-sided posterosuperior cuff tears with concomitant SLAP lesions. Later, Jobe *et al.*^[12] reported on a series of overhead athletes with posterosuperior impingement associated with anterior instability. They

found that these patients did not respond well to standard subacromial decompression, but rather had success with anterior capsulolabral reconstruction. In addition, these patients often presented with a number of associated injuries including superior or inferior labral tears, rotator cuff tears, injury to the greater tuberosity or glenoid, or inferior glenohumeral ligament injury.

In 1992, Walch *et al.*^[7] reported a series of patients with impingement between the articular side of the supraspinatus tendon and the posterosuperior edge of the glenoid cavity, typically noted during maximal abduction and external rotation. At arthroscopy, these patients classically demonstrated a partial articular-sided rotator cuff tear. As a result, they described the "internal impingement" mechanism in which the undersurface of the posterior rotator cuff becomes entrapped between the labrum and the greater tuberosity in the abduction-external rotation position^[7,13].

PATHOPHYSIOLOGY AND

BIOMECHANICS

The biomechanical pathogenesis of internal impingement has been widely debated since its description. Some had posited that acquired anterior instability is the causative factor, while others have refuted this notion citing evidence suggesting no correlation between symptomatic internal impingement and anterior glenohumeral translation^[12,14-20]. More current thinking suggests that symptomatic internal impingement is multifactorial, involving physiologic shoulder remodeling, posterior capsular contracture, and scapular dyskinesis^[21,22].

Kinetic chain

The kinetic chain concept describes the coordinated motion that transmits energy in a synchronized fashion from the lower extremity, through the trunk, to the shoulder, and finally to the ball as it is released^[23-25]. Key elements of the kinetic chain are leg strength, body rotation, core strength, scapular position and motion, and shoulder rotation. Inflexibility, weakness, and imbalance of any point in the kinetic chain can create a situation where the arm lags behind the legs and trunk, placing the throwing shoulder in a vulnerable position, increasing stresses about the shoulder and leading to injury.

This concept is vital to the throwing motion and has been broken down into six distinct phases which comprise the throwing cycle. The first three phases consist of the wind-up, early cocking, and late cocking phases. These phases account for the bulk of the total time spent in the throwing motion (approximately 1.5 s), and they allow proper positioning of the lower extremities, core, and arms in preparation for release of the ball^[15,26,27]. In the late cocking phase, the shoulder is placed in abduction (90-100 degrees) and maximal external rotation (170-180 degrees), the position commonly associated with internal impingement^[7]. The fourth phase, the acceleration phase, is the phase in which the greatest angular velocity change

is seen across the shoulder joint as the ball is propelled forward. This motion occurs at a rotational velocity of over 7250 degrees per second and is the fastest human motion to ever be recorded^[24,28-30]. It follows that this phase results in the most injuries, despite being the shortest phase (0.05 s). As the ball is released, the deceleration phase and follow through phase are completed which result in a distraction force across the joint as the arm is slowed (0.35 s).

Thrower's paradox

The "thrower's paradox" refers to the delicate balance between mobility and stability which allows pitchers to achieve a high level of function^[24,31]. To generate rational velocities upwards of 7000°/s, the arc of motion must be expanded to allow maximal external rotation of the shoulder. The normal arc of rotation of a healthy shoulder from maximal internal rotation to maximal external rotation is 180°^[31]. The arc of motion in a high-level throwing athlete is shifted posteriorly to allow for increased external rotation at the cost of decreased internal rotation by allowing increased clearance of the greater tuberosity over the glenoid during rotation^[2-4,21,23,25]. Increased external rotation is achieved by a number of shoulder adaptations that develop over time including increased retroversion of the humeral head and glenoid, and increased anterior capsular laxity^[1,2,4,5,17,32].

Andrews *et al.*^[33] and Bigliani *et al.*^[34] have suggested that glenohumeral joint laxity is a common finding among throwing athletes. Jobe *et al.*^[12,32,35] originally described "subtle instability", or microinstability, to define the acquired laxity and anterior translation of the humeral head that occurs with the arm in a maximally abducted and externally rotated position. At what point this laxity becomes pathologic is another matter of debate. Subtle instability is postulated to result from repetitive shear stresses during the cocking and acceleration phases and contributes to the development of labral tears and articular-sided rotator cuff tears. Paley *et al.*^[17] stated anterior instability is actually the most significant factor in the development of internal impingement.

Alternatively, some authors have posited that micro-instability of the shoulder actually protects against internal impingement^[3,7,21]. Cadaveric, magnetic resonance imaging (MRI), and arthroscopic studies have consistently shown that contact of the rotator cuff on the posterosuperior labrum is a normal, physiologic occurrence^[3,21,23,28]. This theory postulates that the abnormal laxity of the humerus relative to the glenoid actually prevents impingement between the greater tuberosity and superior glenoid.

The anatomic changes allowing for increased external rotation can also result in remodeling of the posterior soft tissues, leading to contracture of the posterior capsule and posterior band of the inferior glenohumeral ligament^[21]. This increased external rotation creates increased torsional and shear stress upon the biceps anchor and undersurface of the rotator cuff^[1,7,28,32,35,36].

Despite the need for increased laxity, adequate stability must be maintained to prevent symptomatic humeral head subluxation, often achieved through further posterior capsular contracture. When present, these alterations can contribute to internal impingement and lead to rotator cuff tears, labral tears, capsular injuries, chondral injuries, and biceps tendon pathology. These findings were confirmed in a cadaveric model by Grossman *et al.*^[26], who reported on a simulated posterior capsular contraction model which led to GIRD and posterosuperior translation of the humeral head during the late cocking, ultimately resulting in SLAP injuries. Each of these processes can lead to pain, decreased velocity, loss of control, and diminished endurance in the throwing athlete.

Clinically, loss of 15 degrees or more of internal rotation in the throwing shoulder compared to the non-dominant arm is commonly seen. The potential for injury increases once this threshold has been reached. Burkhart *et al.*^[21,37] have reported that shoulders with an internal rotation deficit > 25 degrees are at increased risk for development of SLAP lesions as a result of increased posterosuperior peel back on the labrum. In addition, Dines *et al.*^[38] have shown that throwers with ulnar collateral ligament insufficiency at the elbow demonstrated a significant amount of GIRD as compared to players without a history of elbow injury.

Scapular dyskinesia

Kibler^[39] has defined scapular dyskinesia as an alteration in the normal resting position of the scapula or an alteration in the normal dynamic scapular motion. The scapula serves as an important link in the kinetic chain. When the scapula is unable to effectively transmit energy from the trunk to the pitching arm or stabilize the shoulder properly, pitching mechanics become inefficient and pitching velocity can suffer. Pitchers will then compensate by recruiting other surrounding musculature and increasing stress across the shoulder joint^[39].

Scapular motion has been found to be more intricate than once thought. Instead of the proposed 2:1 ratio of humeral to scapular motion during forward elevation, recent studies have shown the scapula to have a more complex role in shoulder motion. Scapular motion is now defined in three planes: Internal/external rotation around a vertical axis, upward/downward rotation around a horizontal axis, and anterior/posterior tilt around a horizontal axis^[39]. The intricate scapular positioning is controlled dynamically by force couples generated by the trapezius, serratus anterior, latissimus dorsi, and rhomboid musculature. These muscular couples contract before rotator cuff activation, allowing the cuff to contract against a stable scapular base^[40].

Myers *et al.*^[41] have previously reported that throwers normally develop upward rotation, internal rotation, and retraction of the scapula during forward elevation of the humerus. Scapular dyskinesia may result from inflexibility or imbalances in periscapular muscles secondary to fatigue, direct trauma, or nerve injury^[25].

Table 1 Keys to diagnosing internal impingement

History	Shoulder stiffness
	Need for prolonged warm-up
	Decline in performance (loss of velocity of control)
Physical exam	Posterior shoulder pain in late cocking phase
	Posterior glenohumeral joint line tenderness
	Increased external rotation, decreased internal rotation
Imaging	Scapular dyskinesis
	Positive anterior relocation test
	Positive posterior impingement sign
	Bennett lesion (exostosis of posteroinferior glenoid rim)
	Sclerosis of greater tuberosity, posterior humeral head cysts, rounding of posterior glenoid rim
	Posterosuperior labral tears
	Partial-thickness articular-sided rotator cuff tears (supraspinatus, infraspinatus)

Table 2 Jobe's clinical classification of internal impingement^[35]

Stage	Presentation/symptoms
I : Early	Shoulder stiffness and need for prolonged warm-up, no pain with ADLs
II : Intermediate	Pain localized to the posterior shoulder in the late cocking phase, no pain with ADLs
III : Advanced	Similar symptoms to Stage II, but refractory to a period of adequate rest and rehabilitation

ADL: Activities of daily living.

In pitchers with poor scapulothoracic rhythm, there is a trend toward scapular internal rotation and protraction around the rib cage resulting from inflexibility or imbalances in the periscapular musculature^[41]. When the scapula is ineffective in stabilizing the shoulder, the rotator cuff is forced to over-compensate to stabilize the glenohumeral joint. These loads are then transmitted to the superior glenoid and the articular surface of the rotator cuff tendons and can lead to injury. This alteration in function is thought to be an independent factor in the development of internal impingement. It follows that scapular dyskinesis has been reported in up to 100% of patients with internal impingement^[40,42].

HISTORY AND PHYSICAL EXAMINATION

A thorough history is the first step in appropriately diagnosing internal impingement (Table 1). Throwers with internal impingement may complain of shoulder stiffness or the need for a prolonged warm-up. They may also note a decline in performance, including loss of control or decreases in pitch velocity. They may also describe posterior shoulder pain, especially in the late cocking phase. These complaints were outlined by Jobe^[35], who defined three stages in the clinical presentation of internal impingement. Stage I consists of stiffness and difficulty in warming up, but no complaints of pain. Stage II is hallmarked by the complaint of pain during the late cocking phase of the throwing cycle. Those patients that have recurrent pain after a period of adequate rest and rehabilitation are classified as Stage III (Table 2).

The classic presentation and physical exam findings in the throwing shoulder with internal impingement commonly consists of posterior glenohumeral joint line tenderness, increased external rotation, and decreased

internal rotation. Despite this common pattern, a complete and thorough physical exam is important to identify any other associated shoulder pathology. The exam should start with visual inspection. Inspection may demonstrate greater muscular development in the dominant extremity, but assessment for any muscular atrophy must be performed. The scapulae are evaluated for positioning, dyskinesis, and winging. The scapula may have a prominent inferior medial border, and the throwing shoulder may appear to sag inferiorly compared to the non-throwing shoulder. Next, the coracoid process, anterior and posterior joint lines, greater tuberosity, long head of biceps tendon, AC joint, and deltoid should be palpated for tenderness.

Rotational glenohumeral motion should be assessed with the arm at the side and at 90 degrees of abduction. Internal impingement typically leads to posterior shoulder tightness in the throwing shoulder, leading to a loss of internal rotation^[4,43]. This finding has been confirmed in a population of college baseball players with shoulder pain, who demonstrated a 10 degree loss of internal rotation in their throwing shoulder as compared to both their non-dominant shoulder and pain-free controls^[44]. Forward elevation and horizontal abduction should also be evaluated.

Strength examination focusing the rotator cuff should be completed in all patients with suspected internal impingement. Rotator cuff involvement can range from undersurface fraying, to partial articular-sided tears, to full-thickness tears. The most common tendon involved is usually the infraspinatus; therefore special attention should be paid to external rotation strength.

Special testing may include the relocation test, described by Jobe, in which the shoulder is placed in 90 degrees of abduction and maximal external rotation.

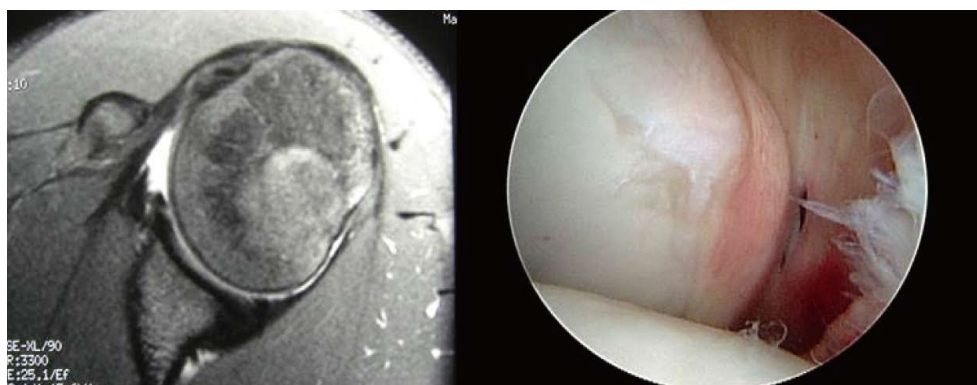


Figure 1 Magnetic resonance image of Bennett lesion and corresponding arthroscopic picture viewing posteriorly from anterosuperior portal.

While in this position, the humerus is loaded in both an anterior and posterior direction. While posterior directed force will provoke pain and impingement, anterior loads will relieve pain (opposite from the traditional findings in patients with anterior shoulder instability)^[35]. A second test, the posterior impingement sign, was described by Meister^[45] and involves placing the shoulder into 90 to 110 degrees of abduction, slight extension, and maximum external rotation. The provocation of deep posterior pain indicates a positive test and is highly correlated with undersurface tearing of the rotator cuff and/or posterior labrum.

In addition to the relocation test described above, a global stability exam is also important in the evaluation of internal impingement. The difficulty with stability examination in this patient population harkens back to the “thrower’s paradox” previously described. Many of these patients have adaptive laxity, which must be distinguished from pathologic laxity. Most often, the most important finding is the patient’s subjective sensation of shoulder subluxation during examination, which may occur while placing the patient in the position of apprehension. Additionally, a good examination under anesthesia at arthroscopy is vital for stability testing.

Lastly, examination of the posterosuperior labrum is an important component of the internal impingement exam, as labral tears in this location are common. Multiple physical exam maneuvers have been described for evaluation of the superior labrum and have shown high sensitivity for detection of tears, but none have shown high specificity for identification of superior labral tears^[46-48].

RADIOGRAPHIC EVALUATION

Radiographic evaluation should begin with standard shoulder radiographs, including internal and external rotation anteroposterior, scapular Y, axillary, and West Point views^[1]. Radiographs may be normal in the setting of internal impingement, but patients may display several radiographic findings in association with this pathologic process. Common radiographic findings include the “Bennett lesion” (exostosis of the posteroinferior glenoid rim), sclerosis of the greater tuberosity, posterior humeral

head osteochondral cysts, and rounding of the posterior glenoid rim^[8,9].

The mainstay of radiographic evaluation is MRI, which has a high sensitivity for capsular, labral, and rotator cuff pathology in the throwing shoulder. At our institution, we routinely perform noncontrast MRI, as it has been shown to have equivalent or superior sensitivity and specificity rates to MR arthrogram, when appropriate pulse sequences are utilized^[49]. Common MRI findings in patients with internal impingement include posterosuperior labral tears, partial-thickness articular-sided rotator cuff tears most notably at the junction between the supraspinatus and infraspinatus as they insert on to the humeral head, and cystic changes in the posterior aspect of the humeral head^[50,51]. Additionally, patients can display calcification at the scapular attachment of the posterior capsule (Bennett lesion), posterior capsular contracture and thickening at the level of the posterior band of the inferior glenohumeral ligament, and subchondral fracture and remodeling of the posterosuperior glenoid^[1] (Figures 1-3).

TREATMENT

Nonoperative management

Once a diagnosis of internal impingement is made, non-operative management should be recommended as the first line treatment. The proper non-operative treatment can be tailored based on stages set forth by Jobe discussed earlier^[35]. Patients in Stage I (those with complaints of stiffness and difficulty warming up but without localized posterior shoulder pain) should be prescribed a course of nonsteroidal anti-inflammatories and rest. Patients in Stage II (those with isolated posterior shoulder pain) usually require four to six weeks of rest and can also benefit from physical therapy^[1]. Physical therapy has been shown to be both therapeutic and protective against further injury in several studies^[21,22,31].

Given the pathophysiologic mechanism of internal impingement, therapy is focused on correction of aberrant shoulder range of motion and scapular dyskinesis. To that end, special attention should be paid to correction of GIRD through the “sleeper stretch” which allows posterior

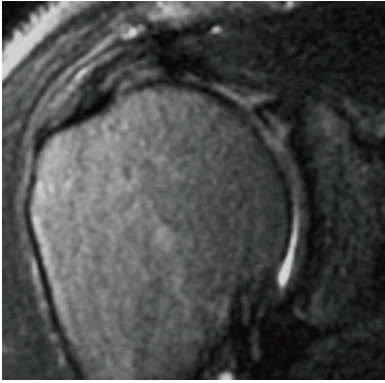


Figure 2 Magnetic resonance image of a Type 2 SLAP tear with concomitant partial thickness rotator cuff tear.



Figure 3 Partial thickness articular-sided tear of infraspinatus as viewed from posterior portal.

capsular stretching. In a study of high-level tennis players performing daily “sleeper stretch” exercises, patients were found to have significant increases in both internal rotation and total rotation, as well as a 38% decrease in the prevalence of shoulder problems^[21] (Figure 4). In addition, scapular stabilization exercises should be recommended. As the throwing motion involves the entire body through the kinetic chain, core strengthening and lower body strengthening must also be stressed simultaneously. Lastly, proper throwing mechanics should be enforced, especially for younger athletes.

Corticosteroid injections directed at the posteroinferior glenoid rim have been described^[52]. These injections have mostly been used for diagnostic, rather than therapeutic, purposes. To this point, no compelling data has been published to support injections for internal impingement and should be used judiciously due to the potential risk of permanent tendon damage.

Operative treatment

Due to the spectrum of pathology seen in internal impingement, multiple operative treatment options exist. Paley *et al.*^[17] published a series demonstrating > 80% incidence of concomitant articular-sided rotator cuff tear in professional overhead athletes with internal impingement. Often, this tear will be associated with an adjacent “kissing

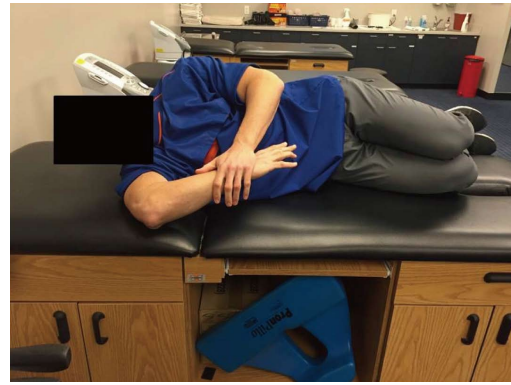


Figure 4 Demonstration of the “sleeper stretch”.

lesion” seen as a labral tear. Historically, treatment of these lesions has included debridement or repair and possible acromioplasty, with mixed results. Andrews *et al.*^[11] published a series of thirty-six overhead athletes with articular-sided partial tears of the supraspinatus that underwent arthroscopic debridement. They found 85% were able to return to their pre-morbid level of function, which they attributed to stimulation of tendon healing *via* debridement. Similarly, Sonnery-Cottet *et al.*^[20] performed arthroscopic debridement on twenty-eight tennis players with articular-sided partial tears and glenoid lesions. Seventy-nine percent were able to return to play, but 91% still had some persistent pain.

Historically, most surgeons have felt that lesions involving more than half of the thickness of a rotator cuff tendon should be repaired. Some authors support completion of these partial tears to aid in soft-tissue mobilization, followed by standard double-row repair^[53-55]. More recent data suggests that partial tears up to 75% in high-level throwing athletes should be debrided unless they involve the anterior cable of the rotator cuff. The high forces that throwing athletes generate subject the cuff to intense stress and threaten the repair integrity^[56]. In cases involving injury to the anterior cable, the anterior cable can be repaired and the partial thickness posterior cuff injury can be debrided. If partial tear completion and repair is indicated, a lateralized double-row repair as described by Dines *et al.*^[57], should be considered as it has shown favorable outcomes in professional overhead throwing athletes, allowing restoration of a more anatomic footprint.

Additional pathology seen in internal impingement includes SLAP tears, biceps tenosynovitis, and degenerative changes in the humeral articular surface^[7,58]. Meister *et al.*^[45] reported their results of twenty-two overhead athletes with internal impingement who underwent debridement of the rotator cuff, biceps, and labrum. A subset of patients also underwent arthroscopic removal of a Bennett lesion. Only 55% of the cohort had returned to their pre-morbid level at 6 years post-op. Neri *et al.*^[59] also evaluated a cohort of high-level overhead athletes who underwent Type II SLAP repairs at a mean of three years follow up and found that only 57% were able to



Figure 5 Type 2B SLAP tear s/p repair.

return to their pre-injury level of competition, while an additional 25% returned to sport but were limited by pain. These marginal results may be due to the higher demand elite overhead athletes place on their shoulders and higher expectations amongst the study group. In addition, Neri *et al.*^[59] showed that a statistically significant correlation did exist between the presence of partial-thickness rotator cuff tears and the inability to return to pre-injury level of play (Figure 5).

Andrews *et al.*^[60] have suggested that the poor results of debridement alone could be attributed to unaddressed subtle anterior laxity. As a result, they advocated for concomitant open anterior stabilization. They reported a 92% success rate at an average of close to three years postoperatively in a cohort of twenty-five athletes. Other studies have had less-optimistic results^[35].

Lastly, some authors have proposed osseous procedures to address the osseous changes which heavily contribute process of internal impingement. Riand *et al.*^[61] reported on humeral osteotomies to increase humeral retroversion in twenty patients who had continued pain after arthroscopic debridement. Eleven of the twenty patients were able to resume sports activities at the same level, and five were able to resume sports at a lower level.

CONCLUSION

Internal impingement is a complex pathologic process secondary to repetitive use in overhead athletes resulting in articular-sided partial-thickness rotator cuff tears and

SLAP lesions. The pathogenesis of internal impingement is multi-factorial. Understanding the etiology and pathogenesis will allow proper diagnosis and treatment.

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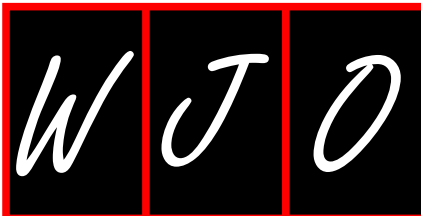
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Lessons learned from study of congenital hip disease in adults

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Abstract

Orthopaedic surgeons specialising in adult hip recon-

struction surgery often face the problem of osteoarthritis secondary to congenital hip disease (CHD). To achieve better communication among physicians, better treatment planning and evaluation of the results of various treatment options, an agreed terminology is needed to describe the entire pathology. Furthermore, a generally accepted classification of the deformities is necessary. Herein, the authors propose the use of the term "congenital hip disease" and its classification as dysplasia, low dislocation and high dislocation. Knowledge of the CHD natural history facilitates comprehension of the potential development and progression of the disease, which differs among the aforementioned types. This can lead to better understanding of the anatomical abnormalities found in the different CHD types and thus facilitate preoperative planning and choice of the most appropriate management for adult patients. The basic principles for improved results of total hip replacement in patients with CHD, especially those with low and high dislocation, are: Wide exposure, restoration of the normal centre of rotation and the use of special techniques and implants for the reconstruction of the acetabulum and femur. Application of these principles during total hip replacement in young female patients born with severe deformities of the hip joint has led to radical improvement of their quality of life.

Key words: Congenital hip disease; Low dislocation of the hip; Hartofilakidis classification; Dysplasia of the hip; High dislocation of the hip; Total hip replacement; Trochanteric osteotomy; Restoration of the normal centre of rotation; Femoral shortening; Patients' satisfaction

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Core tip: This review is based on the knowledge and experience acquired in the long course of the senior author's surgical practice on the complex problem of congenital deformities of the hip in adults.

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INTRODUCTION

Congenital deformities of the hip are the main cause of secondary joint osteoarthritis (OA). Depending on the severity of the deformity, undiagnosed patients or those treated insufficiently in infancy face the problem of OA earlier or later in their adult life.

Initially, before the introduction of total hip replacement (THR), we were treating adult patients with different types of osteotomies, without having enough knowledge of the pathological anatomy and the natural history of the underlying deformity. This explains why the results of osteotomies were not always satisfactory. Later, when we started to treat these patients with THR, we realised that the anatomy of the joint differs between patients, thus requiring adjustment of the surgical technique. Since then, the complex problem of congenital hip deformities became one of the main scientific interests of the senior author (GH). Lessons learned, in the long course of this experience, are summarised in this review article.

Lesson 1

The first lesson involves the understanding of the nature of the hip deformities present at birth. The terms used for these deformities were mostly misleading, causing misunderstandings and confusion. We have been concerned about the term "developmental dysplasia of the hip" (DDH) for two reasons: (1) The term "developmental" is not descriptive of the congenital origin of the deformity; and (2) An indiscriminate use of the term "dysplasia" is not in agreement with the variety of the underlying pathology. Therefore, we recommend the use of the term "congenital hip disease" (CHD) for the entire spectrum of related deformities. These deformities are congenital in nature and have the potential to develop.

Furthermore, we recognized that for better communication, planning of treatment and evaluation of the results of different treatments, a classification system of general acceptance must be used. Weinstein had classified CHD in infancy in three radiographic types: (1) dysplasia (inclination of the acetabulum with centralized ossification centre - Shenton's line intact); (2) subluxation (subluxed ossification centre - Shenton's line broken); and (3) complete dislocation (ossification centre outside the acetabulum) (Figure 1). Based on that classification, we identified in adults three different CHD types of increasing severity: (1) dysplasia; (2) low dislocation; and (3) high dislocation. Depending on the type, adjustment of our surgical technique during THR is required. In dysplasia, the femoral head is contained within the original acetabulum.

In low dislocation, the femoral head articulates with a false acetabulum that partially covers the true acetabulum. In high dislocation, the femoral head is migrated superiorly and posteriorly to the hypoplastic true acetabulum. The proximal part of the femur, in dysplasia, is normal. In low dislocation, the femoral neck is short, and shorter still in a high dislocation, with excessive anteversion. The diaphysis, in high dislocation, is hypoplastic with excessive narrowing of the femoral canal and has thin cortices (Figure 2).

Later, in a refinement of our classification system, we further subdivided low and high dislocation. Low dislocation was subdivided into B1 and B2 subtypes, when the false acetabulum covers more or less than 50% of the true acetabulum, respectively. High dislocation was subdivided into C1 and C2 subtypes, depending on the presence or the absence of a false acetabulum, respectively (Figures 3 and 4). The lesson learned by using this classification system is that "better comprehension of the pathologic anatomy and the specific characteristics of these hips, makes their THR reconstruction easier and more successful"^[1-9].

Lesson 2

The second lesson learned is that knowledge of the natural history of CHD facilitates comprehension of the potential development and progression of the disease, which differs among the three types. This can lead to better understanding of the anatomical abnormalities found in the different types, thus facilitating preoperative planning and choice of the most appropriate management for adult patients.

In our country, before the introduction of screening systems, the majority of infant dysplastic hips remained undiagnosed until the onset of symptoms, usually at the third decade of a patient's life. Degenerative changes progress slowly since that time and, usually, THR can be postponed until the age of 45-50 years. Patients with low dislocation, who had not received previous treatment in infancy, limp since childhood and experience pain later in their lives, usually at the age of 25-30 years. In these cases, degenerative changes develop within the false acetabulum and THR, usually becomes necessary earlier than in dysplastic hips. Patients with high dislocation and no previous treatment also limp since early childhood. Limping is more severe in patients with unilateral involvement. Natural history depends on the presence or absence of a false acetabulum. In patients with a false acetabulum pain starts early, usually around 30 years of age, while in patients without a false acetabulum pain starts much later, around the age of 40-45 years, as a consequence of muscle fatigue (Figures 5-7). In unilateral involvement, the leg-length discrepancy ranges between 4-10 cm and increases the disability of the patient. Also, the ipsilateral knee presents valgus deformity, sometimes severe, and the spine thoracolumbar scoliosis.

The indication for a THR not only depends on the degree of pain and disability, but also includes emotional

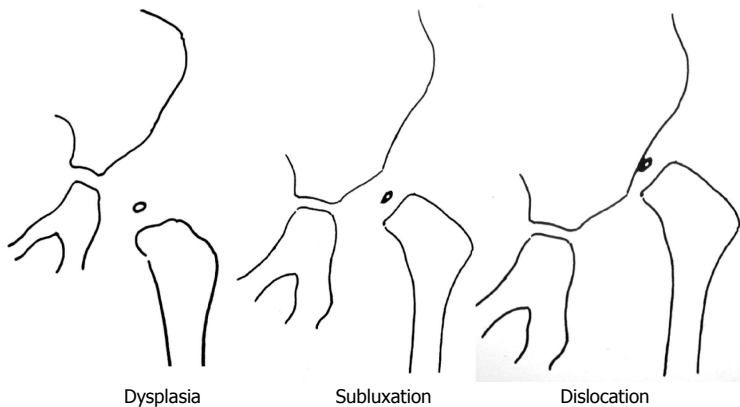


Figure 1 Drawings of the three types of the disease in infants.

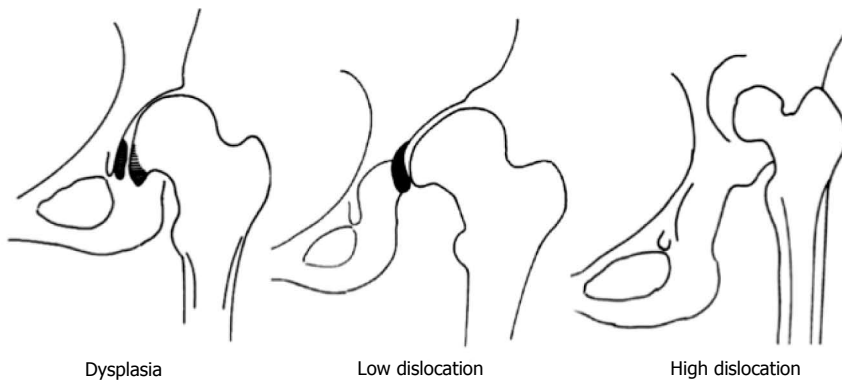
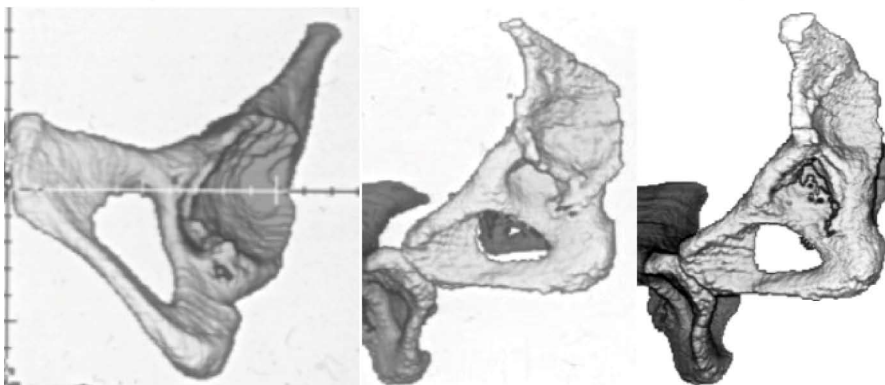


Figure 2 Drawings and three dimensional-computed tomography images of the 3 types of congenital hip disease in adults.

The black-colored areas in dysplasia represents the large osteophyte that covers the acetabular fossa and the medial marginal osteophyte of the femoral head (capital drop), and in low dislocation it is the inferior part of the false acetabulum that is an osteophyte which begins at the level of the superior rim of the true acetabulum.



parameters, given that these patients are young females with an active and productive life^[10,11].

Lesson 3

Another lesson learned is that THR is a difficult operation and should be performed by experienced surgeons and only when there is an absolute indication based on the patient's symptoms, psychological impact, clinical findings and the potential of disease development; "Not to early, not to late". Several technical details should be considered^[9,12-17].

Transtrochanteric approach: Wide exposure is essential for the reconstruction, especially of hips with low and high dislocation. The lateral transtrochanteric approach was introduced by Charnley, who suggested that it would facilitate the access to the joint and the reconstruction of the disturbed anatomy of the acetabulum and the proximal femur. Additionally, lateral reattachment of the trochanter increases the abductor lever arm and

minimises the reactive forces acting on the acetabulum. Four categories of the trochanteric reattachment are recognised: (1) reattachment at the original bed of trochanteric osteotomy; (2) distal reattachment in relation to its original bed, the trochanter having contact with the distal part of the original bed; (3) reattachment on the lateral femoral cortex in cases where the femoral neck was resected to the level of the lesser trochanter; and (4) reattachment proximal to its original bed (Figure 8). The ideal reattachment of the trochanter is the distal reattachment retaining contact with the distal part of the original bed. However, this is not always possible mainly due to shortened abductor muscles, an often small and malpositioned trochanter and a lengthened limb^[16].

The most common complication of trochanteric osteotomy is the non-union of the trochanteric fragment. Other complications include breakage and migration of the wires, heterotopic ossification and dislocation. We learned that the complications of this exposure were less

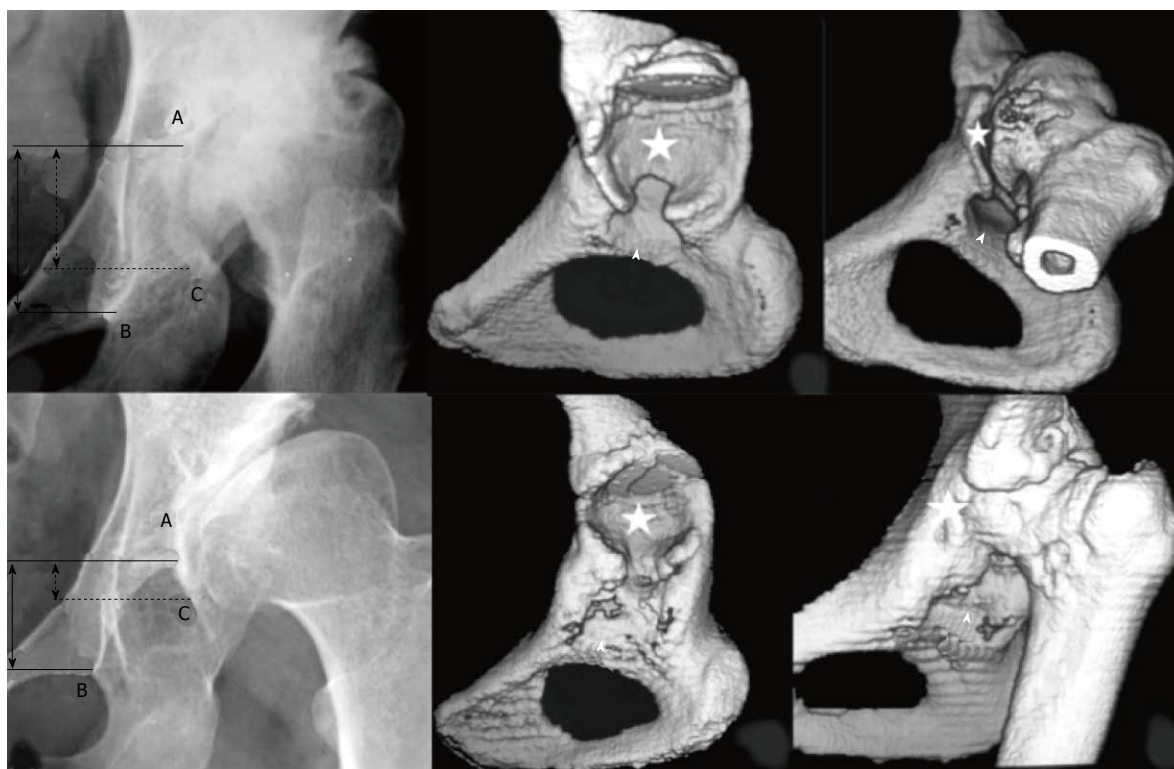


Figure 3 Images illustrating the two subtypes of low dislocation: B1 and B2. Three points must be recognised on radiographs: (A) the superior limit of the true acetabulum; (B) the inferior point of the teardrop; (C) the most inferior point of the false acetabulum. Three dimensional-computed tomography scans may help to determine the superior limit of the true acetabulum, when it is not clear in plain radiographs. Asterisks depict false acetabulum and arrowheads true acetabulum.

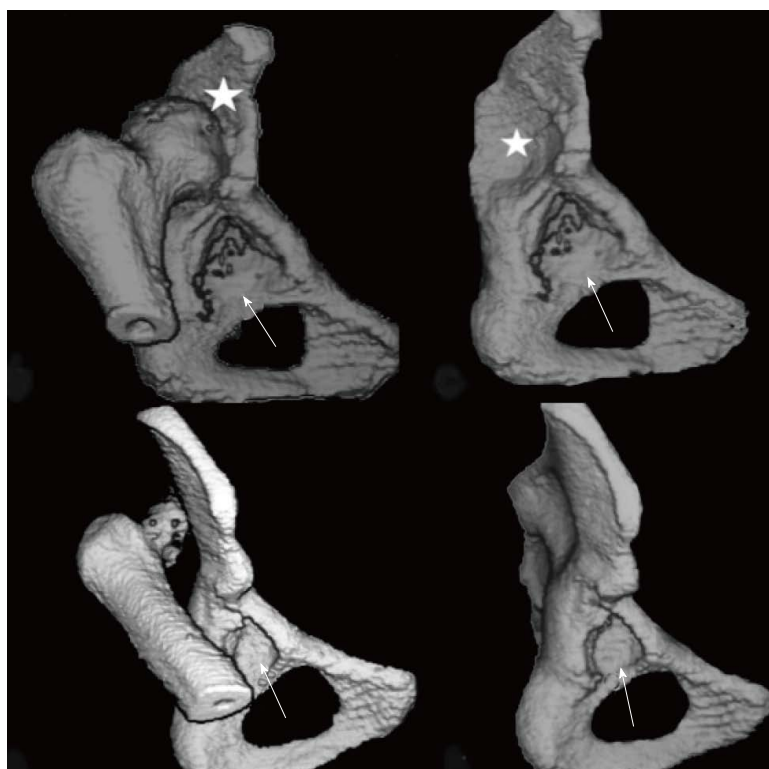


Figure 4 Three dimensional-computed tomography scans of the two subtypes of high dislocation. Arrows indicate the true and asterisks the false acetabulum.

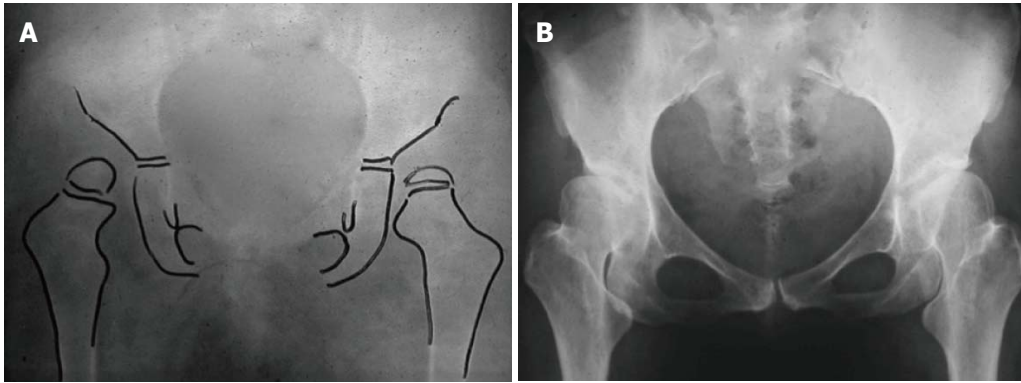


Figure 5 Development of dysplastic hips. A: At the age of 3 years, when the child was first seen by her physician. An abduction frame was applied for 6 mo; B: At the age of 35 years, the patient had the first symptoms, pain and limping, due to the development of secondary hip osteoarthritis.

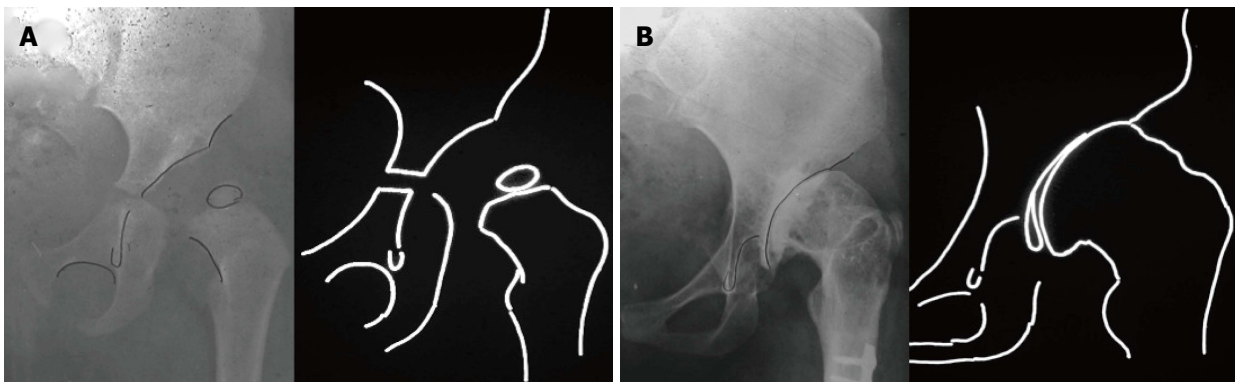


Figure 6 Radiographs and diagrams of a female patient with subluxation of the left hip in infancy, subsequently developed to low dislocation. A: At the age of 2 years; B: Image, when the patient was 37 years old.

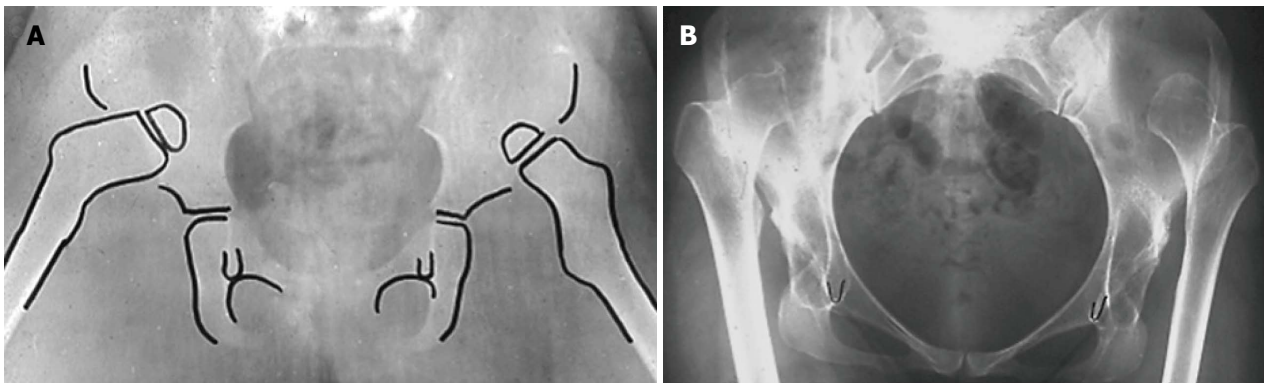


Figure 7 Images illustrating the case of a female patient with bilateral high dislocation. A: Radiograph at the age of 2 years; B: Radiograph, at the age of 33 years, when the patient was consulted for bilateral total hip replacements.

important than the benefits gained^[16].

Restoration of the normal centre of rotation of the joint: Placement of the acetabular component at the level of true acetabulum is essential for restoration of the hip biomechanics and improvement of survival of the prosthesis. However, it is not always possible to achieve bony coverage of the acetabular component at this level. Two alternative techniques have been used to solve this problem. When the reamed acetabulum

can provide at least 80% osseous coverage of the implant, we use an uncemented small 40-42 mm metal backed acetabular component. If this is not feasible, the cotyloplasty technique is an effective alternative. Cotyloplasty involves medialization of the acetabular floor through creation of a comminuted fracture of the entire medial wall, impaction of autogenous cancellous morselized bone grafts and implantation of a small, all-polyethylene (PE) implant, usually the offset-bore acetabular cup (Figure 9)^[3,9,14]. The main mechanical

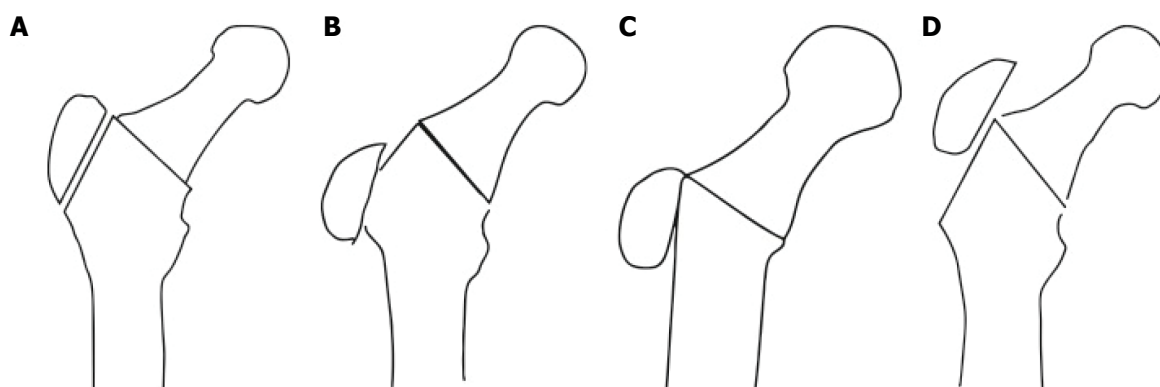


Figure 8 Drawings depicting. A: Reattachment of the trochanter at its original bed; B: Distal reattachment of the trochanter, retaining contact with the distal part of the original bed; C: Reattachment of the trochanter on the lateral femoral cortex; D: Proximal reattachment of the trochanter.

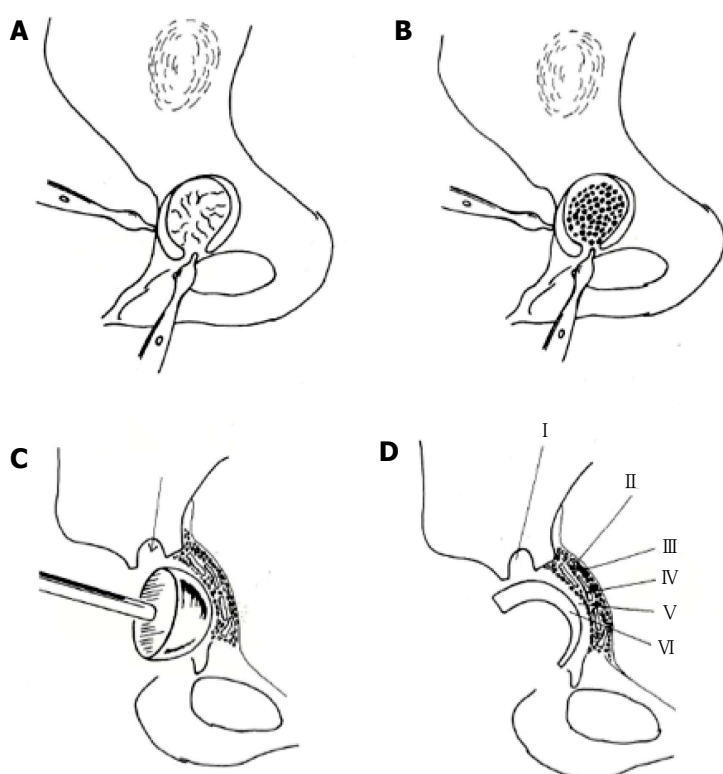


Figure 9 Cotyloplasty technique. A: Comminuted fracture of the entire medial wall; B: Large amount of cancellous morselised graft placed between the fragments of the acetabular floor, onto the periosteum; C: Grafts moulded with a hemispherical pusher; D: Final appearance: I: Anchorage hole; II: Internal layer of the periosteum; III: Autogenous morselised graft; IV: Fragments of the acetabular floor; V: Cement mantle; VI: Offset-bore acetabular component.

advantage of this technique is that the weight-bearing area is allowed to shift to beneath the acetabular roof, while adequate anterior and posterior coverage of the cup is achieved. Moreover, the host-graft interface is biologically active, which may ensure incorporation of the graft and the anatomical placement of the cup. This, combined with carefully controlled medialization optimizes the mechanical environment and influences the long-term survival of the artificial hip.

On the other hand, augmentation of superior segmental defects with structural autograft or allograft and placement of the acetabular component in the anatomical position had been suggested by Harris *et al.*^[18]. Although the short-term results of this technique were excellent, a high failure rate after approximately 12 years has been reported^[19]. This may be related

to the complex pathological anatomy encountered at the level of the true acetabulum and the abnormal distribution of stresses, combined with the unfavourable long-term biological behaviour of structural grafts^[3,9].

Shortening of the femur: For hips with high dislocation, shortening of the femur during THR is inevitable. We favour shortening of the femur by progressive resection of bone at the level of the femoral neck. We argue against leaving the greater trochanter in place and subtrochanteric femoral shortening osteotomy, because, in the majority of hips with high dislocation, the greater trochanter lies above the centre of rotation of the femoral head and its resection and advancement are essential (Figure 10). Besides, subtrochanteric osteotomy resembles an artificial fracture which needs additional osteosynthesis and may

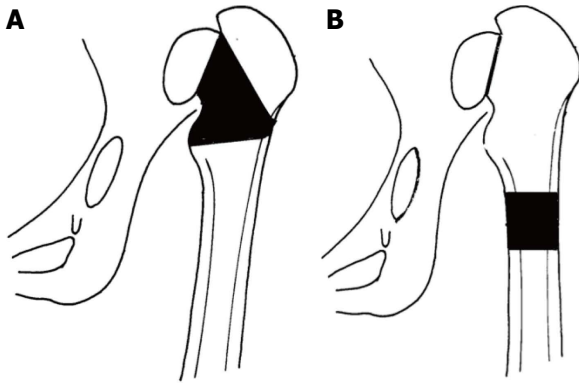


Figure 10 Drawings of the two alternatives of shortening of the femur. A: At the level of femoral neck; B: Distal shortening at the level of femoral shaft.

subsequently cause undesirable complications^[9].

Special implants: Special small implants are needed. We have been using the Chamley's offset bore cup in most of our cases. It is an extra small all-PE implant with a 35 mm face diameter, offset articular surface and approximately 10 mm PE thickness at the upper weight-bearing part, equal to that of a conventional socket, that decreases to a few millimetres in the lower part. In cases with a narrow femoral canal and short and anteverted neck, for the reconstruction of the femur we have mostly used the stainless steel Chamley CDH stems with polished surface, monoblock and collarless, and the Harris CDH stem made of CoCr, precoated at the proximal part, modular and with collar^[20-22]. Currently new cementless designs are used.

Lesson 4

Females with congenital dislocation of the hip represent a special cohort of patients with a problematic life since birth. These patients are of young age and may have pain, severe limping and deformation, major leg-length discrepancies and several psychological disorders, such as anxiety and depression, since their early childhood. We evaluated the quality of life (QoL) of 82 female patients, with low and high dislocation, followed for a minimum of 12 years after THR using clinical scores and QoL questionnaires. We concluded that THR radically improves their QoL for a long period of time. Even patients who subsequently underwent revisions had enjoyed pain relief and functional improvement for an appreciable period of time^[23].

Letters from these patients, many years after surgery, show their satisfaction for the great changes in their physical and psychological status as well as their social and family life. Short excerpts from this communication are the following^[24]: "My childhood was a life of torrent. My mother used to tell me that no man is going to love me. She made me feel useless. My life changed after surgeries at the age of 28. I got married and now I have a 15-year-old son. We are a happy family"; "My childhood and teenage life were very difficult, with many complexities

and insecurities. I was feeling like a child of an inferior God. I took the decision to have an arthroplasty at the age of 29. Even though it was necessary to be operated again after 11 years, I am now fully active leading a normal life. I am very pleased"; "My life was an Odyssey. At school, the children were making fun of me for being so different to them because of my pelvic deformity and my movements. When I had the arthroplasties in my hips, I was 47 years old. My life changed. I look at the mirror and I do not believe my eyes. People who knew me did not recognize me. Twenty-four years have gone since I was operated and I have a normal life".

Concluding messages

The most suitable term for the total spectrum of congenital hip deformities is "CHD", classified in adults into dysplasia (type A), low dislocation (type B) and high dislocation (type C). Types B and C are further subdivided in subtypes B1 and B2, and C1 and C2, respectively, depending on their different anatomic characteristics.

The three types of CHD are the main causes of secondary OA. Degenerative changes develop gradually, usually from the age of 30-35 years, causing pain and increasing functional disability over time. Knowledge of the natural history of the three types of CHD facilitates choosing the most appropriate time for THR.

The transtrochanteric approach is essential in cases with low and high dislocation and in certain dysplastic hips with great limitation of the range of motion.

Restoration of the normal centre of rotation is fundamental for the joint biomechanics and the survival of the prosthesis.

Shortening of the femur, if needed, is better to be performed at the level of the femoral neck.

In the majority of cases, special implants are needed to reconstruct the acetabulum and the femur.

The improvement of the QoL, especially in young females with high dislocation, is impressive. Most of these patients stated that after surgery they feel "like they were born again".

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Fifth metatarsal fractures and current treatment

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on management of fifth metatarsal fractures due to inconsistency with respect to classification of these fractures. This article provides a thorough review of fifth metatarsal fractures with examination of relevant literature to describe the management of fifth metatarsal fractures especially the proximal fracture. A description of nonoperative and operative management for fifth metatarsal fractures according to anatomical region is provided.

Key words: Metatarsal fractures; Fifth metatarsal; Jones fracture; Operative care; Athlete

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Core tip: Nondisplaced fifth metatarsal fractures can be treated nonoperatively depending on fracture location and patient factors. When nonoperative management is utilized improved early functional scores are associated with less rigid immobilization and a shorter period of nonweightbearing. Neck and shaft fractures with greater than ten degrees plantar angulation or three millimeters of displacement in any plane where closed reduction is insufficient require operative management. Operative intervention is recommended for base of the fifth metatarsal avulsion fractures (zone one) with more than three millimeters of displacement. Acute and delayed union zone two fractures may be managed nonoperatively but operative management with an intramedullary screw should be considered in athletes. Zone three (diaphyseal stress fractures) fractures that are Torg type I and type II should be managed with intramedullary screw fixation in the athlete. In the non-athlete these fractures may be managed nonoperatively however prolonged immobilization is often required and a nonunion may still result. Symptomatic nonunions of zone two and zone three fractures should be managed operatively.

Abstract

Metatarsal fractures are one of the most common injuries of the foot. There has been conflicting literature

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INTRODUCTION

Metatarsal fractures are frequently encountered injuries of the foot^[1]. Approximately five to six percent of fractures encountered in the primary care setting are metatarsal fractures^[2]. In adults, metatarsal fractures peak in the second to fifth decades of life. The most frequent fracture seen is the fifth metatarsal, accounting for 68% of metatarsal fractures^[2]. Proximal fifth metatarsal fractures are divided into three zones^[3-5]. Zone one, zone two, and zone three fractures account for 93%, four percent and three percent of proximal fifth metatarsal fractures, respectively^[6]. There is some evidence-based literature to help make decisions with these fracture types, which will be described in this review.

CLASSIFICATION

The first to describe a fracture of the proximal fifth metatarsal was Sir Robert Jones^[7-9]. He described a fracture in the proximal three quarter segment of the shaft distal to the styloid^[7-9]. The Jones fracture as described by Sir Robert Jones was later defined by Stewart^[10,11] as a transverse fracture at the junction of the diaphysis and metaphysis without extension into the fourth and fifth intermetatarsal articulation. Since then there has been a focus in the literature on fractures of the proximal fifth metatarsal due to the propensity for poor healing of some fractures in this region. The blood supply to the proximal fifth metatarsal is important in understanding troublesome fracture healing in this area. The blood supply of the fifth metatarsal was investigated in a cadaver model by Smith *et al.*^[12]. They found that the blood supply arises from three possible sources; the nutrient artery, the metaphyseal perforators, and the periosteal arteries. A watershed area exists between the supply of the nutrient artery and the metaphyseal perforators which corresponds to the area of poor fracture healing in the clinical setting^[12]. A classification system created by Torg *et al.*^[13] is based on healing potential. This classification simplifies proximal fifth metatarsal fractures as either involving the tuberosity or the proximal diaphysis distal to the tuberosity, the latter group being called the Jones fracture^[13,14]. Under this system the Jones fracture is divided into three types based on the radiological appearance of the fracture^[13]. Type I (acute) fractures are characterized by a narrow fracture line and an absence of intramedullary sclerosis^[13,15,16]. The features of acute fractures in this classification are no history of previous fracture, although previous pain or discomfort may be present^[13]. Torg type I fractures are presumed to be acute fractures at a site of pre-existing stress concentration on the lateral cortex that becomes acutely disabling when they extend across the entire diaphysis^[13].



Figure 1 Radiograph of a Torg type II fifth metatarsal fracture.



Figure 2 Radiograph of a fifth metatarsal Torg type III fracture, which has nonunion.

Type II (delayed union) are distinguished by having a previous injury or fracture with radiographic features of a widened fracture line and evidence of intramedullary sclerosis (Figure 1)^[13,15,16]. Type III (nonunion) are characterized by complete obliteration of the medullary canal by sclerotic bone with a history of repetitive trauma and recurrent symptoms (Figure 2)^[13,15,16]. Although the term Jones fracture was applied to the fractures in this classification, based on Torg's description these fractures are more consistent with stress fractures. As a result, proximal fifth metatarsal fractures were re-classified to avoid the confusing term of Jones fractures. Proximal fifth metatarsal fractures can be classified into three zones as described by Lawrence *et al.*^[3] and Dameron^[4,5]. Tuberosity avulsion fractures represent zone one (Figure 3)^[3-5]. Zone two (Jones fracture) is described as a fracture at the metaphysis-diaphyseal junction. Zone three or diaphyseal stress fractures include the proximal 1.5 cm of the diaphysis^[3,4,5,9,17]. This classification is straightforward however, it must be noted that their description of zone two is a slight mis-representation of the true Jones fracture as described by Stewart^[11]. It is important to note that the Jones fracture in this classification system is an acute injury with no prodrome whereas zone three fractures have a variable prodrome^[3]. The distinction between Jones (zone two) and proximal diaphyseal stress fractures



Figure 3 Radiograph of a zone one fifth metatarsal fracture.

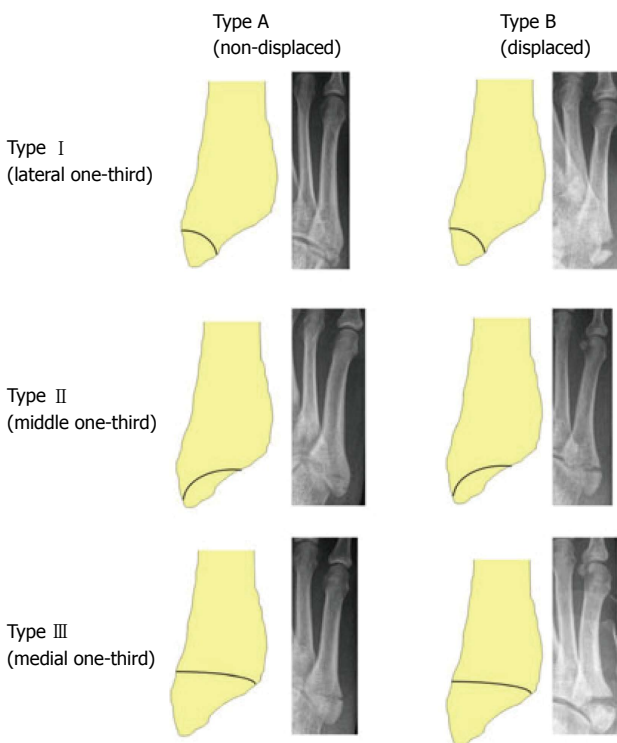


Figure 4 Classification system created by Mehlhorn *et al.*^[18] based on risk of displacement with more medial joint entry of the fracture line. Type I, type II, type III are defined as fracture line entry in the lateral one-third, middle one-third and medial one-third, respectively^[18].

(zone three) is commonly confused in the literature which potentially obscures important differences in prognosis and treatment^[3,4,10,15]. A systematic review done by Dean *et al.*^[14], looked at the classification of Jones fractures in 19 studies. They found that the majority of authors did not differentiate between fractures involving the fourth/fifth intermetatarsal articulation from more distal fractures. They concluded that the Jones fracture is generally applied to all fractures of the proximal fifth metatarsal distal to the tuberosity within 1.5 cm of this region. However, because this is not a universal definition it very difficult to recognize differences in outcomes between operative and nonoperative management of zone two and zone three fractures in the literature. It also indicates that in many cases the literature fails to differentiate the chronicity of

zone two and zone three fractures^[14].

More recently, Mehlhorn *et al.*^[18] proposed another classification for base of fifth metatarsal fractures based on radiomorphometric analysis reflecting the risk for secondary displacement. In this classification the joint surface of the fifth metatarsal base is divided into three equal parts. Type I, type II, and type III fractures represent the lateral third, middle third, and medial third respectively (Figure 4). Adding to this classification they introduced an A type which represents no relevant displacement and a B type which denotes a fracture step off of greater or equal to two millimetres^[18].

CLINICAL PRESENTATION AND ASSESSMENT

An injury to the fifth metatarsal presents with history of acute trauma or repetitive trauma to the forefoot^[19]. Zone one fractures are typically avulsion type injuries. The mechanism of these fractures are an acute episode of forefoot supination with plantar flexion^[3,19,20]. This results in pull from the lateral band of the plantar fascia and peroneus brevis^[20]. Typically, the fracture pattern is transverse to slightly oblique. Occasionally, these fractures are comminuted, significantly displaced or disrupt the cuboid-base of fifth metatarsal joint^[6]. Zone two fractures result from an acute episode^[3-5]. The exact mechanism is not known but is thought to result from a large adduction force applied to the forefoot with the ankle plantar flexed^[3,4,19]. Zone three fractures (diaphyseal stress fractures) typically results from a fatigue or stress mechanism^[3]. Stress fractures of the proximal fifth metatarsal have been defined by DeLee *et al.*^[1] as a spontaneous fracture of normal bone that results from a summation of stresses any of which by itself would be harmless. Multiple factors contribute to the development of stress fractures including systemic factors, anatomic factors, and mechanical factors^[1]. Although multiple factors contribute the exact mechanical mechanism is unclear. It is thought that either muscle creates a localized force that outweigh the stress-bearing capacity of bone or that when muscle fatigues excessive forces are transmitted to the surrounding bone^[19].

Radiographic imaging for a suspected metatarsal fracture includes three standard radiographic views of the foot: Lateral, anteroposterior, and a 45 degree oblique. Acute stress fractures are typically not detected on the standard three views of the foot. It is suggested that repeated radiographs are made at 10 to 14 d after the initial onset of symptoms^[9]. At this time a radiolucent reabsorption gap around the fracture confirms the diagnosis^[9]. In the case of more complex midfoot trauma, a CT scan is recommended to rule out the Lisfranc fracture dislocation^[9].

INDICATION FOR SURGERY

Management of fifth metatarsal fractures depends on the classification of the fracture, the nature of other



Figure 5 Radiograph showing healing in a nonunion treated with a percutaneous 3.5-mm cortical lag screw.

injuries sustained, and patient demographics. Taking everything into consideration, along with patient activity level, treatment can be nonoperative or operative.

If there is more than three to four millimeters displacement or ten degrees of plantar angulation of neck or shaft fractures and closed reduction is not sufficient, operative intervention is recommended^[10].

Avulsion fractures of the base of the fifth metatarsal that are displaced greater than three millimetres or comminuted should be reduced and operatively fixed^[7]. Fracture reduction and fixation should be considered if the fracture fragment involves more than 30% of the cubometatarsal joint^[10]. Mehlhorn *et al*^[18] recommended (based on their radiomorphometric analysis) that fractures with larger than a two millimeter step off involving the joint surface be fixed with open reduction internal fixation given the risk for posttraumatic osteoarthritis. They found that fractures they classified as type III A (medial joint fracture) had a 45% risk of secondary displacement. Therefore, they recommend that for these fractures, open reduction and internal fixation be a consideration.

Displaced zone two fractures require operative management. Less consensus exists on acute nondisplaced Jones fractures (zone two). There are many studies that advocate for early intramedullary screw fixation for acute Jones fractures in the active population^[21-24]. Porter *et al*^[21] demonstrated that acute Jones fractures treated operatively resulted in quicker return to sport and clinical healing in competitive athletes. In this same study, athletes returned to sports at a mean of 7.5 wk (range 10 d to 12 wk). This time period is shorter than the average time to healing with nonoperative management. Mindrebo *et al*^[22] described nine athletes that underwent early percutaneous intramedullary screw fixation and the patients were full weightbearing within seven to ten days. They found that on average the patients were able to return to full sport by 8.5 wk and all had radiographic union by an average of six weeks^[22]. Literature published by Quill^[23] reports that one in three nonoperatively treated Jones fractures re-fractured and therefore recommended early surgical management.

Another study by Mologne *et al*^[24] compared non-

operative management with a nonweightbearing cast or early intramedullary screw fixation and weightbearing within 14 d. The operative treatment group demonstrated a reduced time to return to sport and faster clinical union by almost 50% compared to the nonoperative group^[24]. The incidence of treatment failure in the cast treatment group was 44%. However, it should be noted that in this study a Jones fracture was defined as a type I Torg fracture and did not account for the difference between zone two and three fractures^[24].

Zone two delayed unions are a relative indication for surgical intervention. While delayed unions may eventually heal the detrimental effects of prolonged immobilization and nonweightbearing is a reason to consider operative intervention^[3].

Surgical intervention is typically recommended for type II and type III diaphyseal stress fractures, as seen in Figure 5^[4,25]. Delayed unions may eventually heal by nonoperative means but often require prolonged immobilization and nonweightbearing which is a reason to consider operative management^[3,25]. In highly active individuals with a type II diaphyseal stress fracture, operative management is recommended^[3,10,25].

Additionally, when making decision between operative and nonoperative management of zone two and zone three fractures hindfoot varus should be excluded. Evidence of hindfoot varus is thought to be a predisposing factor for these fractures as well as re-fracture following fixation. In one study done by Raikin *et al*^[26], 90% of patients with Jones fractures had evidence of hindfoot varus, whereas the incidence of hindfoot varus in the normal population is approximately 24%. In this study Jones fractures included acute and stress fractures of zone two and zone three. It can be concluded that patients presenting with zone two and zone three fractures and clinical hindfoot varus require correction of the varus to prevent re-fracture after operative or nonoperative management. Raikin *et al*^[26] corrected hindfoot varus with a lateral heel wedge and forefoot post inserts which resulted in no re-fractures of operatively managed fractures.

NONOPERATIVE TREATMENT

There are a number of nonoperative treatment modalities used for metatarsal fractures. They vary by anatomical region, patient history and radiological findings but evidence based medicine has helped with this treatment type. Isolated nondisplaced shaft and neck fractures of the fifth metatarsal are treated nonoperatively^[10,27]. A variety of nonoperative modalities include elastic dressing and a rigid shoe, short leg walking cast, posterior splint, or a hard plastic cast shoe with weightbearing as tolerated^[26]. A study done by O'Malley *et al*^[28] demonstrated that active individuals do well when treated nonoperatively. The study looked at 35 ballet dancers with distal shaft fractures treated nonoperatively and 31 of the patients returned to dance without limitations or pain^[28].

Table 1 Studies comparing nonoperative management of zone 1 proximal fifth metatarsal fractures

Ref.	Treatment modality	Outcome
Shahid <i>et al</i> ^[29]	Airboot compared to below knee walking cast	Pain and function recovered quicker with airboot No difference in time to union between groups
Clapper <i>et al</i> ^[30] Gray <i>et al</i> ^[31]	Hard-soled shoe compared to below knee cast Plastic slipper compared to tubi-grip support	No difference between clinical healing results Fractures treated with a plaster slipper resulted in significantly better pain and function at 2 wk At 6 and 12 wk the outcomes were similar for both treatment groups
Wiener <i>et al</i> ^[32]	Below knee casting compared to soft "Jones" dressings	The average time to union was 33 d vs 46 d respectively for soft dressings compared to rigid casting

Evidence based studies suggest that nondisplaced zone one fractures at the base of the fifth metatarsal are treated with protected weightbearing utilizing one of the many modalities varying from a short leg cast to elastic dressing and rigid shoe only as seen in Table 1^[3,9,26,29-32]. The outcomes of nonoperative treatment for nondisplaced zone one fractures are good with low nonunion rates reported between 0.5% and 1%^[33].

Acute zone two fractures are managed with non-weightbearing in a short leg cast for 6 to 8 wk. Torg type I diaphyseal stress fractures (zone three) are also managed the same however, prolonged immobilization up to twenty weeks may be required^[3,10,13,31]. Despite prolonged immobilization zone three diaphyseal stress fractures may still go on to nonunion. Delayed unions of zone two and type II zone three fractures may eventually heal by nonoperative treatment but operative management is recommended in highly active patient populations^[3,10,13,25]. Additionally, despite prolonged immobilization in zone three fractures it is not uncommon for a nonunion to occur^[3].

There is a clear lack of randomized controlled trials comparing various nonoperative treatment modalities. As a result, the choice of nonoperative management should be based on the patient and the individual fracture type. A retrospective study done by Konkel *et al*^[33], demonstrates the results obtained from nonoperative management of fifth metatarsal fractures. They found that the average time to bony union for tuberosity fractures, Jones, stress, segmental shaft, and oblique distal shaft/neck fractures was 3.7, 3.5, 4.8, 3.6 and 3.4 mo respectively. There was only one nonunion out of the 66 metatarsal fractures, which was a tuberosity fracture. There was delayed union in ten tuberosity fractures, two Jones fractures, two stress fractures and four oblique distal shaft/neck fractures. Overall they found that delayed union was seen in 27% of the patients with an overall union rate of 98.5%. The long-term satisfaction rate with nonoperative management was 100% in 40 patients with long-term follow-up^[33].

One consideration is patients factors associated with less favourable outcomes. Female gender, diabetes mellitus and obesity are associated with adverse outcomes of metatarsal fractures^[34]. One prospective cohort study showed that Torg type III fractures, displacement, and weight were significant independent predictors of poor

outcomes at six weeks^[35]. Additionally, this study showed that at 20 wk in addition to the above factors gender and diabetes were also significant independent predictors of poor outcome^[35].

OPERATIVE TREATMENT

There are a variety of modalities for operative management of proximal fifth metatarsal fractures including percutaneous fixation with an intramedullary screw, corticocancellous bone graft, closed reduction and cross-pinning with Kirschner-wire (K-wire) fixation, or open reduction and internal fixation with minifragment plate and screws^[3,4,9].

Zone one fractures that require operative fixation based on the indications specified in a previous section can be fixed using K-wires, tension band wiring, or small ASIF screws. If an avulsed fragment is too small for fixation excision may be required if chronic irritation results^[3,4,19].

Percutaneous fixation with an intramedullary screw has become the preferred treatment choice for zone two and three fractures requiring operative fixation as specified in the indications for surgery section^[3,15,19,24,25]. The advantage of this construct is that it is minimally invasive and compression across the fracture site can be obtained^[1,36]. It also has been shown to have decreased healing time with accelerated mobilization^[19,36]. DeLee *et al*^[1] were the first to describe the use of percutaneous intramedullary screw fixation with a solid 4.5-mm malleolar screw for diaphyseal stress fractures in ten athletes. The study reported an average healing time of 7.5 wk and return to sport in an average of 8.5 wk with no postoperative complications or re-fractures^[1,2,4]. It is important to note that this study was published prior to the introduction of the Torg *et al*^[13] classification. Historically, the treatment of choice for symptomatic Torg type II and type III fractures as first described by Torg *et al*^[13], in their study that introduced the Torg classification was cortico-cancellous bone graft. In this series 95% of patients treated with cortico-cancellous bone graft had healed radiographically and clinically at a mean of 12.3 wk^[13]. Currently, the most accepted technique for nonunions is open curettage of the nonunion site followed by intramedullary screw placement^[8].

Currently, there exists a variety of intramedullary

screws a surgeon can select from for fixation of a proximal fifth metatarsal fracture. Solid and cannulated screws exist. The theoretical advantage of a cannulated screw is the precision and ease of screw placement over a guidewire^[25,37]. However, a study done by Glasgow *et al.*^[38] reported the risk of re-fracture with cannulated screws. The study examined operative failure of three delayed unions, three nonunions, and five acute Jones fractures. A variety of intramedullary screws were used including: 4.0-mm cancellous, 4.5-mm malleolar, 4.5-mm cannulated screw, and a 6.5-mm cancellous screw. They concluded that intramedullary fixation with other than a 4.5-mm malleolar screw resulted in re-fracture and failure. In this same study two of the nonunions and all of the delayed unions treated with cortico-cancellous bone graft failed. They concluded failure was due to undersized cortico-cancellous grafts and incomplete reaming of the medullary canal. Additionally, early return to vigorous physical activity with the bone graft procedure and screw fixation was associated with re-fractures and delayed union^[38]. It should be noted that in this study the definition of Jones fracture included zone two and zone three fractures. On the contrary, a more recent study by Porter *et al.*^[21] described 100% union using a partially threaded cancellous 4.5 mm cannulated screw for fixation of acute zone two fractures in self-reported athletes with high satisfaction rates and no re-fractures^[21].

Pietropaoli *et al.*^[37] compared the strength of 4.5-mm malleolar screws and 4.5-mm partially threaded cancellous cannulated screws in a simulated Jones fracture (zone two) cadaver model. The study reported no difference in the two screws from a biomechanical standpoint. They also found that the forces to cause displacement in both screws were much higher than the peak force experienced by the lateral aspect of the foot. Therefore, they concluded that early return to function after intramedullary screw fixation of these fractures should be considered^[37].

Portland *et al.*^[25] demonstrated 100% union rate after immediate intramedullary fixation with 4.5-mm or 5.0-mm cannulated screws in acute Jones fractures (zone two) and Torg type I diaphyseal stress fractures, with an average time to union of 6.2 wk. Immediate intramedullary fixation of type II diaphyseal stress fractures resulted in 100% union and average time to union of 8.3 wk. They advocate for immediate intramedullary fixation of Jones fractures and acutely presenting Torg type I and II diaphyseal stress fractures^[25].

A more recent study done by DeSandis *et al.*^[36] investigated screw sizing of zone two fractures using CT and radiographic analysis of fifth metatarsal morphology in 241 patients. The fifth metatarsal has a lateral curvature and a plantar bow and its shaft morphology is variable which makes choosing the correct screw challenging. They recommended using the largest diameter screw possible keeping in mind using a large diameter medullary screw in a narrow canal can result in diaphyseal fracture. The analysis found a range of canal widths between 2.2- and 5.9-mm and they concluded

most canals can accommodate a 4.0- or 4.5-mm diameter screw. They recommend using AP radiographs for preoperative templating of screw diameter to ensure the screw is an adequate size for the patient. Screw length they concluded should be as short as possible with 16 mm of distal threads. Based on their imaging analysis screw length should rarely be larger than 50-mm and typically should be 40-mm or less to prevent fracture distraction that can result due to the natural plantar bow of the fifth metatarsal. Additionally, special attention should be paid to larger individuals which based on their analysis were found to have more bowing. In this population excessively long screws should be avoided to reduce the risk of medial cortex perforation which may lead to fracture distraction^[36].

In some cases, tension band wiring may be favoured if there are small fragments that are not amenable to screw fixation^[32]. Sarimo *et al.*^[39] treated 27 zone two fractures with tension band wiring with good results. Patients started weightbearing at three weeks and the mean time to union was 12.8 wk^[36].

Postoperatively after fixation of proximal fifth metatarsal fractures the foot should be immobilized and kept nonweightbearing. The period of nonweightbearing is 1 to 2 wk with progressive weightbearing in a short-leg walking cast or aircast for four to six weeks^[3,4,10,13,19,25]. A functional brace or foot orthoses may be worn if the patient is returning to strenuous competitive activity^[19,26]. Following inlay cortico-cancellous bone grafting the patient should be immobilized and nonweightbearing for six weeks as specified by Torg *et al.*^[13].

CONCLUSION

The treatment of fifth metatarsal fractures is evolving and evidence-based medicine is directing care which can be nonoperative or operative. The choice of treatment varies by anatomical region, patient history and radiological findings. All nondisplaced fractures including stress fractures can be treated nonoperatively^[3,9,10,19]. Fifth metatarsal neck and shaft fractures may be treated with a variety of nonoperative modalities and weightbearing as tolerated^[19]. There is no definitive literature that describes the exact amount of translation and angulation that is acceptable for metatarsal neck and shaft fractures. The criteria cited is more than 10 degrees of plantar angulation or three to four millimeters of translation in any plane^[19]. Displacement greater than this, should be corrected by open reduction if closed reduction fails^[19]. The recommended treatment for nondisplaced zone one fractures is symptomatic care in a walking cast, air-boot, or compression wrap with protected weightbearing until discomfort subsides^[3,19]. Based on the current literature, less rigid immobilization and shorter period of nonweightbearing can be associated with better early functional outcomes (Table 1). These fractures should be treated operatively if displaced more than three millimeters or if more than 30% of the cubometatarsal joint is involved. The treatment of zone two and zone three

fractures is more complex because they are recognized for prolonged healing time and nonunion. Nonoperative treatment of zone two and zone three fractures includes immobilization and nonweightbearing for 6 to 8 wk or longer in the case of zone three stress fractures^[3,9]. Torg type I diaphyseal stress fractures and acute zone two fractures are managed nonoperatively, however in the highly active population operative management should be considered due faster clinical healing and return to sport^[3,4,21-24]. Zone two and zone three delayed unions may be managed operatively or nonoperatively^[3,4]. Operative intervention is recommended in the active population^[3]. Symptomatic zone two and zone three nonunions should be managed operatively^[3,4].

Evidence based decisions are difficult in this anatomic area as there is a paucity of good randomized control trials comparing treatment options. In the studies that do exist zone two and zone three fractures are often confused. In addition to fracture classification inconsistencies there are also inconsistencies with respect to chronicity of fractures and differentiating between stress fractures and an acute traumatic mechanism^[39]. Regardless, when making decisions on treatment, special attention should be paid to the athlete with operative and nonoperative approaches to treatment being outlined and the treatment modality should be based on the patient's preference.

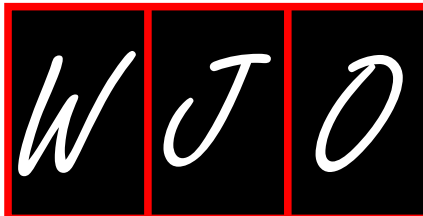
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Basic Study

Extrinsic visual feedback and additional cognitive/physical demands affect single-limb balance control in individuals with ankle instability

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Abstract

AIM

To investigate the impact of extrinsic visual feedback and additional cognitive/physical demands on single-limb balance in individuals with ankle instability.

METHODS

Sixteen subjects with ankle instability participated in the study. Ankle instability was identified using the Cumberland Ankle Instability Tool (CAIT). The subject's unstable ankle was examined using the Athletic Single Leg Stability Test of the Biodex Balance System with 4 different protocols: (1) default setting with extrinsic visual feedback from the monitor; (2) no extrinsic visual feedback; (3) no extrinsic visual feedback with cognitive demands; and (4) no extrinsic visual feedback with physical demands. For the protocol with added cognitive demands, subjects were asked to continue subtracting 7 from a given number while performing the same test without extrinsic visual feedback. For the protocol with added physical demands, subjects were asked to pass and catch a basketball to and from the examiner while performing the same modified test.

RESULTS

The subject's single-limb postural control varied significantly among different testing protocols ($F = 103$; $P = 0.000$). Subjects' postural control was the worst with added physical demands and the best with the default condition with extrinsic visual feedback. Pairwise

comparison shows subjects performed significantly worse in all modified protocols ($P < 0.01$ in all comparisons) compared to the default protocol. Results from all 4 protocols are significantly different from each other ($P < 0.01$) except for the comparison between the “no extrinsic visual feedback” and “no extrinsic visual feedback with cognitive demands” protocols. Comparing conditions without extrinsic visual feedback, adding a cognitive demand did not significantly compromise single-limb balance control but adding a physical demand did. Scores from the default protocol are significantly correlated with the results from all 3 modified protocols: No extrinsic visual feedback ($r = 0.782$; $P = 0.000$); no extrinsic visual feedback with cognitive demands ($r = 0.569$; $P = 0.022$); no extrinsic visual feedback with physical demands ($r = 0.683$; $P = 0.004$). However, the CAIT score is not significantly correlated with the single-limb balance control from any of the 4 protocols: Default with extrinsic visual feedback ($r = -0.210$; $P = 0.434$); no extrinsic visual feedback ($r = -0.450$; $P = 0.081$); no extrinsic visual feedback with cognitive demands ($r = -0.406$; $P = 0.118$); no extrinsic visual feedback with physical demands ($r = -0.351$; $P = 0.182$).

CONCLUSION

Single-limb balance control is worse without extrinsic visual feedback and/or with cognitive/physical demands. The balance test may not be a valid tool to examine ankle instability.

Key words: Ankle; Balance; Instability; Motor control; Rehabilitation

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Core tip: Single-limb balance control with the Biodex Balance System (BBS) was significantly worse without extrinsic visual feedback and with cognitive or physical demands in those with ankle instability. Clinicians should consider a patient’s activity and incorporate proper additional demands in ankle stability testing. In addition, the Athletic Single Leg Stability Test of the BBS may not be a valid tool to examine ankle instability. Further research is needed to examine the validity and reliability of the Athletic Single Leg Stability Test in testing ankle instability.

Hung Y, Miller J. Extrinsic visual feedback and additional cognitive/physical demands affect single-limb balance control in individuals with ankle instability. *World J Orthop* 2016; 7(12): 801-807 Available from: URL: <http://www.wjgnet.com/2218-5836/full/v7/i12/801.htm> DOI: <http://dx.doi.org/10.5312/wjo.v7.i12.801>

INTRODUCTION

Ankle sprain is one of the most common musculoskeletal injuries, especially for active individuals and athletes^[1-3]. An estimated 23000 ankle sprains occur daily in the

United States alone^[1]. Ankle sprains often occur as the result of trauma (e.g., landing on an uneven surface from a jump), compromising the physical structural and functional integrity of the tissues surrounding the joint^[4]. It was reported that lateral ankle sprains comprise up to 83% of all ankle injuries^[5]. They are likely the result of a fast perturbation of ankle plantar flexion and inversion, contributing to complete or partial tears of the 3 lateral ankle ligaments (anterior talo-fibular ligament, calcaneo-fibular ligament, and posterior talo-fibular ligament). Moreover, compromised mechanical restraints (e.g., injured ligaments, joint capsule), muscle strength, and/or neuromuscular control (e.g., proprioception deficits) after the initial injury may further compromise ankle stability^[6-18]. As the result, 73% of the individuals who had sprained their ankles before are likely to experience recurrent injuries and ankle instability^[19].

Several screening tools [e.g., The Cumberland Ankle Instability Tool (CAIT), The Ankle Instability Instrument, The Functional Ankle Instability Questionnaire] have been developed to identify individuals with ankle instability^[18]. A panel of experts concluded that CAIT is based on the highest level (level I) of evidence according to guidelines described by the Centre of Evidence-Based Medicine, Oxford, United Kingdom^[18]. The CAIT is a 9-item questionnaire with the score ranges from 30 (best) to 0 (worst)^[19]. A subject scores lower than 28 would be considered having ankle instability. It has a test-retest intraclass correlation coefficient (ICC) of 0.96, and it also has a good sensitivity (82.9) and specificity (74.7) in differentiating participants with or without ankle instability^[19].

Proper balance control is crucial to ensure safe functional activities. It is achieved with the integration of sensory inputs (e.g., visual, vestibular, and somatosensory information), muscle activations, and cognitive function in human bodies^[20]. For individuals with ankle instability, compromised muscle strength and proprioception around the ankle joint may hamper balance control^[21,22]. A variety of laboratory equipment (e.g., force plates) and clinical tests (e.g., Y Balance Test, Star Excursion Balance Test, Foot Lift Test) have been developed to examine balance control. One of the commonly used devices to examine both static and dynamic balance control is the Biodex Balance System (BBS; Biodex, Inc, Shirley, NY)^[23]. This apparatus has a good test-retest reliability and provides quantitative measures of balance control^[24,25]. In addition, the BBS uses a multi-axial testing platform which can be set at various degrees of instability/difficulty (from the static protocol of 0° surface tilt to a dynamic protocol of 20° surface tilt) to challenge the subjects with various fitness levels and injury severities^[25]. Compared to other balance testing equipment, the unstable platform of the BBS can simulate unexpected external perturbations (such as landing on an uneven surface) in various activities. However, the monitor of the BBS also provides extrinsic visual feedback (information about the center of gravity location in relation to the base of support) to the subject and compromises the test’s functional significance. In real life scenarios, individuals don’t receive concurrent visual

information about their performance. Therefore, balance control measured with the BBS may not truly reflect balance control in daily activities.

Individuals often sprain their ankles while engaging a sport activity (e.g., basketball), in which additional cognitive demands (e.g., whom to pass the ball to) and/or physical demands (e.g., catching or passing the ball) are often present. It was suggested that balance activities take place in association with at least one concurrent task in daily activities, and cognitive function can have an impact on balance control^[26]. The impact of adding a cognitive loading on functional activities such as gait and balance control is inconclusive, depending on many factors such as the difficulty of the primary/secondary tasks and subject conditions^[26-34]. Examining single-leg balance control with the BBS, Rahnama *et al.*^[30] (2010) reported adding a cognitive task decreased postural stability in subjects with ankle instability. However, they used a modified protocol with their subjects' eyes closed during the testing. Eliminating all visual inputs does not resemble functional activities and common ankle injury mechanisms. Moreover, their protocol can further increase anxiety and unnecessary muscle activation, therefore compromising balance control. In addition, no study had examined the impact of adding a physical demand with a functional significance to individuals performing the single-leg balance test with the BBS.

The first aim of this study was to investigate the impact of extrinsic visual feedback and additional cognitive/physical demands on single-limb balance in individuals with ankle instability. The second aim of the study was to investigate if any of the 4 single-limb balance testing protocols correlates to ankle stability measured by the CAIT. It was hypothesized that taking away the extrinsic feedback and additional cognitive/physical demands could compromise single-limb balance control. Results of the study can provide clinicians useful information regarding testing and rehabilitation regimens for individuals with ankle instability.

MATERIALS AND METHODS

Participants

Sixteen subjects (12 females and 4 males, ranged from 19-30 years old) with ankle instability participated in the study. Subjects were recruited from the campus of a local university. The inclusion criteria for the subjects includes: (1) have one or more ankle sprains over the same ankle resulted in pain, swelling, and/or loss of function when it occurred; (2) have the latest ankle sprain occurred within the past year; (3) have no other prior injury that received medical attention for the injured ankle; (4) have no pain or discomfort during single leg standing over the injured ankle at participation; and (5) answer "no" to all questions on the Physical Activity Readiness Questionnaire (PAR-Q and YOU)^[35]. All participants signed a consent form approved by the Institutional Review Board of the local university at the beginning of the study.

Procedures

At the beginning of the testing session, subjects were asked to fill out the CAIT questionnaire (Table 1). Only subjects who scored 27 or less (an indication of ankle instability) were asked to participate in the study. The subject's unstable ankle was examined using the Athletic Single Leg Stability Test of the BBS. The single-leg test was chosen because all subjects were recreational athletes and other double foot support and/or static protocols of the BBS lack functional significance. Subjects were examined with 4 different protocols: (1) default setting with extrinsic visual feedback; (2) no extrinsic visual feedback; (3) no extrinsic visual feedback with cognitive demands; and (4) no extrinsic visual feedback with physical demands (Figure 1). Subjects were tested at dynamic level 4, which provided moderate balance control difficulty.

After adopting a single-limb stance on the BBS platform without shoes, subjects performed a total of 3 trials with 20 s/trial while receiving extrinsic visual feedback (their center of gravity location in relation to the base of support) concurrently from the monitor. For the remaining 3 modified protocols, subjects were positioned on the platform facing the opposite direction while keeping the same relative foot position/alignment in relation to the platform as in the default protocol. In the modified protocols, subjects were able to use vision to assist maintaining the balance but without the direct extrinsic visual feedback from the monitor. For the protocol with cognitive demands, subjects were asked to continue subtracting 7 from 121 (trial 1), 119 (trial 2), and 116 (trial 3) to 0 without feedback from the monitor. For the protocol with physical demands, subjects were asked to pass and catch a basketball to and from an examiner standing 6 feet away. The pace was standardized at once every second (guided by a metronome). All 3 modified protocols consisted of 3 trials with 20 s/trial. Subjects were asked to sit and relax for 2 min between protocols to avoid fatigue.

Statistical analysis

Statistical analyses were conducted using IBM SPSS Statistics (Armonk, NY) Version 21.0. The overall stability index (OSI) produced by the BBS was used for analyses. One-way Analysis of Variance (ANOVA) with repeated measures was used to compare the 4 different testing protocols. Post hoc comparisons were performed with the Paired-Samples *T* test. Pearson Correlation was used to examine the correlations between the OSI and the CAIT scores. Significance level (*P*-values) was set at 0.05 for all comparisons.

RESULTS

The subject's single-limb balance control varied significantly among different protocols ($F = 103$; $P = 0.000$). Subjects' postural control was the worst with added physical demand (passing and catching a basketball) and the best

Table 1 The Cumberland Ankle Instability Tool

Please check the one statement in each question that best describes your ankles			
	Left	Right	Score
1 I have pain in my ankle			
Never	<input type="checkbox"/>	<input type="checkbox"/>	5
During sport	<input type="checkbox"/>	<input type="checkbox"/>	4
Running on uneven surfaces	<input type="checkbox"/>	<input type="checkbox"/>	3
Running on level surfaces	<input type="checkbox"/>	<input type="checkbox"/>	2
Walking on uneven surfaces	<input type="checkbox"/>	<input type="checkbox"/>	1
Walking on level surfaces	<input type="checkbox"/>	<input type="checkbox"/>	0
2 My ankle feels unstable			
Never	<input type="checkbox"/>	<input type="checkbox"/>	4
Sometimes during sport (not every time)	<input type="checkbox"/>	<input type="checkbox"/>	3
Frequently during sport (every time)	<input type="checkbox"/>	<input type="checkbox"/>	2
Sometimes during daily activity	<input type="checkbox"/>	<input type="checkbox"/>	1
Frequently during daily activity	<input type="checkbox"/>	<input type="checkbox"/>	0
3 When I make SHARP turns, my ankle feels unstable			
Never	<input type="checkbox"/>	<input type="checkbox"/>	3
Sometimes when running	<input type="checkbox"/>	<input type="checkbox"/>	2
Often when running	<input type="checkbox"/>	<input type="checkbox"/>	1
When walking	<input type="checkbox"/>	<input type="checkbox"/>	0
4 When going down the stairs, my ankle feels unstable			
Never	<input type="checkbox"/>	<input type="checkbox"/>	3
If I go fast	<input type="checkbox"/>	<input type="checkbox"/>	2
Occasionally	<input type="checkbox"/>	<input type="checkbox"/>	1
Always	<input type="checkbox"/>	<input type="checkbox"/>	0
5 My ankle feels unstable when standing on one leg			
Never	<input type="checkbox"/>	<input type="checkbox"/>	2
On the ball of my foot	<input type="checkbox"/>	<input type="checkbox"/>	1
With my foot flat	<input type="checkbox"/>	<input type="checkbox"/>	0
6 My ankle feels unstable when			
Never	<input type="checkbox"/>	<input type="checkbox"/>	3
I hop from side to side	<input type="checkbox"/>	<input type="checkbox"/>	2
I hop on the spot	<input type="checkbox"/>	<input type="checkbox"/>	1
When I jump	<input type="checkbox"/>	<input type="checkbox"/>	0
7 My ankle feels unstable when			
Never	<input type="checkbox"/>	<input type="checkbox"/>	4
I run on uneven surfaces	<input type="checkbox"/>	<input type="checkbox"/>	3
I jog on uneven surfaces	<input type="checkbox"/>	<input type="checkbox"/>	2
I walk on uneven surfaces	<input type="checkbox"/>	<input type="checkbox"/>	1
I walk on a flat surface	<input type="checkbox"/>	<input type="checkbox"/>	0
8 Typically, when I start to roll over (or "twist") on my ankle, I can stop it			
Immediately	<input type="checkbox"/>	<input type="checkbox"/>	3
Often	<input type="checkbox"/>	<input type="checkbox"/>	2
Sometimes	<input type="checkbox"/>	<input type="checkbox"/>	1
Never	<input type="checkbox"/>	<input type="checkbox"/>	0
I have never rolled over on my ankle	<input type="checkbox"/>	<input type="checkbox"/>	3
9 After a typical incident of my ankle rolling over, my ankle returns to "normal"			
Almost immediately	<input type="checkbox"/>	<input type="checkbox"/>	3
Less than one day	<input type="checkbox"/>	<input type="checkbox"/>	2
1-2 d	<input type="checkbox"/>	<input type="checkbox"/>	1
More than 2 d	<input type="checkbox"/>	<input type="checkbox"/>	0
I have never rolled over on my ankle	<input type="checkbox"/>	<input type="checkbox"/>	3

with the default condition with extrinsic visual feedback (Figure 2). Pairwise comparison shows subjects performed significantly worse in all modified protocols ($P < 0.01$ in all comparisons) compared to the default protocol. Results from all 4 protocols are significantly different from each other ($P < 0.01$), except for the comparison between the "no extrinsic visual feedback" and "no extrinsic visual feedback with cognitive demands" protocols. Comparing conditions without extrinsic visual feedback, adding a cognitive demand did not significantly compromise single-limb balance control but adding a physical demand did.

Scores from the default protocol are significantly correlated with the results from all 3 modified protocols: no extrinsic visual feedback ($r = 0.782$; $P = 0.000$); no extrinsic visual feedback with cognitive demands ($r = 0.569$; $P = 0.022$); no extrinsic visual feedback with physical demands ($r = 0.683$; $P = 0.004$). However, the CAIT score is not significantly correlated with the OSI from any of the 4 protocols: default with extrinsic visual feedback ($r = -0.210$; $P = 0.434$); no extrinsic visual feedback ($r = -0.450$; $P = 0.081$); no extrinsic visual feedback with cognitive demands ($r = -0.406$; $P = 0.118$); no extrinsic

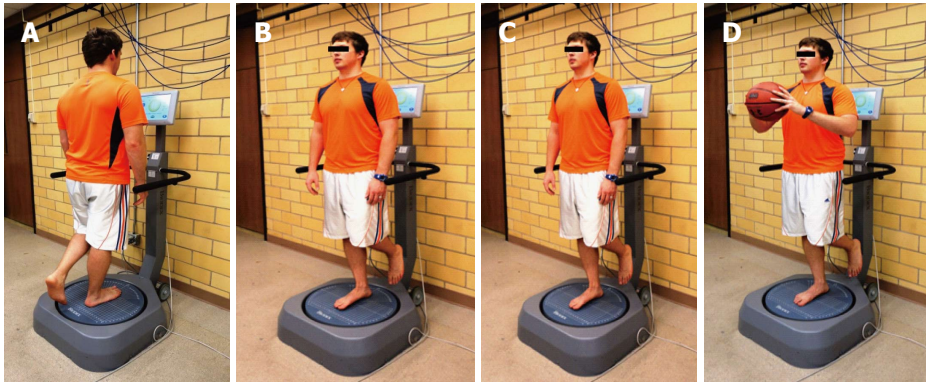


Figure 1 Single-limb balance testing protocols with the Biodex Balance System. A: The Athletic Single Leg Stability Test (default) with extrinsic visual feedback from the monitor; B: Modified test without extrinsic visual feedback; C: Modified test without extrinsic visual feedback and with cognitive demands; D: Modified test without extrinsic visual feedback and with physical demands.

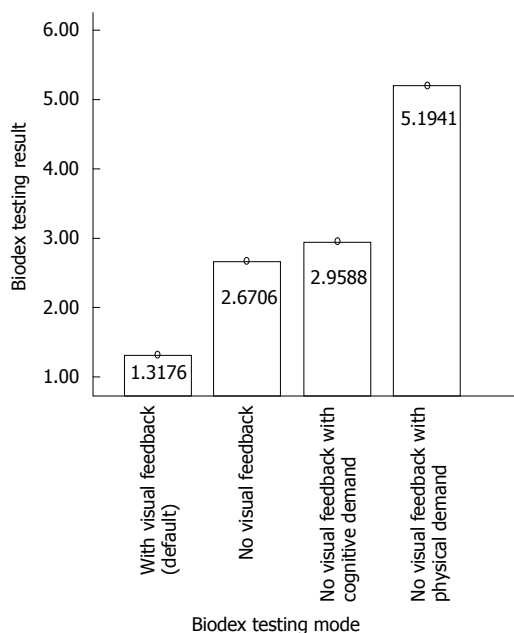


Figure 2 Overall stability Index of the Biodex Balance System from the 4 testing protocols.

visual feedback with physical demands ($r = -0.351$; $P = 0.182$).

DISCUSSION

Compared to the default setting, results of the present study show single-limb balance control was significantly worse without extrinsic visual feedback with the BBS. The BBS is a common testing and training apparatus for balance control. However, the monitor of the BBS provides concurrent extrinsic visual feedback about the performance, which is not available in most activities. In other words, performance with the BBS may overestimate the true capability of balance control in a functional setting. Although providing additional extrinsic visual feedback could be beneficial during training sessions for those with balance control deficits (e.g., patients with severe Parkinson disease), its result may not accurately

reflect the performance in other conditions without additional visual feedback. Moreover, individuals who train exclusively with the BBS may grow accustomed to rely too much on extrinsic visual cues for balance control.

The results show that adding a cognitive demand did not significantly compromise single-limb balance control with the BBS. Literature is very limited about the role of cognitive function on balance control for individuals with ankle instability^[30]. The impact of adding a cognitive loading on functional activities depends on many factors such as the difficulty of the primary/secondary tasks and subject conditions^[26-34]. Rahnama *et al.*^[30] (2010) reported that adding a cognitive task decreased single-limb postural stability in subjects with ankle instability. However, their subjects were asked to close their eyes during the testing. Despite not having extrinsic visual feedbacks, subjects in the current study could still use their vision in a subconscious matter to adjust their body alignment in relation to surrounding objects. Therefore, an easier primary task (maintaining the balance) may explain the lack of cognitive effect in the current study. Another explanation for the difference between the two projects is the difficulty of the secondary task. Instead of performing a simple mathematic calculation task, their subjects were asked to remember the sequence of 7 digits and then repeat the digits in the exact reverse order. The more difficult secondary task could also have a greater impact on single-limb balance control in their study.

No study had examined the impact of adding physical demands on single-limb balance control in individuals with ankle instability. Considering basketball players are more vulnerable to ankle sprains (41.1% prevalence) than other athletes^[3], the present study adopted a physical demand (catching and passing) that is similar to playing basketball. The results show that adding a physical demand significantly compromised single-limb balance control with the BBS. In order to catch and pass the basketball properly in a timely fashion, subjects could not solely focus on balance control. Engaging an upper extremity movement/perturbation also moved their center of gravity away from the base of support more

often, therefore making it more challenging to maintain single-limb balance. Based on the results, clinicians should incorporate physical demands in ankle stability testing and rehabilitation protocols to better simulate functional activities and sports.

Results of the present study indicate a poor correlation between single-limb balance control and ankle instability severity. It was suggested that ankle instability can have a negative impact balance control^[21,22]. Because after the initial ankle sprain, overstretched ligaments and joint capsule may hamper the function of mechanoreceptors (e.g., muscles spindles and Golgi Tendon Organs) and compromise the proprioception of the ankle joint^[6-18]. However, other studies found no proprioception difference between unstable and healthy ankles^[10,36-38]. Moreover, balance control can also rely on other motor control strategies (e.g., hip strategy), and the coordination of other joints (e.g., hip and knee) and muscles (e.g., trunk muscles). In conclusion, many factors other than ankle stability can contribute to single-limb balance control. The results of the current study suggest that OSI measured with the BBS may not be a good indicator of the severity of ankle instability.

A limitation of the present study is the small sample size. In addition, future studies may consider adding a separate group of subjects without ankle instability to examine if subjects with ankle instability respond to added demands differently from healthy subjects. Although the BBS is a commonly used apparatus in a rehabilitation setting, further research is needed to examine the validity and reliability of the Athletic Single Leg Stability Test in testing ankle instability.

Single-limb balance control is compromised without extrinsic visual feedback and/or with added cognitive/physical demands. Clinicians should consider eliminating excessive extrinsic visual feedback and incorporating physical demands in ankle stability testing and rehabilitation protocols to better simulate functional activities and sports. In addition, many factors other than ankle stability may impact single-limb balance control. Single-limb balance tests with the BBS may not be a valid tool to categorize the severity of ankle instability.

COMMENTS

Background

Ankle sprain is one of the most common musculoskeletal injuries. Recurrent ankle sprains can cause ankle instability, and potentially contribute to poor balance control. The purpose of the research was to examine the impact of extrinsic visual feedback and additional cognitive/physical demands on single-limb balance control, and to examine if those testing results can correlate to the severity of ankle instability.

Research frontiers

Ankle instability is a well-studied pathology. However, it is still unclear if ankle instability would have a significant impact on single-limb balance control. In addition, some of the commonly used balance testing protocols provide too much visual feedback and lack functional significance.

Innovations and breakthroughs

In order to provide more functional significance of a commonly used protocol for

single-limb balance testing, the default protocol was modified to better resemble daily activities and sport movements.

Applications

Clinicians should consider eliminating excessive extrinsic visual feedback and incorporating physical demands in ankle stability testing and rehabilitation protocols to better simulate functional activities and sports.

Terminology

Extrinsic visual feedback in the current study refers to the visual information about the center of gravity location in relation to the base of support displayed by the Biodex Balance System monitor. Proprioception includes both position sense and movement sense of a joint.

Peer-review

Authors aimed to investigate the impact of extrinsic visual feedback and additional cognitive/physical demands on single-limb balance in individuals with ankle instability. Sixteen subjects with ankle instability participated in the study.

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Case Control Study

Benefits of the use of blood conservation in scoliosis surgery

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Abstract

AIM

To investigate whether autologous blood transfusion (ABT) drains and intra-operative cell salvage reduced donor blood transfusion requirements during scoliosis surgery.

METHODS

Retrospective data collection on transfusion requirements of patients undergoing scoliosis surgery is between January 2006 and March 2010. There were three distinct phases of transfusion practice over this time: Group A received "traditional treatment" with allogeneic red cell transfusion (ARCT) in response to an intra- or post-operative anaemia (Hb < 8 g/dL or a symptomatic anaemia); Group B received intra-operative cell salvage in addition to "traditional treatment". In group C, ABT wound drains were used together with both intra-operative cell salvage and "traditional treatment".

RESULTS

Data from 97 procedures on 77 patients, there was no difference in mean preoperative haemoglobin levels between the groups (A: 13.1 g/dL; B: 13.49 g/dL; C: 13.66 g/dL). Allogeneic red cell transfusion was required for 22 of the 37 procedures (59%) in group A, 17 of 30 (57%) in group B and 16 of 30 (53%) in group C. There was an overall 6% reduction in the proportion of patients requiring an ARCT between groups A and C but this was not statistically significant ($\chi^2 = 0.398$). Patients

in group C received fewer units (mean 2.19) than group B (mean 2.94) ($P = 0.984$) and significantly fewer than those in group A (mean 3.82) ($P = 0.0322$). Mean length of inpatient stay was lower in group C (8.65 d) than in groups B (12.83) or A (12.62).

CONCLUSION

When used alongside measures to minimise blood loss during surgery, ABT drains and intra-operative cell salvage leads to a reduced need for donor blood transfusion in patients undergoing scoliosis surgery.

Key words: Blood conservation; Scoliosis; Autologous blood; Cell salvage; Transfusion

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Core tip: To our knowledge this is the first report of autologous blood transfusion (ABT) drain use in scoliosis surgery and suggests that its use is both safe and cost effective. When used as part of a systematic programme to minimise blood loss during surgery, the use of ABT drains and intra-operative cell salvage leads to a reduced need for donor blood transfusion in patients undergoing scoliosis surgery.

Loughenbury PR, Berry L, Brooke BT, Rao AS, Dunsmuir RA, Millner PA. Benefits of the use of blood conservation in scoliosis surgery. *World J Orthop* 2016; 7(12): 808-813 Available from: URL: <http://www.wjgnet.com/2218-5836/full/v7/i12/808.htm> DOI: <http://dx.doi.org/10.5312/wjo.v7.i12.808>

INTRODUCTION

Autologous blood reinfusion techniques have become commonplace in many fields of orthopaedic surgery, especially in hip and knee arthroplasty^[1,2]. These techniques can reduce the need for donor blood and the risks that accompany its use, such as incorrect blood component transfused, transfusion reactions, viral/prion disease transmission, and the metabolic and immunological consequences of transfusing allogeneic blood^[3].

The use of autologous blood reinfusion in spinal surgery is less commonly reported. Intra-operative cell salvage has been associated with reduced transfusion requirements in scoliosis surgery^[4,5], but the use of post-operative reinfusion devices is not well documented. Reports are limited to short segment spinal fusion and have not included corrective surgery to treat scoliosis^[6]. This study was designed to determine the effect of sequentially introducing intra-operative cell salvage and post-operative autologous blood transfusion (ABT) drains in scoliosis surgery. These were used alongside a number of efforts to reduce the need for allogeneic red cell transfusion (ARCT) during scoliosis surgery including a

move from two-stage (anterior and posterior) surgery to a single stage (posterior) technique, and measures such as optimisation of pre-surgical haemoglobin levels, oral iron supplements and the antifibrinolytic drug tranexamic acid.

This study aimed: To examine the impact of using ABT drains and intra-operative cell salvage on the allogeneic transfusion requirements for scoliosis surgery in our unit; to evaluate the volume of blood drained and re-transfused using ABT drains.

MATERIALS AND METHODS

This is a retrospective analysis of all patients undergoing surgery for scoliosis in a single centre between January 2006 and March 2010. All patients undergoing corrective scoliosis surgery were included, including revision procedures. Patients under the age of six were excluded, as autologous re-transfusion systems are not recommended for use below this age.

Data was collected from the patient case notes, the transfusion laboratory database and the hospital results server. A data collection form was developed and completed by either the Transfusion Practitioner or Specialist Registrar working in the unit. Patient demographics, details of any preoperative optimisation required, pre- and post-operative haemoglobin levels, donor and autologous transfusion requirements, length of hospital stay and post-operative complications were documented.

The transfusion trigger remained constant across the series. An intra- or post-operative anaemia (Hb < 8 g/dL) or symptomatic anaemia initiated treatment with ARCT. The volume and timing of these transfusions was recorded.

Over this time period there were three distinct phases of transfusion practice: (1) Group A: January to December 2006: "Traditional treatment": These patients received ARCT in response to the transfusion trigger. No autologous blood was transfused ($n = 37$); (2) Group B: January to December 2007. Intra-operative cell salvage was implemented using the Sorin Dideco (Sorin Group, Mirandola, Italy) system ($n = 30$); (3) Group C: January 2008 to March 2010. In addition to intra-operative cell salvage a Bellovac™ (Astra Tech, AB) autologous blood transfusion wound drain was used. Blood collected in the Bellovac™ (Astra Tech) drain was reinfused in accordance with manufacturers guidelines within 6 h of the procedure. The volume of blood collected and reinfused was recorded using the Bellovac™ ABT Management chart provided with the drain ($n = 30$).

Administration of pharmacological agents, such as tranexamic acid, was led by the Consultant Anaesthetist for the case, with no standard guidelines. The doses of any preoperative antifibrinolytics were therefore variable, but were recorded for all patients.

Statistical analysis was carried out using Microsoft Excel. χ^2 tests were used to determine the significance of changes in the proportion of patients requiring an

Table 1 Case mix and patient demographics

Group	n	Median age (range)	Case mix
A	37	15 (8-33)	8 posterior 3 anterior 12 anterior/posterior staged 2 anterior/posterior combined
B	30	16 (9-57)	8 posterior 3 anterior 6 anterior/posterior staged 7 anterior/posterior combined
C	30	15 (11-74)	22 posterior 1 anterior 2 anterior/posterior 3 anterior/posterior combined

ARCT and produce a two-tailed *P* value of significance. Where means are compared a paired *t*-test was used.

RESULTS

Data was collected for 97 procedures on 77 patients between January 2006 and March 2010. Median age of patients was 15 (range 8-74). Group A (January 2006 to December 2006) contained 37 procedures with a median patient age of 15 (range 8-33); group B (January 2007 to December 2007) contained 30 procedures with a median patient age of 16 (range 9-57) and group C (January 2008 to March 2010) contained 30 procedures with a median patient age of 15 (range 11-74). The case mix in each group is detailed in Table 1.

Pre-operative haemoglobin levels

There was no difference in the mean preoperative haemoglobin level. The mean preoperative haemoglobin was 13.09 g/dL (95%CI: 12.60-13.58) in group A, 13.49 g/dL (95%CI: 12.89-14.08) in group B and 13.66 g/dL (95%CI: 13.19-14.13) in group C.

Antifibrinolytics

In group A one patient received a 50 mg dose of tranexamic acid before surgery but no other patients received antifibrinolytic therapy. No patients in group B received antifibrinolytic therapy. In group C, 9 out of 30 patients (30%) were given tranexamic acid before surgery with the dose varying from 350 mg to 1 g. There were no patients in any group taking antiplatelet medication or other agents that would increase bleeding risk.

Volume of autologous blood received

Patients in group A did not receive any autologous blood. Those in group B received intra-operative transfusion of blood collected through the cell saver. The mean volume of blood reinfused was 413.4 mL (SD 287.34 mL). There were technical problems that interrupted reinfusion in 4 out of 30 cases.

Patients in group C received autologous blood from both the cell saver and ABT drains. Intraoperative cell-

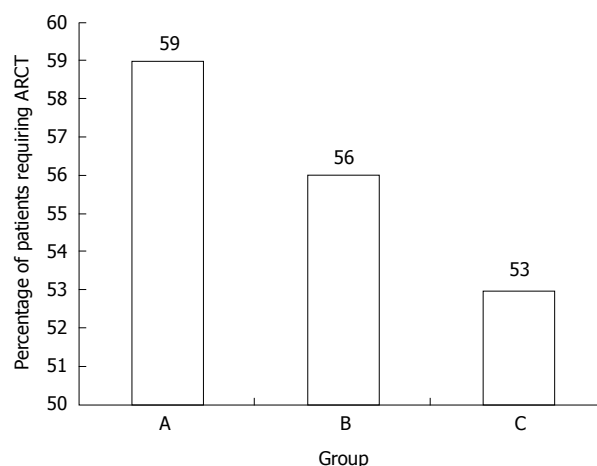


Figure 1 Percentage of patients requiring an allogeneic red cell transfusion. ARCT: Allogeneic red cell transfusion.

salvage was used successfully during 26 out of 30 procedures (87%). Technical problems or insufficient volume collected prevented reinfusion in the remaining four cases. The mean volume of cell saver blood reinfused was 525.4 mL (SD 302.96 mL). ABT drains were used successfully in 28 out of 30 cases. Technical problems and unavailability of equipment prevented reinfusion in the other two cases. The mean volume of blood received through the ABT drain was 442 mL (SD 169.38 mL).

ARCT requirements

A comparison of the ARCT requirements in groups A, B and C is shown in Table 2 and Figures 1 and 2.

There was an overall 6% reduction in the proportion of patients requiring an ARCT between groups A and C (Figure 1) but this was not statistically significant ($\chi^2 = 0.398$). The percentage of patients receiving an ARCT fell from 59% in group A to 56% in group B (not significant; $\chi^2 = 0.068$) and 53% in group C (not significant; $\chi^2 = 0.134$).

In patients requiring allogeneic transfusion, the mean number of units transfused fell from 3.82 units (SD 3.79) in group A to 2.94 (SD 1.82) in group B (not significant; $P = 0.894$) and 2.19 units (SD 0.91) in group C (not significant; $P = 0.0843$). However, the reduction in the mean number of allogeneic units transfused in groups A and C was statistically significant: - 3.82 units in group A to 2.19 units in group C (Figure 2; $P = 0.0322$). There was no difference in ARCT requirements when comparing primary and revision procedures.

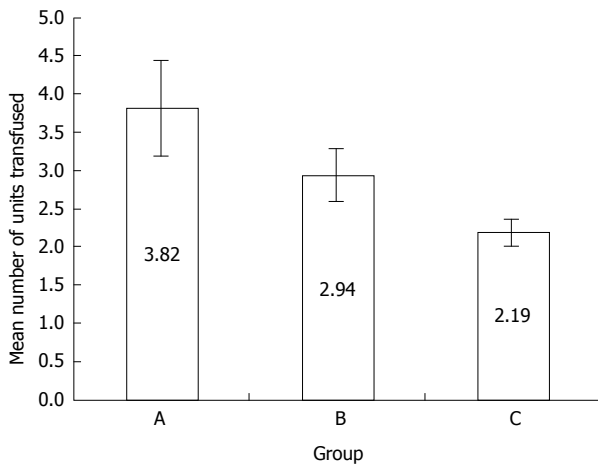
A reduction in the length of hospital stay between the groups was also noted. The mean length of stay was 12.62 d (range 8-21) in group A, 12.83 d (range 6-31) in group B and 8.65 d (range 5-23) in group C.

There were no complications relating to allogeneic or autologous transfusion in groups A and B. One patient in group C developed a pyrexia of unknown origin following surgery but this settled without the need for medical intervention. This patient received reinfused blood through the ABT drain but not the cell saver and

Table 2 Allogeneic red cell transfusion requirements

Group	Proportion requiring ARCT	Total number of units received	Mean ARCT (standard deviation)
A	22/37 (59%)	84 (73 units packed red cells, 8 units fresh frozen plasma, 2 units of cryoprecipitate and one unit of platelets)	3.82 units (3.79)
B	17/30 (57%)	50 units packed red cells	2.94 units (1.82)
C	16/30 (53%)	37 units packed red cells	2.19 units (0.91)

ARCT: Allogeneic red cell transfusion.

**Figure 2** Mean number of units transfused.

did not receive any allogeneic blood. There were no other complications in group C.

DISCUSSION

As part of an integrated blood conservation programme, autologous blood reinfusion can reduce the need for, and the potential complications of, donor blood transfusion. These include transfusion of incorrect blood components, transfusion reactions, viral/prion disease transmission, coagulopathy, and the metabolic and immunological consequences of transfusing allogeneic blood^[3].

Autologous blood reinfusion in the form of cell salvage has become commonplace in many fields of orthopaedic surgery^[1,2]. Its use in other adult elective orthopaedic procedures has been shown to reduce the rate of exposure to allogeneic red cell transfusion (RR = 0.42; 95%CI: 0.32-0.54)^[7]. In adult spinal surgery, transfusion requirements in both elective surgery and operative fracture fixation are significantly lower when cell salvage is used^[1]. The benefits of cell salvage in scoliosis surgery are less clear, but have been reported in both posterior and anterior scoliosis surgery^[4,8]. In their retrospective case-controlled study of 137 consecutive patients, Mirza *et al*^[8] underlined the benefits of cell salvage in instrumented anterior scoliosis surgery. The need for allogeneic blood fell from 39.4% in the control group ($n = 33$) to 6.7% in the study group ($n = 104$) ($P < 0.0001$). Bowen *et al*^[4] reported a retrospective case control study in which the routine use of the cell saver during posterior

spinal fusion for paediatric idiopathic scoliosis, was associated with decreased allogeneic transfusion rates, particularly for procedures lasting over 6 h and where expected blood loss was over 30% of blood volume.

The use of ABT drains has also become more popular in orthopaedic procedures over the last ten years. Post-operative collection and reinfusion of red cells has been shown to reduce the need for donor blood transfusion in both knee and hip arthroplasty^[9,10]. In spinal surgery, re-transfusion drains have been shown to be safe and reduce the need for donor blood during short segment fusion in adults^[6]. The use of ABT drains in scoliosis surgery has not previously been reported.

In our study both cell salvage and ABT drains were sequentially introduced and lead to a modest, but not statistically significant, reduction in the percentage of patients receiving an allogeneic transfusion. However, those patients who were transfused were exposed to significantly fewer allogeneic units (donor exposures), falling from 3.82 to 2.19 units when both intraoperative cell salvage and ABT drains were used.

In common with all retrospective studies, it is impossible to adjust for all confounders, both known and unknown. The reduction in donor blood exposure we achieved occurred alongside a number of other changes to minimise blood loss during surgery during the study period. There was a trend towards less invasive surgical procedures in groups B and C, partly due to changes in the implant systems used which provided a greater ability to achieve an adequate curve correction using a single stage posterior procedure. There was also a trend towards an increased use of antifibrinolytics in group C (30% of whom received tranexamic acid), and there is good evidence that this can lead to lower transfusion rates in paediatric scoliosis surgery^[11].

In the absence of randomised controlled trials, the results of retrospective series, such as this, must be interpreted with caution. However, the methods described were found to be safe and practical in routine practice and contributed to a clinically and financially worthwhile reduction in the use of donor red cells.

We also observed a fall in the length of inpatient hospital stay in patients in group C. This is probably multifactorial, including changes to surgical practice that lead to earlier postoperative mobilisation and discharge. However, changes in transfusion practice may have contributed as several previous studies have reported reduced length of hospital stay in orthopaedic patients

Table 3 Cost analysis comparing groups A, B and C

	Group A donor blood only	Group B cell salvage	Group C cell salvage + ABT drain
Number of RBC units transfused	84	50	37
Total cost of RBC transfusion (£)	10416	6200	4588
Number of patients receiving cell salvage	0	30	30
Total cost of cell Salvage (£)	0	2100	2100
Number of patients receiving ABT drains	0	0	30
Total cost of ABT drains (£)	0	0	1620
Overall transfusion cost	10416	8300	8308
Number of patients	37	30	30
Overall cost per patient (£)	281.51	276.67	276.93

RBC = £124; cost of cell salvage ACD-A, collection and processing kit = £70; costs of a single ABT drain = £54. ABT: Autologous blood transfusion; RBC: Cost of one unit of blood; ACD-A: Acid citrate-dextrose anticoagulant.

exposed to less donor blood^[12].

Cost analysis

There is good evidence to suggest that cell salvage is a cost effective method to reduce the need for donor blood transfusion^[13]. A cost analysis of the data presented in our study is detailed in Table 3. This confirms that the use of cell salvage alone and the use of cell salvage with ABT drains was cost neutral in our study.

In conclusion, to our knowledge this is the first report of ABT drain use in scoliosis surgery and suggests that its use is both safe and cost effective. When used as part of a systematic programme to minimise blood loss during surgery, the use of ABT drains and intra-operative cell salvage lead to a reduced need for donor blood transfusion in patients undergoing scoliosis surgery in our unit. The lowest donor blood transfusion rates were seen when both modalities were used, leading to a 6% reduction in the number of donor transfusion episodes and a significant reduction in the mean units of red cells transfused per episode (2.19 compared to 3.82). The programme proved practical and cost-effective to implement in a busy surgical unit.

COMMENTS

Background

Autologous blood reinfusion techniques have become commonplace in many fields of orthopaedic surgery, especially in hip and knee arthroplasty. These techniques can reduce the need for donor blood and the risks that accompany its use.

Research frontiers

Intra-operative cell salvage has been associated with reduced transfusion requirements in scoliosis surgery, but the use of post-operative reinfusion devices is not well documented.

Innovations and breakthroughs

To the authors' knowledge this is the first report of autologous blood transfusion (ABT) drain use in scoliosis surgery and suggests that its use is both safe and cost effective.

Applications

When used as part of a systematic programme to minimise blood loss during

surgery, the use of ABT drains and intra-operative cell salvage can reduce the need for donor blood transfusion in patients undergoing scoliosis surgery.

Terminology

ABT and allogeneic red cell transfusion.

Peer-review

This is a case control study reporting the use of ABT drains in scoliosis surgery and suggests that its use is both safe and cost effective. When used as part of a systematic programme to minimise blood loss during surgery, ABT drains and intra-operative cell salvage lead to a significantly reduced need for donor blood transfusion in patients undergoing scoliosis surgery.

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Retrospective Study

Windsurfing vs kitesurfing: Injuries at the North Sea over a 2-year period

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Abstract

AIM

To analyze all windsurfing and kitesurfing (kiteboarding) injuries presented at our coastal hospital over a 2-year period.

METHODS

Twenty-five windsurfers (21 male; aged 31 ± 8 years) and 32 kitesurfers (23 male; aged 29 ± 11 years) presented at our hospital during the 2-year study period. Various injury data were recorded, including transport to hospital and treatment. After a median follow-up of 16 mo (range, 7-33 mo), 18 windsurfers (72%) and 26 kitesurfers (81%) completed questionnaires on the trauma mechanisms, the use of protective gear, time spent on windsurfing or kitesurfing, time to return to sports, additional injuries, and chronic disability.

RESULTS

Most patients sustained minor injuries but severe injuries also occurred, including vertebral and tibial plateau fractures. The lower extremities were affected the most, followed by the head and cervical spine, the upper

extremities, and the trunk. The injury rates were 5.2 per 1000 h of windsurfing and 7.0 per 1000 h of kitesurfing ($P = 0.005$). The injury severity was the same between groups ($P = 1.0$). Less than 30% of the study population used protective gear. Kitesurfers had a higher number of injuries, and required transport by ambulance, inpatient hospital stay and operative treatment more often than windsurfers, but these differences were not statistically significant ($P > 0.05$). The median time to return to windsurfing and kitesurfing was 5 and 4 wk, respectively ($P = 0.79$). Approximately one-third of the patients in each group experienced chronic symptoms.

CONCLUSION

Kitesurfing results in a significantly higher injury rate than windsurfing in the same environmental conditions but the severity of the injuries does not differ.

Key words: General sports trauma; Extreme sports; Surfing; Epidemiology; Prevention

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Core tip: To our knowledge, this is the first study that directly compares kitesurfing and windsurfing in the same weather and environmental conditions, giving a unique insight in the injuries associated with these sports. Kitesurfing resulted in a significantly higher injury rate than windsurfing in the same environmental conditions but the severity of the injuries did not differ. The presented results may assist the health-care professional and the athlete in taking measures to prevent injuries and in advising or choosing the safer sport.

van Bergen CJA, Commandeur JP, Weber RIK, Haverkamp D, Breederveld RS. Windsurfing vs kitesurfing: Injuries at the North Sea over a 2-year period. *World J Orthop* 2016; 7(12): 814-820 Available from: URL: <http://www.wjgnet.com/2218-5836/full/v7/i12/814.htm> DOI: <http://dx.doi.org/10.5312/wjo.v7.i12.814>

INTRODUCTION

Windsurfing has a long history as a popular water sport since its introduction in 1971. It has been recognized as an Olympic sport since 1984. Various reports on windsurfing injuries have been published since 1988^[1-6]. Due to the technical developments, lighter and more sophisticated material, windsurfers have more possibilities to push out their frontiers, possibly exposing them to more dangerous situations (Figure 1).

Kitesurfing (or kiteboarding) is possibly the fastest growing water sport today. Using a small board and a kite of up to 20 m², surfers can achieve huge jumps and great speeds up to 55 knots (Figure 2). This extreme sport has been increasingly popular since it was developed in the late 1990s. In 2006, the total number



Figure 1 Windsurf action.



Figure 2 Kitesurf action.

of kitesurfers worldwide was estimated at around 210000, with 114465 inflatable kites sold that same year according to an in-depth worldwide industry survey^[7]. By 2008, the participation rate growth was between 35% and 50%^[7]. Despite the increasing popularity and possible dangers of this sport, little is known about injury prevalence or prevention^[8]. However, some reports did analyse kitesurfing injuries and even reported fatal accidents^[9-12].

Windsurfing and kitesurfing are sports that have many fundamentals in common; wind is used to generate forward motion on the water, opening the possibility of performing tricks both on the water and in the air. However, the equipment used for these sports are completely different. Windsurfers use a sail with a mast directly connected to the board, whereas kitesurfers use a kite, which is attached to the athlete by multiple lines. Because of these differences, rates and severity of injuries may differ. Interestingly, these sports have never been compared in one study or in the same weather and environmental conditions. Providing comparative data may assist the athlete in taking measures to prevent injuries and in choosing the safer sport.

The aim of this study was to analyze and compare all kitesurfing and windsurfing injuries at our hospital over a 2-year period. We hypothesized that kitesurfing results in more frequent and more severe injuries than windsurfing.



Figure 3 Picture of the popular spot at the North Sea on a typical day, showing windsurfers and kitesurfers mixed.

MATERIALS AND METHODS

This is a retrospective study carried out according to a study protocol approved by the local medical ethics committee. All patients with windsurfing or kitesurfing injuries who presented at our hospital from September 2009 through September 2011 were included. September 2009 was chosen as the starting point of the study since our hospital then introduced the Electronic Patient Data (EPD) system, allowing for reliable inclusion of patients by search criteria.

A place at the North Sea close to our hospital is probably the most popular and most visited kitesurfing and windsurfing location of The Netherlands (Figure 3). Wind speeds are ≥ 4 Beaufort most of the days (60%-70%), and there are waves of up to 4 m. Injured individuals from this place typically present at the Emergency Department of our hospital because it is very close, between the beach and the highway.

Patient identification

The EPD was searched for windsurfing and kitesurfing injuries using the terms "surf" and "kite" with any possible prefix or extension. Thus, each patient with any of these terms - also as part of another word - mentioned in the patient charts was identified.

Data collection

Each patient's chart was manually reviewed. Numerous patient data were recorded, including gender, date of birth, injury data [trauma mechanism, type of injury, affected body part(s)], site of injury (beach or water), transport to hospital, type of treatment, submission to hospital, or outpatient treatment.

Additionally, the patients were sent questionnaires with prepaid envelopes, accompanied by an information letter, containing the following items: (1) skill level (beginner, intermediate, advanced, expert); (2) number of years of experience; participant hours (number of days a year and mean number of hours a day); (3) cause of the injury; wind speed; (4) use of protective gear (helmet, vest) at the time of injury; (5) availability and use of a quick-release system (kitesurfing only);

(6) time to return to windsurfing or kitesurfing; (7) additional injuries during the study period; and (8) chronic symptoms.

For windsurfing, the skill level was assessed by the study participants according to the following scale: Beginner, learning to windsurf, basic tacking and gybing; intermediate, basic beach starting, planing techniques, use of foot straps and harness; advanced, water starting, carve gybe, launching and landing in waves; expert, advances aerial and freestyle manoeuvres^[1]. For kitesurfing, the skill level was assessed by the study participants according to the following scale: Beginner, starting and landing the kite with help, downwind rides on the water; intermediate, easy manoeuvres such as jibes, upwind rides, and small jumps; advanced, high jumps and transition jumps; expert, high jumps with rotation and board-off jumps^[9].

The questionnaire included any information that could not be retrieved from the charts for that specific patient.

The severity of the injury was classified as follows^[9]: (1) catastrophic, injuries leading to permanent disability or death; (2) severe, injuries resulting in absence from kitesurfing or windsurfing for more than 6 wk; (3) medium, injuries resulting in absence from kitesurfing or windsurfing for more than 1 d; and (4) mild, injuries resulting in incapacity to train or compete on a normal basis.

Statistical analysis

Data with a normal distribution are presented as mean \pm standard deviation. Data with a skewed distribution are presented as median and range. Injury rates were calculated per 1000 h of windsurfing or kitesurfing. Various data were compared between windsurfing and kitesurfing with use of parametric or nonparametric statistical tests, depending on the number and distribution of data (Student *t*-test for age; Mann-Whitney *U* test for other continuous data; χ^2 or Fisher's Exact test for nominal and categorical data, where appropriate). A *P*-value of 0.05 was considered to be statistically significant.

RESULTS

The computer-generated search and manual chart review identified 57 patients (25 windsurfers and 32 kitesurfers) who had presented at our Emergency Department during the study period.

Baseline characteristics and protective gear

There were 44 male and 13 female patients with a mean age of 30 ± 10 (range, 11-57) years. Eighteen windsurfers (72%) and 26 kitesurfers (81%) completed the questionnaire. The median time to follow-up was 16 mo (range, 7-33 mo). Most baseline characteristics were not significantly different between the groups, but windsurfers had significantly more experience and

Table 1 Baseline characteristics

	Windsurfing	Kitesurfing	P-value
Age (yr)	31 ± 8	29 ± 11	0.437
Gender (No. and % of patients)			0.279
Male	21 (84%)	23 (72%)	
Female	4 (16%)	9 (28%)	
Experience (yr)	15 (0-30)	2 (0-12)	< 0.001
Annual participant hours	106 (1-480)	70 (1-810)	0.184
Skills level			0.001
Beginner	1 (4%)	11 (34%)	
Intermediate	0 (0%)	6 (19%)	
Advanced	9 (36%)	3 (9%)	
Expert	8 (32%)	6 (19%)	
Wind speed (Bft)	6 (2-8.5)	5 (2-9)	0.362
Protective gear (No. and % of patients)	3 (17%)	7 (28%)	0.480

Data are presented as mean ± standard deviation in case of normal distribution, and as median (range) in case of skewed distribution.

a higher skills level than kitesurfers (Table 1). Only 17% in the windsurf group and 28% in the kitesurf group used protective gear such as helmets and body protectors (impact vests). A quick-release system was present in 24 (92%) out of 26 kitesurfers, but it was deployed by only eight (31%).

Injury rates

In the windsurf group, 32 injuries occurred during a total of 6146 windsurfing hours during the study period of 2 years. This corresponds with an overall injury rate of 5.2 injuries per 1000 h of windsurfing (Table 2). In the kitesurf group, 49 injuries occurred during a total of 6978 kitesurfing hours in 2 years, which corresponds with an overall injury rate of 7.0 injuries per 1000 h of kitesurfing. The difference between the groups was statistically significant ($P = 0.005$).

Injury patterns and severity

In the windsurf group, most injuries generally occurred during difficult manoeuvres such as forward and back loops, or due to unexpected wind gusts. Likewise, in the kitesurf group most injuries were caused by high jumps or wind gusts; additionally, many of the injuries in this group were caused by technical difficulties controlling the kite and/or board. Only few injuries in both groups were the result of collisions with other surfers.

Most patients in both groups sustained minor injuries, such as contusions, lacerations, and ankle sprains. However, numerous serious injuries also occurred. In the windsurf group, serious injuries included fractures of the tibial plateau and os trapezium. In the kitesurf group, there were fractures of the dens, L3 vertebral body, olecranon, scaphoid, tibial plateau, and talus. The lower extremities were affected the most, followed by the head and cervical spine, the upper extremities, and the trunk, respectively (Table 2).

The severity of the injuries was graded as medium or severe in most cases of both groups (Table 2).

Table 2 Injury data

	Windsurfing	Kitesurfing	P-value
Number of injuries per 1000 h	5.2	7	0.005
Affected body site (No. and % of patients)			0.413
Head and cervical spine	9 (36%)	11 (34%)	
Upper extremity	6 (24%)	5 (16%)	
Trunk and thoracolumbar spine	0 (0%)	3 (9%)	
Lower extremity	10 (40%)	13 (41%)	
Injury severity (No. and % of patients)			1.000
Mild	3 (17%)	5 (19%)	
Medium	6 (33%)	9 (35%)	
Severe	8 (44%)	10 (39%)	
Catastrophic	1 (6%)	2 (8%)	
Location of accident (No. and % of patients)			0.014
Water	17 (94%)	15 (58%)	
Beach	1 (6%)	11 (42%)	
Type of transport (No. and % of patients)			0.161
Ambulance	2 (11%)	8 (31%)	
Own transport	16 (89%)	18 (69%)	
Inpatient hospital stay (No. and % of patients)	1 (4%)	5 (16%)	0.215
Operative treatment (No. and % of patients)	1 (4%)	4 (13%)	0.372
Total number of injuries during study period (No. and % of patients)			0.237
1	20 (80%)	19 (59%)	
2	3 (12%)	9 (28%)	
3	2 (8%)	4 (13%)	
Return to sports in weeks (median and range)	5 (0-chronic)	4 (0-chronic)	0.792
Chronic symptoms (No. and % of patients)	7 (39%)	8 (31%)	0.576

The injury severity was neither significantly different between the groups, nor associated with patient age, skills level, or the use of protective gear.

Although kitesurfers had a higher number of injuries and required transport by ambulance, inpatient hospital stay and operative treatment more frequently than windsurfers, these differences were not statistically significant (Table 2). Kitesurfing accidents happened significantly more on the beach than windsurfing accidents ($P = 0.014$).

The median time to return to sports was approximately 1 mo (Table 2). About one-third of the patients experienced chronic symptoms due to the accident at the time of follow-up (Table 2).

DISCUSSION

Windsurfing and kitesurfing are sports that are closely connected to the elements. As such, these sports have a certain level of unpredictability. These properties might be the reasons why these sports are so popular. However, the unpredictability might also impose a key factor in the risks associated with these sports. Therefore, finding typical trauma mechanisms and

handles for prevention is challenging.

Injury rates

The main objective of this study was to investigate the incidence of injuries among both windsurfers and kitesurfers and thus generate an objective image of the risk incorporated in these sports. The results suggest that kitesurfing has a significantly higher injury rate than windsurfing. We found an injury rate of 5.2 per 1000 h of windsurfing compared to 7.0 injuries per 1000 h of kitesurfing.

The groups were comparable with regard to age, gender, weather conditions, and participant hours. However, the average experience and skills level were higher amongst windsurfers. One might speculate that less experienced athletes sustain more severe injuries, which could partially explain the outcomes of our study. However, another study found that having more experience increases the risk of significant injuries among recreational surfers^[13]. Therefore, the explanation for the different injury rates between the groups in our study might be found in the technical differences between sports and in the risks associated with the gear used, rather than the experience of the sportsmen.

In the literature, a few previous studies reported injury rates. A review of the literature identified an overall injury rate of 5.9 to 7.0 injuries per 1000 noncompetitive kitesurfing hours^[12]. A prospective kitesurf study showed an injury rate of 7 injuries per 1000 h of practicing the sport^[9]. This rate is the same as that found for kitesurfing in our retrospective study. For windsurfing, however, lower injury rates have been reported in retrospective studies. McCormick *et al*^[4] in 1988 found an injury rate of 0.22 injuries per 1000 h of practicing windsurfing. Other reported injury rates in windsurfing ranged from 1.1 to 2.0 injuries/person per year^[1,6]. These rates are somewhat lower than ours. Possible interpretations for these differences might be found in the different weather and water conditions between studies and in an evolution of the sport since 1988, possibly adding more risk. Furthermore, the rates found in our study might be overestimated because only injured individuals were included.

Both windsurfing and kitesurfing clearly have lower injury rates compared with other extreme sports and contact sports. For example, American football, motocross, and football (soccer) have injury rates of 36, 23, and 19 per 1000 h, respectively^[14-16]. However, conclusions from comparing injury rates between different studies should be interpreted with caution due to the lack of a uniform injury definition in sports science.

Injury patterns and severity

We found no difference in injury severity between the kitesurf and windsurf groups. In both groups, the majority of injuries were relatively minor, with the highest prevalence in the lower extremities. Previous studies

showed comparable patterns, severity and location of injuries^[1,9,11]. The injury severity was not associated with patient age, skills level or the use of protective gear. However, the study may not have enough numbers in the subgroups to support this with confidence (causing a statistic error type II).

In windsurfing, the most common causes of injuries were technical manoeuvres and unexpected wind gusts. The leading causes of injuries in kitesurfing were jumps, wind gusts and lack of controlling the kite. These findings concur with other studies^[9,11]. The fact that kitesurfers lost control more often might be due to less experience, a difference in the technical aspects of controlling a kite, or a larger surface of a kite than a windsurf sail.

Dyson *et al*^[1] reported collision with equipment to be the major contributor to injuries of windsurfers. Nickel *et al*^[9] suggested separating designated areas for windsurfers and kitesurfers to prevent collisions. However, in our study that addressed both sports, collisions were reported infrequently. According to these results, separation of windsurfing and kitesurfing areas seems unnecessary.

Injury prevention

Little is known about the effect of protective gear in the prevention of windsurf and kitesurf related injuries. The equipment of both sports has evolved during the past decades. In windsurfing, the changes have been subtle. Booms are smaller and foot straps are designed for easier exit. Given the fact that kitesurfing is a relatively young sport, equipment evolution is most notable in this sport; the major advancement being a quick-release safety system, enabling kitesurfers to release the kite easily in case of emergency (Figure 4). Although most kitesurfers had a quick-release system on their kite, the minority deployed it. The use of protective gear such as helmets, impact vests and flotation devices has been accepted more and more. However, only still a small percentage of the athletes use protective equipment, as is shown in our study as well as previous studies^[8,11]. Better use of quick-release systems and protective gear offers possibilities for education and counseling.

In kitesurfing, significantly more accidents occurred on the beach (42% vs 6% for windsurfing). Launching and landing the kite on the beach are the times that kitesurfers are classically at risk, particularly if inexperienced, as an accident at this time means the kiter lands on a hard surface (beach) as opposed to water. These data provide another handle for prevention, with focus on education and awareness regarding the risk of injury during launching and landing the kite.

Other sources of injury prevention are exercises and warming up. Dyson *et al*^[1] reported a relatively high incidence of lower back problems amongst windsurfers and stated a possible role for back exercises in the prevention of these injuries. Lundgren *et al*^[17] found a protective effect of a warming-up before starting a

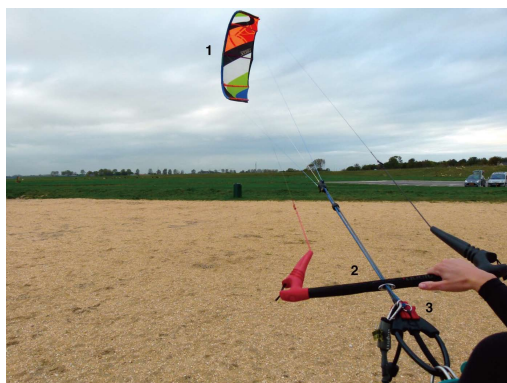


Figure 4 Picture showing (1) the kite, (2) the handlebar, and (3) the quick-release system.

kitesurf session.

Limitations

The identification of study patients through searching the EPD might not have detected patients in case the words “surf” or “kite” were not used in the patient’s file. The injury rates in this study were calculated retrospectively. It might be difficult for participants to estimate the hours they practiced sports during the time period of the study, although any uncertainty would affect both groups. Furthermore, only athletes who presented at our hospital were included. It might be conceivable that these windsurfers and kitesurfers are more prone to injuries than others. The total amount of windsurfers and kitesurfers is unknown. These factors might result in injury rates that are reported higher than they actually are. However, the results for kitesurfers are the same as those found in a prospective study^[9], which might support the validity of our findings. Lastly, the group sizes in our study were limited. This may partially explain the fact that differences between groups concerning transport by ambulance, inpatient hospital stay and operative treatment did not reach statistical significance. Future studies following large groups of surfers prospectively throughout multiple seasons may provide the most reliable data to analyse and compare injuries.

In conclusion, to our knowledge, this is the first study that directly compares kitesurfing and windsurfing in the same conditions, giving a unique insight in the injuries associated with these sports during a 2-year period. The outcomes suggest that kitesurfing has a higher injury rate than windsurfing. The severity of injuries was similar.

COMMENTS

Background

Windsurfing has been a popular water sport for over three decades. Kitesurfing is possibly the fastest growing water sport today. Windsurfing and kitesurfing are sports that have many fundamentals in common; wind is used to generate forward motion on the water, opening the possibility of performing tricks both on the water and in the air. Thus, both extreme sports pose the athlete at risk for

injuries. However, the equipment used for these sports are completely different. Windsurfers use a sail with a mast directly connected to the board, whereas kitesurfers use a kite, which is attached to the athlete by multiple lines. Because of these differences, rates and severity of injuries may differ.

Research frontiers

Various reports on windsurfing injuries have been published since 1988. Some other reports have analysed kitesurfing injuries and even reported fatal accidents. Current research aims to further identify trauma mechanisms and provide handles for prevention.

Innovations and breakthroughs

To the authors' knowledge, this is the first study that directly compares kitesurfing and windsurfing in the same conditions, giving a unique insight in the injuries associated with these sports during a 2-year period. The outcomes suggest that kitesurfing has a higher injury rate than windsurfing. Injury data are presented and preventive measures are suggested.

Applications

The provided comparative data may assist the athlete and sport physician in taking measures to prevent injuries and in choosing the safer sport. This study forms a base for future research investigating larger groups of surfers prospectively to provide the most reliable data on injuries.

Peer-review

It is an interesting manuscript on investigating and comparing windsurf vs kite surf in the same conditions giving a description of the associated injuries. Overall the structure of the manuscript is very good and the language of the manuscript reaches the standard of publishing.

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Retrospective Study

Seasonal variation in adult hip disease secondary to osteoarthritis and developmental dysplasia of the hip

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Author contributions: Sueyoshi T primary contributed to the work and wrote the manuscript; Ritter MA did the surgery and organized the study; Davis KE did the data collection and statistics; Loder RT made some of developmental dysplasia of the hip comments and gave suggestion.

Institutional review board statement: The study was reviewed and approved by the St. Francis Hospital Institutional Review Board.

Informed consent statement: All study participants or their legal guardian provided consent about personal and medical data collection prior to study enrolment.

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Data sharing statement: The original anonymous dataset is available on request from the corresponding author at jrsiresearch1@gmail.com.

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Abstract

AIM

To determine if there was a seasonal variation in adults undergoing total hip arthroplasty for end stage hip disease due to osteoarthritis (OA) or sequelae of developmental dysplasia of the hip (DDH).

METHODS

The total hip registry from the author's institution for the years 1969 to 2013 was reviewed. The month of birth, age, gender, and ethnicity was recorded. Differences between number of births observed and expected in the winter months (October through February) and non-winter mo (March through September) were analyzed with the χ^2 test. Detailed temporal variation was mathematically assessed using cosinor analysis.

RESULTS

There were 7792 OA patients and 60 DDH patients who underwent total hip arthroplasty. There were more births than expected in the winter months for both the DDH ($P < 0.0001$) and OA ($P = 0.0052$) groups. Cosinor analyses demonstrated a peak date of birth on 1st October.

CONCLUSION

These data demonstrate an increased prevalence of DDH and OA in those patients born in winter.

Key words: Seasonal trend; Winter; Osteoarthritis; Birth month; Developmental hip dysplasia

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Core tip: The purpose of this study was to determine if there was a seasonal variation undergoing total hip arthroplasty for osteoarthritis (OA) or developmental dysplasia of the hip (DDH). Differences between number of births observed and expected in the winter months and non-winter months were analyzed with the χ^2 test. There were 7792 OA and 60 DDH, and more births than expected in the winter months for both the DDH ($P < 0.0001$) and the OA ($P = 0.0052$) cohorts. These data clearly demonstrated an increased prevalence of DDH and OA in those patients born during the winter months.

Sueyoshi T, Ritter MA, Davis KE, Loder RT. Seasonal variation in adult hip disease secondary to osteoarthritis and developmental dysplasia of the hip. *World J Orthop* 2016; 7(12): 821-825 Available from: URL: <http://www.wjgnet.com/2218-5836/full/v7/i12/821.htm> DOI: <http://dx.doi.org/10.5312/wjo.v7.i12.821>

INTRODUCTION

Total hip arthroplasty (THA) is a standard, successful treatment for end stage hip disease. The etiology of such disease is frequently due to osteoarthritis (OA), osteonecrosis/avascular necrosis (ON/AVN), rheumatoid arthritis (RA) and developmental dysplasia of the hip (DDH). Studies of hip osteoarthritis suggest that childhood deformities or residuals of childhood hip disease often experience accelerated or premature joint degeneration^[1,2]. DDH is a wide spectrum of deformity ranging from mild acetabular dysplasia to complete and fixed dislocation^[3]. Many studies have noted that DDH is more frequent in patients born in the colder months^[4-6]. DDH with residual acetabular dysplasia is often followed by OA of the hip. OA has also been reported to be more frequent in people born in the winter^[7]. The authors wished to determine if there was any seasonal variation in adults who are treated for OA and DDH in the Midwest region of the United States. This will further augment a previous study which noted a seasonal variation in children with DDH in this region^[3].

MATERIALS AND METHODS

A retrospective review of all THAs performed from November 1969 through November 2013 at the author's institution was performed. This review was approved by the local Institutional Review Board. There were 10572 THAs in 8579 patients that met the initial inclusion criteria of primary OA, RA, ON or DDH diagnosis. There were 192 THAs in 157 patients that did not have adequate

Table 1 Month of birth for 7852 patients undergoing total hip arthroplasty for osteoarthritis and sequelae of developmental hip dysplasia

Month of birth	DDH		OA		Temperature (°F)	Precipitation (ins)
	n	%	n	%		
January	10	17	745	9.6	26	2.5
February	6	10	622	8	30.5	2.3
March	3	5	617	7.9	40.7	3.4
April	3	5	585	7.5	51	3.9
May	3	5	582	7.5	61.4	4.5
June	4	7	613	7.9	70.5	4.2
July	3	5	667	8.6	74.3	4.2
August	0	0	675	8.7	72.2	3.9
September	3	5	682	8.8	65.2	3.1
October	14	23	702	9	53.6	3
November	3	5	610	7.8	42.3	3.4
December	8	13	692	8.9	31.2	3.1
Total	60	100	7792	100		41.5

OA: Osteoarthritis; DDH: Developmental hip dysplasia.

demographic data and were excluded from any analysis. The diagnosis was OA in 7792 patients, ON/AVN in 523, RA in 204, and DDH in 60. Those with ON/AVN and RA were then excluded, leaving 9671 THAs in 7852 patients for analysis. Date of birth, age, and gender were gathered from the database of total joint surgeries performed at the authors' institution based on medical records and operation note data at the time of surgery.

The months of the year were divided into winter and non-winter. Winter was defined as October through February, and non-winter as March through September based on local temperatures. The source for the average monthly temperature and precipitation was from the Indiana State Climate Office in Purdue University (<https://climate.agry.purdue.edu/climate/facts.asp>) (Table 1). Differences by winter and non-winter months were evaluated for both the DDH and OA groups. Temporal variation was analyzed using cosinor analysis (see below).

Statistical analysis

Continuous data are reported as the mean \pm standard deviation. Categorical data are reported as frequencies or percentages. Statistical differences between continuous variables were analyzed by the student's *t*-test and between categorical variables by the Pearson's χ^2 test. Statistical Analysis System (SAS) version 9.2 (Cary, NC) was used for *t*-tests and categorical analysis where the expected value was set as the null hypothesis. The Statistical Package for Social Sciences (SPSS) version 12.0 for Windows (SPSS Inc., Chicago, IL, United States) was used for further statistical analysis. The data were subjected to cosinor analysis in an effort to mathematically define any seasonal variation when present. Cosinor analysis^[8,9] determines the best mathematical fit of the data to an equation defined by $F(t) = M + A \cos(\omega t + \phi)$, where *M* = the mean level (termed mesor), *A* = the

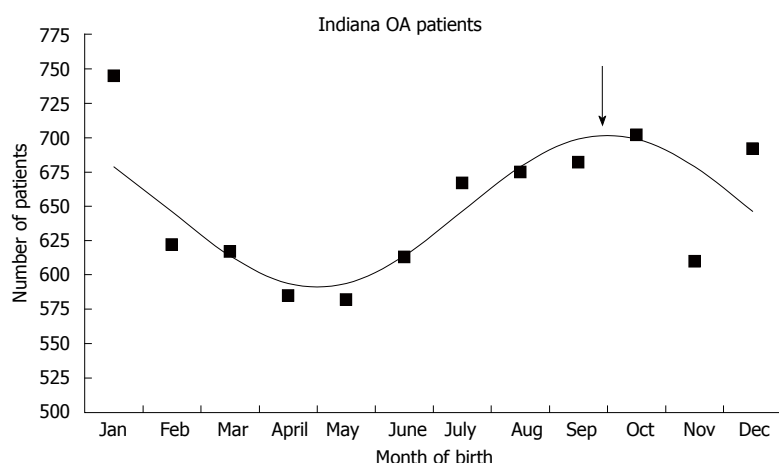


Figure 1 The cosinor fit for Indiana patients with hip osteoarthritis and month of birth. The best fit is represented by the equation: Number of OA births = $646.4 + 55.3 [\cos(36t - 18) - 324]$, where $t = 1$ is January, 2 is February, 3 is March, etc. This was statistically significant ($r^2 = 0.56$, $P = 0.025$). The peak is 1st October (solid arrow). The data points are the black triangles and the best fit represented by the bold black line. OA: Osteoarthritis.

amplitude of the cosine curve, ϕ = acrophase (phase angle of the maximum value), ω = the frequency (which for monthly analysis is $360^\circ/12 = 30^\circ$), and t = time (which in this case is each month). The overall P and r^2 value for the distribution are given for the rhythmic pattern described by the cosinor equation for M, A, and ϕ . The data was analyzed for the entire period of 12 mo as well as decreasing increments of 1 mo. A best monthly fit may not be over a period of 12 mo, but a different time span (e.g., 7 or 6 mo periodicity). Cosinor analyses were performed with ChronoLab 3.0™ software (see Acknowledgement). For all statistical analyses the level of significance was set at $P < 0.05$.

RESULTS

The average age at the time of THA was 55.4 ± 13.6 years in the DDH group and 67.9 ± 11.4 years in the OA group. This age difference was statistically significant ($P < 0.0001$). There were 49 females (81.7%) and 11 males (18.3%) in the DDH and 4160 females (53.4%) and 3632 males (46.6%) in the OA group ($P < 0.0001$). The distribution of birth months for the 7792 OA and 60 DDH patients can be found in Table 1. The number of births in the winter months was much higher than expected in both the DDH [41 of 60 (68.3%) actual, 25 of 60 (41.7%) expected, $P < 0.0001$] and the OA [3371 of 7792 (43.3%) actual, 3247 of 7792 (41.7%) expected, $P < 0.0052$] groups. Cosinor analysis demonstrated an excellent fit for the OA group with a peak on October 1 (Figure 1). There was no good cosinor fit for the DDH group, likely due to the small number.

DISCUSSION

This study demonstrates that both adult DDH and OA patients with end stage hip disease are more frequently born in the colder months. This is similar to pediatric DDH in the previous report^[3] and adult Japanese OA^[7].

Children with DDH undergoing operative treatment demonstrated a peak birth on 15th October^[3], and is essentially the same as the adult OA patients peak on 1st October. Nagamine *et al.*^[7] noted that adult hip osteoarthritis in the Japanese population was more prevalent when the patient was born in the colder months. The authors subjected their data to cosinor analysis and found a peak on 20th January (Figure 2). The Japanese OA peak of 20th January is identical to the 26th January peak in Japanese children with DDH from Kochi, Japan^[3,10].

There are certain limitations to this study. This is a retrospective study and diagnosis of DDH and OA was determined mainly by radiographs and patient history. The diagnosis of OA includes both primary and secondary OA^[11]. Patients undergoing THA are usually at the end stage of hip arthritis making it difficult to distinguish primary from secondary OA. Some patients will have shallow acetabulae, but once the joint space disappears at the lateral aspect of the acetabulum, it is impossible to differentiate between primary and secondary OA. The patient's history may help, but most patients with DDH treated in infancy with a Pavlik harness or other abduction bracing may not be aware of that history; they would not be able to remember it due to their young age at the time of treatment and their parents may not have told them of such treatment. Although the DDH group was small, we still believe that the findings are valid due to the highly significant $P < 0.0001$.

These findings have several possible explanations. First, it is very likely that many of the OA patients had underlying, subtle mild DDH (e.g., minimal acetabular dysplasia). The population of OA, technically the secondary OA, may have just more of DDH than what the authors think. Another possible explanation is that both DDH and OA patients share some common, but as yet unknown, etiology. The possible factors/etiologies that can be invoked are pre-/perinatal maternal factors as well as postnatal environmental factors.

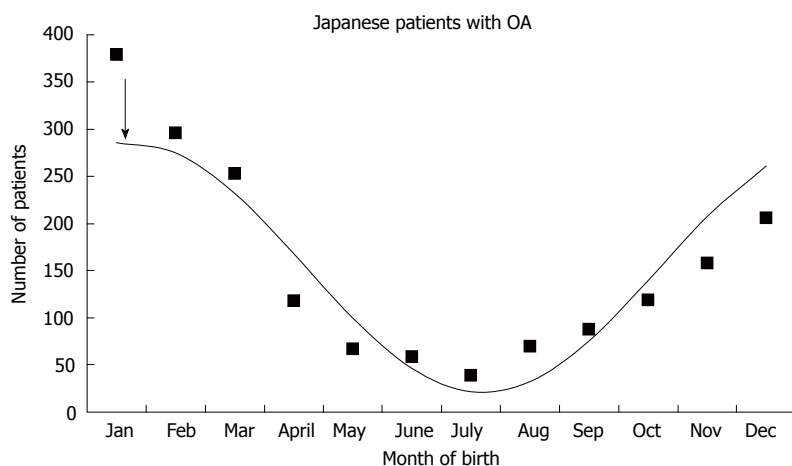


Figure 2 The cosinor fit for Japanese patients with osteoarthritis and month of birth. Data extracted from the study of^[7]. The best fit is represented by the equation: number of OA births = $153.7 + 132.7 (\cos 30t - 15) - 21$, where $t = 1$ is January, 2 is February, 3 is March, etc. This was statistically significant ($r^2 = 0.83$, $P < 0.001$). The peak is 20th January (solid arrow). The data points are the black triangles and the best fit represented by the bold black line. OA: Osteoarthritis.

Potential pre-/perinatal factors are a breech birth or difficult delivery^[12]. First born and high-birth-weight babies are at an increased risk of DDH, but those do not exhibit seasonal trend^[6]. Obstetric pelvic insufficiency shows a seasonal variation with a peak in November to December^[13]. In the current study, the authors did not investigate the patients' birth history (pregnancy number or breech/vertex delivery). Since there been no report showing a relationship between osteoarthritis and breech/difficult delivery, it is most likely that residual DDH (minimal acetabular dysplasia) influenced the incidence of OA.

Another potential perinatal factor is relaxin, which stimulates collagenase and alters connective tissue^[14,15]. Collagen metabolism is altered in DDH^[16] with increased joint laxity^[17]. However, the correlation between DDH and relaxin is controversial^[18,19]. In addition, no seasonal variation in relaxin levels has been shown leading to the conclusion that relaxin is not a critical factor. Another possible factor is vitamin D, which is well known to have a seasonal variation with peak levels in the summer^[20,21]. Low maternal levels of vitamin D result in low birth weight infants and increased levels in heavy infants^[22,23]. A report from Norway suggests hip and knee osteoarthritis is more common in those born in the spring months^[24], perhaps due to vitamin D issues.

In conclusion, this study demonstrated that patients undergoing THA for end stage hip disease due to OA or the sequelae of DDH are more commonly born in the colder months. This confirms previous reports noting the same phenomenon in both adult OA and childhood DDH. Further research will be needed to understand this association.

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COMMENTS

Background

Winter birth has been associated with developmental dysplasia of the hip (DDH) and it was reported recently to be related to hip osteoarthritis (OA). This study aims to investigate the epidemiology of DDH and OA according to the registry of total hip arthroplasty in the authors' institution.

Research frontiers

To the authors' knowledge, this is the first study to present both OA and DDH were associated with winter birth. Reviewing the registry over 40 years, the average age at the surgery was statistically younger in DDH patients than in OA patients. Both DDH and OA patients were more born in winter months than in non-winter months.

Innovations and breakthroughs

Several reports have shown DDH is often caused by swaddling during infancy, which was reported to be one reason DDH is associated with winter birth. The study demonstrated OA also was associated with winter birth. The etiology is still unknown why winter birth influences hip OA, but the study will contribute to elucidate the mechanism of OA and the prevention of hip surgery in the future.

Peer-review

It is an interesting and well-written article.

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Observational Study

Effect of introducing an online system on the follow-up of elbow arthroplasty

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Abstract

AIM

To evaluate the effect of introducing a structured online follow-up system on the response rate.

METHODS

Since June 2015 we have set up an electronic follow-up system for prosthesis in orthopedic patients. This system allows prospective data gathering using both online and paper questionnaires. In the past all patients received questionnaires on paper. This study includes only patients who received elbow arthroplasty. Response rates before and after introduction of the online database were compared. After the implementation, completeness of the questionnaires was compared between paper and digital versions. For both comparisons Fisher's Exact tests were used.

RESULTS

A total of 233 patients were included in the study. With the introduction of this online follow-up system, the overall response rate increased from 49.8% to 91.6% ($P < 0.01$). The response rate of 92.0% in the paper group was comparable to 90.7% in the online group ($P > 0.05$).

Paper questionnaires had a completeness of 54.4%, which was lower compared to the online questionnaires where we reached full completeness ($P < 0.01$). Furthermore, non-responders proved to be younger with a mean age of 52 years compared to a mean age 62 years of responders ($P < 0.05$).

CONCLUSION

The use of a structured online follow-up system increased the response rate. Moreover, online questionnaires are more complete than paper questionnaires.

Key words: Patient reported outcome measures; Elbow; Follow-up; Arthroplasty; Online; Database

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Core tip: Since the last decade, increasing attention is paid on patient reported outcome measures (PROMs) and several online follow-up systems became available to collect PROMs. The purpose of this article was to evaluate the introduction of a structured online follow-up system in order to facilitate analysis of data.

Viveen J, Prkic A, The B, Koenraadt KLM, Eygendaal D. Effect of introducing an online system on the follow-up of elbow arthroplasty. *World J Orthop* 2016; 7(12): 826-831 Available from: URL: <http://www.wjgnet.com/2218-5836/full/v7/i12/826.htm> DOI: <http://dx.doi.org/10.5312/wjo.v7.i12.826>

INTRODUCTION

In 2002, the Swedish Hip Arthroplasty Registry started using patient reported outcome measures (PROMs) on a national scale. In 2011, patients' response rates of up to 90.2% were reported for this registry. Several other national registries have followed their model. In general, most PROMs data are collected from total hip replacement patients. None of the registries included mandatory PROMs on elbow arthroplasty in their dataset.

Nevertheless, PROMs are increasingly used to evaluate the outcome of surgical procedures in patient care and in clinical orthopaedic research projects^[1-4]. These subjective outcomes may be even of more clinical importance than objective outcomes such as range of motion, since patient satisfaction is determined in a complex multifactorial way and does not always correlate well with easy-to-measure objective endpoints.

Unfortunately, in all medical fields researchers and clinicians have to deal with non-responders. This can potentially lead to biased results. And although statistical methods can be employed in an attempt to reduce this effect, it is better to achieve a high response rate to begin with. Therefore encouragement of patients to complete questionnaires is necessary, ideally by using simple and attractive tools.

Especially in patients who received elbow prostheses a structured follow-up is important, as the survival rate of elbow prosthesis is not as high as the rate of hip- and knee arthroplasties^[3]. Despite of several technical improvements of the implants, complications still plague patients with a total elbow arthroplasty. Symptoms of a loosened elbow prosthesis are sometimes unclear and unspecific. This makes recognition at an early stage difficult. Nevertheless, it is essential to have the earliest detection as possible, since loosening of the prosthesis may cause irreversible bone destruction^[5-8].

Since the last decade, several online follow-up systems became available to collect PROMs. Before these systems became available, all questionnaires were sent manual on paper. In the current study we want to report on the advantages and disadvantages of using an online follow-up system. The aim of this study is to evaluate the effect of introducing a structured online follow-up system, as our hypothesis was to increase the response rate in order to obtain a complete follow-up of patients who received elbow arthroplasty.

MATERIALS AND METHODS

This study includes patients who have received surgery at our hospital for four types of elbow arthroplasties; total elbow prosthesis, radial head prosthesis, radio-capitellar prosthesis and revisions of aforementioned prosthesis. Patients were excluded if they were cognitive or physical impaired and therefore unable to complete questionnaires or to visit the outpatient clinic. In total 233 patients met the inclusion criteria and were included for the study.

Demographic data are presented in Table 1. Ninety-nine patients received a total elbow arthroplasty, 14 patients a radiocapitellar prosthesis, 68 patients a radial head prosthesis and 52 patients received revision surgery of aforementioned prosthesis. There were 60 men and 173 woman included. Mean age at surgery was 61 years (SD 13). For further details for each type of elbow arthroplasty see Table 1.

Since June 2015 we have set up an electronic follow-up system (online PROMs, Interactive Studios, Rosmalen, The Netherlands), which allows prospective data gathering using online questionnaires. Online questionnaires are sent automatically at multiple follow-up moments after surgery or manual if patients prefer paper questionnaires. All questionnaires we use are valid in our language. Additional physical examination results can be added manually during reassessment at the outpatient clinic.

Until this introduction all patients received questionnaires on paper. Questionnaires were sent regarding the same follow-up moments, but monitoring was poor since no structured (online) system was available. Patients who received surgery before implementation of the online system are added to the system as well. Questionnaires sent after June 2015 for this group of patients counted as post implementation questionnaires.

When elbow arthroplasty surgery is planned at the

Table 1 Demographic data of all patients divided per type of prosthesis

	Total elbow prosthesis	Radiocapitellar prosthesis	Radial head prosthesis	Revision surgery	Total
Patients (n)	99	14	68	52	233
Sex					
Man	22	7	19	12	60
Woman	77	7	49	40	173
Age (yr)					
Mean (SD)	69 (4)	56 (7)	50 (13)	61 (13)	61 (13)

outpatient clinic and the patient gives written informed consent for use of the data for research, the patient is added to our database. The email addresses are added to the online-system. The database sends a preoperative questionnaire automatically close before surgery. In case of missing email addresses, patients who visit the outpatient clinic for reassessment are asked for their email address, so they can receive the questionnaires regarding the next follow-up moments by email. When the patient prefers paper questionnaires, results are manually added to the electronic system.

This online planning system makes it possible to remind patients and nurse practitioners (NP) to send paper questionnaires and to schedule an appointment as well. Thereby a notification on the dashboard of the online-system shows up if we did not receive response to the questionnaire within two weeks. At our institution we use a protocol for non-responders both in the email and paper group. A second questionnaire is sent after two weeks and if we still do not receive response after four weeks since the first effort, the NP calls the patient to remind them. If patients do not want to participate anymore they can be deactivated in the system.

We intend to achieve a full preoperative PROMs data capture rate. Because we collect PROMs in the trauma department too, we are used to complete the questionnaires retrospectively with maximum scores in baseline in case of acute trauma. The pre-operative questionnaires serve as a baseline measurement. The post-operative follow-up protocol consists of a visit to the outpatient clinic after one, three, five, seven and ten years including questionnaires. On every occasion, the surgeon or the NP sees the patient and a plain X-ray is made.

Since the introduction the pre- and postoperative patients' part of the questionnaires are slightly different. The preoperative questionnaire consists of the Euroqol five dimensions (EQ-5D), the Oxford Elbow Score (OES) and the Visual Analogue pain Scales (VAS 0-10) in rest and activity. Thereby the patient is asked if they did have surgery on their elbow before. If the answer is yes, we want to know when the surgery took place and what kind of surgery it was.

The postoperative questionnaires consist of the EQ-5D, the OES and the VAS 0-10 in rest and activity too. In addition, the patient is asked how satisfied they are with the result of the surgery and whether they would recommend the received therapy for their elbow to

colleagues, friends or family members. These questions are answered on a seven-point scale (satisfaction) and ten-point scale (recommendation). Before the introduction the pre- and postoperative questionnaires were the same and consisted of the OES and VAS pain in rest and activity.

Statistical analysis

For every follow-up moment in each group we determined the number of questionnaires sent and the number of questionnaires we received. We have added these numbers for every group. We compared response rates before and after the introduction of the online database in June 2015. After implementation of the online system we compared the online- and the paper questionnaires group on response rate, completeness and mean age. In addition, we compared the mean age of responders and non-responders in both groups after implementation. Differences on outcome parameters before and after the introduction of the online database were compared using T-tests for normally distributed data and Fisher's Exact test for dichotomous data.

RESULTS

Before vs after the introduction of the online database

Before June 2015 we achieved an overall response rate of 49.8% on 616 sent questionnaires. After implementation of the online database 143 questionnaires were sent and the response rate increased to 91.6% ($P < 0.01$) (Figure 1). Using the online system we captured full response rates both in patients who received radiocapitellar prosthesis and patients who received revision surgery. In patients who received a total elbow arthroplasty and those who received a radial head prosthesis response rates of respectively 90.0% and 83.3% were revealed (Table 2).

Paper vs online after the introduction of the online database

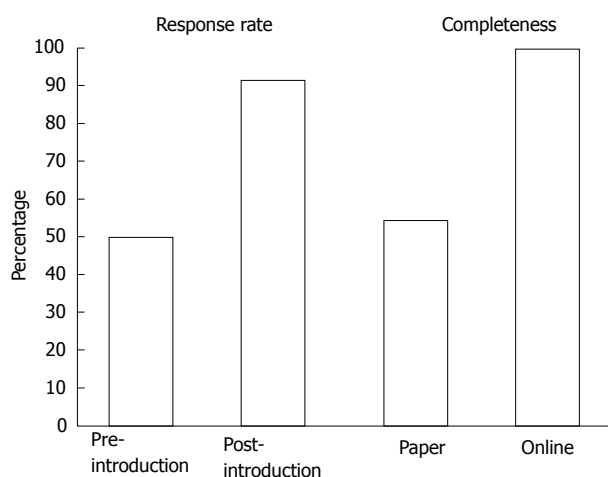
Since the implementation 100 questionnaires were sent on paper and 43 online. We received 92 paper questionnaires and 39 online questionnaires. With a completeness of 54.4% of paper questionnaires, completeness was lower compared to the online questionnaires where we reached full completeness ($P < 0.01$) (Figure 1). The mean age of 63 years (SD 12) of patients who completed the questionnaires on paper was comparable to the mean age

Table 2 Results before *vs* after introduction of the online database

	Total elbow prosthesis	Radiocapitellar prosthesis	Radial head prosthesis	Revision surgery	Total
Questionnaires sent in total					
Before introduction	212	24	245	135	616
After introduction	69	8	36	30	143
Response rate (%)					
Before introduction	57.90%	45.80%	40.40%	54.10%	49.80%
After introduction	89.90%	100%	83.30%	100%	91.60%
Email address known (<i>n</i>)	43	5	26	21	95

Table 3 Paper *vs* online after the introduction of the online database

	Total elbow prosthesis	Radio-capitellar prosthesis	Radial head prosthesis	Revision surgery	Total
Questionnaires sent in total					
Paper	49	5	24	22	100
Online	20	3	12	8	43
Response rate					
Paper	91.90%	100.00%	83.30%	100.00%	92.00%
Online	90.00%	100.00%	83.30%	100.00%	90.70%
Completeness of questionnaires					
Paper	48.90%	40.00%	65.00%	59.10%	54.40%
Online	100.00%	100.00%	100.00%	100.00%	100.00%
Mean age (yr)					
Paper (SD)	68 (8)	58 (8)	50 (14)	63 (10)	63 (12)
Online (SD)	68 (8)	50 (3)	50 (13)	55 (16)	59 (14)
Mean age (yr)					
Responders (SD)	68 (8)	55 (7)	50 (13)	61 (13)	62 (13)
Non-responders (SD)	67 (10)	-	36 (13)	-	52 (19)

**Figure 1** Response rate and completeness.

of 59 years (SD 14) of patients in the online group. If we combine non-responders in the paper and online group, non-responders proved to be younger with a mean age of 52 years (SD 19) compared to a mean age of 62 years (SD 13) of responders ($P < 0.05$). For further details see Table 3.

Online questionnaires

Since the introduction we have collected 95 (41%) email addresses of all elbow arthroplasty patients. The mean age of patients using an email address was 59

years (SD 14). The mean completion time of online questionnaires was 5.2 min (SD 3). Completion times of paper questionnaires were not available.

DISCUSSION

The results of the current study are promising with an increase in response rate from 49.8% to 91.6%. After implementation good response rates are reported both for paper and online questionnaires. In addition online questionnaires were more complete compared to paper questionnaires and non-responders proved to be younger compared to responders.

Several factors might have increased the response rate after the introduction of the online system. As the dashboard function reminds the NP to send paper questionnaires and to be aware of non-response of sent questionnaires, structure is provided regarding follow-up moments. Furthermore, the effort that was previously put into collecting and storing paper versions, is now put into attempts to collect the data in non-responders, since this information is easily fed back by the system. In addition, the amount of paper questionnaires decreased since the online system sends online questionnaires automatically. Therefore, less printing of questionnaires, enveloping, filing, and manual transfer of data from the paper questionnaire to the database is needed anymore. Hence, all these factors together resulted in a huge increase of response rate up to 91.6%.

We also observed some interesting findings on questionnaires sent after the introduction of the online system. At first, we obtained full completeness of online questionnaires compared to 54.4% completeness of paper questionnaires. This is another benefit of using an online system. Online it is impossible to skip questions, while in paper questionnaires researchers and clinicians have to deal with incomplete questionnaires, which accounts for data loss. Secondly, in the current study non-responders proved to be younger compared to the mean age of responders. This is comparable to the results of two other studies^[5,6]. On the contrary, other studies associated non-response with older age^[2,9-11]. The reason of higher non-response rates in younger patients is unclear, but it could be a result of busy lifestyle and low priority in completing questionnaires. With a mean completion time of 5.2 min we think it is reachable to encourage patients to complete our questionnaires. Moreover, in literature the completion times of paper questionnaires are reported to be significant longer compared to online questionnaires when data entry time is studied^[7].

The last noticeable results of the study was the relatively high mean age of patients in the current study was, which probably leads to fewer email addresses, because this generation is not used to computers and/or email^[9]. Nevertheless, the decision to use this group of patients was conscious since a structured follow-up for patients who received an elbow arthroplasty is important as asymptomatic aseptic loosening may cause irreversible loss of bone stock. Hence, in addition to the increase in response rate by using an online system, the current study revealed: A higher completeness using digital-compared to paper questionnaires, that non-response rate was higher in younger subjects and that online questionnaires only took 5.2 min to complete.

While the response rate increased greatly in questionnaires, no show on follow-up moments at the outpatient clinic is still a problem. Arguments we frequently encounter are high costs of the deductible, not having any complaints of the elbow and dependency of elderly people on relatives for driving. Unfortunately, at the moment there is no scientific evidence on the number of reassessments we need to perform to be sufficient in follow-up. However, if patients do not want to be reassessed at the outpatient clinic, questionnaires can be sent anyway. For future research, with a prolonged follow-up, patterns in PROMs outcomes of elbow arthroplasties may be distinguished, so deterrence may be detected earlier and attacked more effectively by, for example, revision surgery. Therefore, with more data, we hope to be able to predict the clinical course per patient and to provide still better patient care.

The current study has two limitations. At first the time since the introduction of the online system in June 2015 is short. Although time is short, we can already report promising results. Thereby in the future we predict more patients to have an email address. We

are convinced this transition from a less online oriented population to a more online oriented population will ensure even better results using an online follow-up system. Further studies are required to evaluate the results on long-term. In addition, the response rate after the implementation of the system could be biased by the effort that was put into collecting questionnaires. Although, this effort could have been put into collecting, since structure was given to follow-up by the dashboard function of the system.

In conclusion, the results of the current study are promising. A structured online system could increase both the response rate and completeness of questionnaires. Further studies are required to report on the mid- and long-term results of the introduction of a structured online follow-up system.

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COMMENTS

Background

Since the last decade, several online follow-up systems became available to collect patient reported outcome measures (PROMs). Before these systems became available, all questionnaires were sent manual on paper. In the current study the authors want to report on the advantages and disadvantages of using an online follow-up system.

Research frontiers

In 2002, the Swedish Hip Arthroplasty Registry started using PROMs on a national scale. Since good response rates were reported in 2011, several other national registries have followed their model. However no literature is available on the effect of introducing an online database. This is the first study reporting on the increased response rate using an online follow-up system.

Innovations and breakthroughs

In the recent years several online databases became available to collect PROMs. The present study represents the effect of introducing an online database on the follow-up of patients who received elbow arthroplasty. The major findings of the study were both an increased response rate and completeness of questionnaires using an online database.

Applications

The data in this study suggested that an online follow-up system could both increase the response rate and completeness of questionnaires. According to the findings of the current study, they can recommend the use of an online database in order to improve the follow-up of patients who have received (elbow) arthroplasty.

Terminology

PROMs are patient reported outcome measures, which are frequently used for follow-up of patients who have received surgery.

Peer-review

It is a well presented, interesting early stage pilot study. Available papers concerning the use of online follow-up databases are rare or non-existing. However, it is about a relevant topic in orthopaedic surgery.

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Observational Study

Surgical apgar score predicts early complication in transfemoral amputees: Retrospective study of 170 major amputations

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Abstract

AIM

To assess whether the surgical apgar score (SAS) is a prognostic tool capable of identifying patients at risk of major complications following lower extremity amputations surgery.

METHODS

This was a single-center, retrospective observational cohort study conducted between January 2013 and April 2015. All patients who had either a primary transtibial amputation (TTA) or transfemoral amputation (TFA) conducted at our institution during the study period were assessed for inclusion. All TTA patients underwent a standardized one-stage operative procedure (ad modum Persson amputation) performed approximately 10 cm below the knee joint. All TTA procedures were performed

with sagittal flaps. TFA procedures were performed in one stage with amputation approximately 10 cm above the knee joint, performed with anterior/posterior flaps. Trained residents or senior consultants performed the surgical procedures. The SAS is based on intraoperative heart rate, blood pressure and blood loss. Intraoperative parameters of interest were collected by revising electronic health records. The first author of this study calculated the SAS. Data regarding major complications were not revealed to the author until after the calculation of SAS. The SAS results were arranged into four groups (SAS 0-4, SAS 5-6, SAS 7-8 and SAS 9-10). The cohort was then divided into two groups representing low-risk ($SAS \geq 7$) and high-risk patients ($SAS < 7$) using a previously established threshold. The outcome of interest was the occurrence of major complications and death within 30-d of surgery.

RESULTS

A logistic regression model with SAS 9-10 as a reference showed a significant linear association between lower SAS and more postoperative complications [all patients: OR = 2.00 (1.33-3.03), $P = 0.001$]. This effect was pronounced for TFA [OR = 2.61 (1.52-4.47), $P < 0.001$]. A significant increase was observed for the high-risk group compared to the low-risk group for all patients [OR = 2.80 (1.40-5.61), $P = 0.004$] and for the TFA sub-group [OR = 3.82 (1.5-9.42), $P = 0.004$]. The AUC from the models were estimated as follows: All patients = [0.648 (0.562-0.733), $P = 0.001$], for TFA patients = [0.710 (0.606-0.813), $P < 0.001$] and for TTA patients = [0.472 (0.383-0.672), $P = 0.528$]. This indicates moderate discriminatory power of the SAS in predicting postoperative complications among TFA patients.

CONCLUSION

SAS provides information regarding the potential development of complications following TFA. The SAS is especially useful when patients are divided into high- and low-risk groups.

Key words: Surgical apgar score; Mortality; Transfemoral amputation; Post-operative complication; Lower extremity amputation

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Core tip: This study presents new knowledge regarding the use of Surgical Apgar Score (SAS) in dysvascular lower extremity amputations (LEA) surgery. There is a significant increase in complications with a low SAS after LEA surgery. This is even more pronounced when the transfemoral amputation (TFA) sub-group is analyzed separately. Thus, for a TFA patient with a $SAS < 7$, the odds of a major complication or death is four times greater than for a patient with a $SAS \geq 7$. ROC analysis confirms the discriminatory power of the SAS approach among the TFA patients. However, the SAS model proved to be of no prognostic value in the transtibial amputation group.

Wied C, Foss NB, Kristensen MT, Holm G, Kallemose T, Troelsen A. Surgical apgar score predicts early complication in transfemoral amputees: Retrospective study of 170 major amputations. *World J Orthop* 2016; 7(12): 832-838 Available from: URL: <http://www.wjgnet.com/2218-5836/full/v7/i12/832.htm> DOI: <http://dx.doi.org/10.5312/wjo.v7.i12.832>

INTRODUCTION

The surgical apgar score (SAS) has a strong correlation with the occurrence of major complications or death within 30-d following general and vascular surgery^[1-4]. The scoring system is based on three intraoperative variables: Lowest mean arterial pressure, lowest heart rate and estimated intraoperative blood loss. The score seems to be irrelevant to several orthopedic sub-specialties^[5,6]. However, there is a lack of knowledge regarding the utility of the score in the field of lower extremity amputations (LEA). An increasing number of high-risk patients are undergoing amputations^[7]. They diverge from standard orthopedic patients due to their high age and many comorbidities^[8]. Studies show that their associated 30-d postoperative mortality is up to 30%^[9], a rate unmatched in the orthopedic specialty. Our aim with this study is to assess whether the SAS is a prognostic tool capable of identifying patients at risk of major complications following LEA surgery.

MATERIALS AND METHODS

This was a single-center, retrospective observational cohort study conducted between January 2013 and April 2015. All patients who had either a primary transtibial amputation (TTA) or transfemoral amputation (TFA) at our institution during the study period were assessed for inclusion. Exclusion criteria included: Re-amputations, a combination of amputation and removal of intramedullary nails and patients with incomplete data registrations. Patients were identified through our local operation database. All patients eligible for inclusion were included. The decision regarding the amputation level was made by senior consultants taking into account skin perfusion pressure measurement results and patient general condition. All TTA patients underwent a standardized one-stage operative procedure (admodum Persson amputation)^[10] performed approximately 10 cm below the knee joint. All TTA procedures were performed with sagittal flaps. The TFA procedures were performed in one stage approximately 10 cm above the knee joint, performed with anterior/posterior flaps. Trained residents or senior consultants performed the surgical procedures. The tourniquet, when used, was inflated around the femur to a pressure of 100 mmHg above systolic blood pressure^[11]. Standardized care was provided for all patients including standards for fluid replacement and thromboprophylaxis. Initial postoperative rehabilitation programs were initiated

Table 1 Calculation of surgical apgar score

	0 point	1 point	2 points	3 points	4 points
Estimated blood loss, mL	> 1000	601 - 1000	101-600	< 100	-
Lowest mean arterial pressure, mmHg	< 40	40-54	55-69	> 70	-
Lowest heart rate, beats per min	> 85	76-85	66-75	56-65	< 55

on the first postoperative day if patients could cooperate.

Intraoperative parameters of interest were collected by reviewing electronic health records. The first author of this study calculated the SAS by following the algorithm described in Table 1. Data regarding major complications were not revealed to the author until after the calculation of SAS. SAS results were arranged into four groups (SAS 0-4, SAS 5-6, SAS 7-8 and SAS 9-10) as proposed by Gawande *et al.*^[2]. The cohort was then divided into two groups representing low-risk (SAS ≥ 7) and high-risk (SAS < 7) patients using a previously established threshold^[5,6,12]. The outcome of interest was the occurrence of major complications and death within 30-d of surgery. The definitions of major complications were in accordance with Gawande *et al.*^[2] from their original study of the field^[2]. This included the following: Bleeding requiring ≥ 4 units of red cell transfusion within 3 d following the operation, acute renal failure (postoperative creatinine > 200 $\mu\text{mol/L}$), acute myocardial infarction, X-ray verified pneumonia, pulmonary embolism, stroke, sepsis and death. The data collectors within this group were cautious not to register preexisting diseases as postoperative complications. The findings were double-checked by two independent researchers to avoid over-registration of complications. The outcomes were omitted compared to Gawande's original work due either to their rare occurrence in amputation surgery or lack of registration in the electronic charts. The study was approved by the local ethics committee and registered at the regional data protection agency (04.12.2012) (j. no. 01975 HVH-2012-053).

Continuous data are presented as median values with interquartile ranges (IQR) or mean values with standard deviations (SD). Comparison between TTA and TFA patients was performed using the t-test or the Wilcoxon rank sum test. Categorical data are presented as numbers with percentages. Comparison between TTA and TFA patients was performed using the chi-squared test or Fisher's exact test. Logistic regression models analyzed the relationship between complications and SAS. Odds ratios (OR) were estimated both for each group level with SAS 9-10 as a reference level and as an average change between levels in the SAS groups to estimate a linear effect across the groups. Both models were analyzed for all patients as one group and were stratified for TFA and TTA procedures.

The discriminatory accuracy of the SAS was evaluated by ROC analysis. The results were expressed

as area under the curve (AUC) and related 95%CI. Logistic regression for complications was made based on the high- and low-risk grouping to evaluate the level of increased risk based on the threshold. This was performed both for all patients as one group and on a stratified basis for the TFA and the TTA procedures. The fits of the logistic regression models were evaluated using the Hosmer-Lemeshow goodness-of-fit test. A p-value of 0.05 was considered statistically significant. All analyses were performed by a biostatistician working with R 3.2.0 (R Foundation for Statistical Computing, Vienna, Austria).

RESULTS

One hundred seventy out of 228 consecutive patients who underwent LEA surgery during the two-year study period were included in the final analysis. In total, 58 patients were excluded due to re-amputation ($n = 37$) or missing data ($n = 21$). The preoperative characteristics are shown in Table 2 and the intraoperative characteristics in Table 3. The overall incidence of major complications and deaths is shown in Table 4 and in relation to the SAS groups in Table 5, which also includes the results from the regression model comparing the individual groups to the reference group. There was a significant linear association demonstrated between SAS group level [OR = 2.00 (1.33-3.03), $P = 0.001$] and complications or death, Table 5. This effect was statistically significant for the TFA group [OR = 2.61 (1.52-4.47), $P < 0.001$] but not for the TTA sub-group [OR = 1.12 (CI: 0.55-2.29), $P = 0.76$]. A significant increase in complications or deaths was observed in the high-risk group (SAS < 7) compared to the low-risk group (SAS ≥ 7) [OR = 2.80 (1.40-5.61), $P = 0.004$]. Corresponding results when the TFA and TTA groups were analyzed separately were [TFA: OR = 3.81 (1.5-9.42), $P = 0.004$], [TTA: OR = 1.69 (0.56-5.12), $P = 0.35$]. The results of the Hosmer-Lemeshow Goodness-of-fit test for the four models all had P -values close to 1, supporting the model assumptions. The results of ROC analysis are shown in Figure 1. The cut-off point of SAS ≥ 7 was found to be optimal when compared to all other cut-off points based on the specificity and sensitivity as evaluated by the Youden index. The AUC from the models were estimated as follows: All patients = 0.648 (0.562-0.733, $P = 0.001$), TFA = 0.710 (0.606-0.813, $P < 0.001$) and TTA = 0.472 (0.383-0.672, $P = 0.528$).

DISCUSSION

This study presents new knowledge regarding the use of the SAS in dysvascular LEA surgery. There was a statistically significant increase in complications with a low SAS after LEA surgery. This is even more pronounced when the TFA sub-group is analyzed separately. Thus, for a TFA patient with a SAS < 7, the odds of a major complication or death are four times larger than for a patient with a SAS ≥ 7 . ROC analysis confirms the discriminatory power of the SAS approach

Table 2 Pre-operative characteristics

	All patients (<i>n</i> = 170)	TTA patients (<i>n</i> = 70)	TFA patients (<i>n</i> = 100)	<i>P</i> -value
Sex (women/men)	74/96	26/44	48/52	0.21
%	(44/56)	(37/63)	(48/52)	
Age, yrs	74 (12)	72 (12)	76 (12)	0.01
Body mass index	24.5 (6.5)	24.9 (5.3)	24.2 (7.2)	0.47
Cause for amputation				
Diabetes/arteriosclerosis/other	58/105/7	30/36/4	28/69/3	0.03
%	(34/62/4)	(43/51/6)	(28/69/3)	
ASA-groups, 1-2/3-4	25/142	16/54	9/88	0.03
%	(15/84)	(23/77)	(9/88)	
Dementia	20 (12)	6 (9)	14 (14)	0.34
Cardiovascular disease	46 (27)	22 (31)	24 (24)	0.30
Pulmonary disease	30 (18)	12 (17)	18 (18)	1.00
Cerebral apoplexy	40 (24)	11 (16)	29 (29)	0.07
Kidney disease	45 (26)	17 (24)	28 (28)	0.72
Diabetes mellitus	72 (42)	37 (53)	35 (35)	0.02
Diagnosed with cancer	14 (8)	6 (9)	8 (8)	1.00
Transfused preoperatively (patients)	34 (20)	10 (14)	24 (24)	0.17
NSAID or acetylsalicylic acid (yes)	82 (48)	38 (54)	44 (44)	0.21
Clopidogrel (yes)	35 (21)	13 (19)	22 (22)	0.70
Preoperative hemoglobin, g/L (SD)	108 (16.1)	108 (16.1)	106 (16.1)	0.48
Preoperative thrombocytes (SD)	354 (151)	366 (157)	345 (146)	0.46

Values are the mean (SD) and number (%), ASA: American Society of anesthesiologists; TTA: Transtibial amputation; TFA: Transfemoral amputation.

Table 3 Intraoperative characteristics

	All patients (<i>n</i> = 170)	TTA patients (<i>n</i> = 70)	TFA patients (<i>n</i> = 100)	<i>P</i> -value
Bilateral amputation procedure	9 (5)	4 (6)	5 (5)	
Rank of surgeon, resident/consultant %	116/54	45/25	71/29	0.40
	(68/32)	(64/36)	(71/29)	
Duration of surgery, minutes (SD)	81 (23)	85 (26)	79 (20)	0.06
Neuraxial/general anesthesia	121/49	56/14	65/35	0.04
%	(71/29)	(80/20)	(65/35)	
Vasopressor agents during surgery (yes)	106	37 (52)	69 (69)	0.037
Tourniquet use (yes)	49 (29)	35 (50)	11 (11)	< 0.001
Initial heart rate (beats/min) (SD)	85 (16)	84 (15)	85 (17)	0.77
Initial blood pressure (mmHg)	Sys: 131 (21)	Sys: 131 (20)	Sys: 131 (21)	0.83
	Dia: 73 (13)	Dia: 74 (13)	Dia: 73 (12)	0.552
Lowest heart rate (beats/min)	72 (SD: 1.2)	70 (SD: 1.9)	74 (SD: 1.5)	0.13
Lowest mean arterial pressure (mmHg)	64 (SD: 0.9)	65 (SD: 1.4)	64 (SD: 1.1)	0.32
Estimated blood loss (mL), median (IQR)	300 (IQR: 125-475)	248 (IQR: 88-408)	400 (IQR: 231-569)	0.15
Perioperative blood lactate (SD)	1.2 (1.2)	1.0 (0.5)	1.3 (1.5)	0.33
Perioperative acid-base balance (SD)	7.39 (0.01)	7.41 (0.05)	7.38 (0.09)	0.05

TTA: Transtibial amputation; TFA: Transfemoral amputation.

Table 4 Surgical APGAR score complications (%)

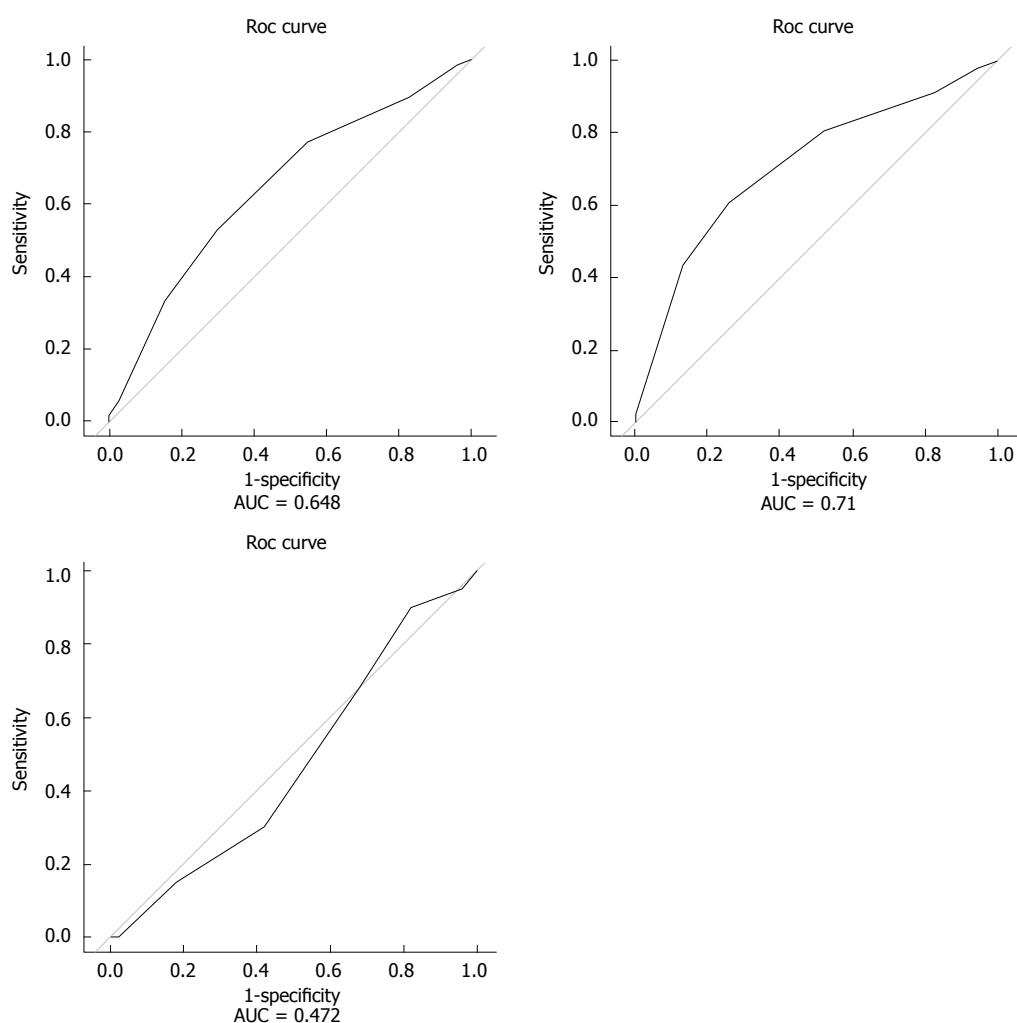
	Total (<i>n</i> = 170)	TTA group (<i>n</i> = 70)	TFA group (<i>n</i> = 100)
Death	30 (18)	9 (13)	21 (21)
Bleeding requiring ≥ 4 units of RBC transfusion within 3 d following operation	28 (16)	7 (10)	21 (21)
Sepsis	12 (7)	4 (6)	8 (8)
Acute myocardial infarction/acute heart failure	7 (4)	0 (0)	7 (7)
Acute renal failure	6 (4)	1 (1)	5 (5)
Pneumonia	18 (11)	5 (7)	13 (13)
Stroke	1 (1)	0 (0)	1 (1)
Pulmonary embolism	0 (0)	0 (0)	0 (0)

TTA: Transtibial amputation; TFA: Transfemoral amputation.

Table 5 Logistic regression analysis of surgical APGAR score groups and complications/death

Score	SAS 9-10	SAS 7-8	SAS 5-6	SAS 0-4
All patients (%)	5 (3)	57 (34)	70 (41)	38 (22)
Complications and deaths within SAS group (%)	1 (20)	14 (25)	29 (41)	22 (58)
Odds Ratio between groups (reference SAS 9-10)	(ref.)	OR = 1.30 CI: 0.13-12.64 P = 0.82	OR = 2.83 CI: 0.30-26.64 P = 0.36	OR = 5.50 CI: 0.56-53.99 P = 0.143
Linearity of the model, OR (95%CI), P-value		2.00 (CI: 1.33-3.03) P = 0.001		
Transfemoral procedure, n = 100 (%)	4 (4)	31 (31)	38 (38)	27 (27)
Complications and deaths within SAS group (%)	1 (25)	8 (26)	17 (45)	20 (74)
Odds ratio between groups (reference SAS 9-10)	(ref.)	OR = 1.04 CI: 0.10-11.52 P = 0.97	OR = 2.43 CI: 0.23-25.51 P = 0.46	OR = 8.57 CI: 0.76-96.52 P = 0.08
Linearity of the model, OR (95%CI), P-value		2.61 (CI: 1.52-4.47) P < 0.001		
Trans tibial procedure, n = 70 (%)	1 (1)	26 (37)	32 (46)	11 (16)
Complications and deaths within SAS group (%)	0 (0)	6 (23)	12 (38)	2 (18)
Odds Ratio between groups (reference SAS 7-8)	¹	(ref.)	OR = 0.74 CI: 0.13-4.41 P = 0.74	OR = 2.00 CI: 0.63-6.38 P = 0.24
Linearity of the model, OR (95%CI), P-value		1.12 (CI: 0.55-2.29) P = 0.76		

¹Only one TTA in the SAS 9-10 group. SAS 7-8 is therefore used as the reference group. TTA: Transtibial amputation; TFA: Transfemoral amputation.

**Figure 1** Receiver operating characteristic curve for all patients, transfemoral amputation patients, transtibial amputation patients.

among the TFA patients. However, the SAS model proved to be of no prognostic value in the TTA group.

The population requiring LEA is old, fragile and has several co-morbidities, as clearly shown by this study. When compared to the cohort of general and vascular surgery patients^[1,2,12], our average LEA patient is approximately 20 years older and 84% of patients have an ASA score classifications > 2 (possibly one of the highest reported in any orthopedic cohort), which compares to 34% in the study by Gawande *et al.*^[2]. The mortality rate in the present study is also high, with 18% of patients dead within the first 30 postoperative days. This clearly shows how vulnerable LEA patients are and how an easily applied risk stratification tool would be of great value for individualizing postoperative monitoring and care. Such a score was suggested in 2007^[2,3] and the SAS has proved to be useful in several surgical specialties including gynecologic, urology and colorectal surgery^[1,2,13,14].

However, only recently has the predictive value of the SAS on patients undergoing orthopedic surgery been evaluated^[6,15]. Furthermore, only in spine surgery was the SAS model found to be of value^[6]. Despite these findings, it was expected that the SAS could prove useful in the LEA cohort due to the similarities in demographics shared with patients undergoing general and vascular surgery^[1]. To some extent, the results from this study did confirm our expectations, although it was surprising to see how poorly the score predicted outcome for the TTA patients. In the TFA group, however, approximately four times as many patients with complications were SAS high-risk patients compared to low-risk and the specificity as analyzed by the ROC model confirmed the difference. It is reasonable to conclude that the SAS model has discriminatory power on the TFA sub-group. The TFA group is significantly different from the TTA group in several important variables regarding the SAS model, which increases the risk of postoperative complications or death. For example, TFA patients are significantly older, more often classified as ASA 3-4, more frequently amputated due to severe arteriosclerosis, have procedures more frequently performed under general anesthesia and more frequently require the aid of vasopressor agents such as Ephedrine or Phenylephrine to secure a stable mean arterial pressure compared to TTA patients (Tables 1 and 2). These differences signal that the TTA group is in markedly better pre-operative condition and therefore less exposed to postoperative complications. Another important matter regarding the low specificity of TTA analysis could be superior hemostatic control in the TTA group where 50% had a pneumatic tourniquet applied during surgery. This lower intraoperative blood loss significantly^[16] and potentially reduces the risk of intraoperative tachycardia^[17]. Most acute LEA procedures are performed with a TFA approach^[18], which to some extent is backed by the results from registration of the blood lactate and acid-base balance. Perioperative blood lactate is 0.3 higher in the TFA group with a markedly higher standard deviation

pointing out several high outliers. Furthermore, the blood acid-base balance was found to be lower. In the event of an acute amputation, the operating staff would be challenged to maintain intact vital parameters during surgery. A larger drop in mean arterial pressure or a sudden rise in heart rate or intraoperative bleeding would affect the SAS and the outcome.

Limitations of this cohort study include those associated with its retrospective design and the missing data. Since there are many co-morbidities within this study, there is a risk that some post-operative complications could already have been present before amputation. However, the data collectors within this group have been highly aware of this matter. The study was performed on a unique group of patients often considered poor candidates for intensive care treatment with high post-operative morbidity and mortality. Randomized controlled trials and prospective studies can be a dubious task with this population, and we found the retrospective design sufficient to answer our research question of the study. The study provides some evidence of the value of the SAS in the post-operative treatment. However, further prospective studies examining the performance of the SAS seem warranted.

In conclusion, it seems warranted that the SAS approach provides the medical staff with information regarding the potential postoperative course after TFA surgery, especially when the patients are divided into high- and low-risk groups. The scoring system could prove useful in guiding preventive strategies such as optimizing intraoperative blood pressure or heart rate. The previously established threshold of SAS < 7 to define high-risk patients and SAS ≥ 7 to define low-risk patients was confirmed to be the optimal cut-point by ROC analysis within this study. The SAS showed no discriminatory power in the TTA sub-group, most likely due to optimized hemostatic control, fewer acute amputations and overall better condition compared to TFA patients.

COMMENTS

Background

There is an increasing number of high-risk elderly and severely comorbid patients scheduled for dysvascular lower extremity amputations. An easily applied risk stratification tool would be of great value for individualizing postoperative monitoring and care. The surgical apgar score (SAS, 0-10 points) has a strong correlation with the occurrence of major complications or death within 30-d following general and vascular surgery. However, a similar correlation has not been demonstrated in general orthopedic surgery. The primary aim of this study is to assess whether the SAS is a prognostic tool capable of identifying patients vulnerable to major complications (including death) following lower extremity amputations surgery.

Research frontiers

The authors have reported the first series in the literature of patients with lower extremity amputation who were evaluated with the SAS. The scoring system could prove useful in guiding preventive strategies such as optimizing intraoperative blood pressure or heart rate.

Innovations and breakthroughs

When divided into four groups (SAS: 0-4, 5-6, 7-8 and 9-10), a logistic

regression model shows a significant linear association between decreasing SAS and increasing postoperative complications in transfemoral amputation patients. This effect is even more pronounced when the patients were compared in high-risk and low-risk SAS groups.

Applications

The SAS should be considered during postoperative care of transfemoral amputation patients.

Peer-review

The paper is well-written and this observational study is informative for readers.

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Recurrent missense mutation of *GDF5* (*p.R438L*) causes proximal symphalangism in a British family

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Informed consent statement: Consent was taken from the patients for their history analysis and genetic testing.

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Abstract

Proximal symphalangism (SYM1B) (OMIM 615298) is an autosomal dominant developmental disorder affecting joint fusion. It is characterized by variable fusions of the proximal interphalangeal joints of the hands, typically of the ring and little finger, with the thumb typically being spared. SYM1 is frequently associated with coalition of tarsal bones and conductive hearing loss. Molecular studies have identified two possible genetic aetiologies for this syndrome, *NOG* and *GDF5*. We herein present a British caucasian family with SYM1B caused by a mutation of the *GDF5* gene. A mother and her three children presented to the orthopaedic outpatient department predominantly for feet related problems. All patients had multiple tarsal coalitions and hand involvement in the form of either brachydactyly or symphalangism of the proximal and middle phalanx of the little fingers. Genetic testing in the eldest child and his mother identified a heterozygous missense mutation in *GDF5* c.1313G>T (*p.R438L*), thereby establishing SYM1B as the cause of the orthopaedic problems in this family. There were no mutations identified in the *NOG* gene. This report highlights the importance of thorough history taking, including a three generation family history, and detailed clinical examination of children with fixed planovalgus feet and other family members to detect rare skeletal dysplasia conditions causing pain and deformity, and provides details of the spectrum of problems associated with SYM1B.

Key words: Proximal symphalangism; *GDF5*

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Core tip: This report highlights the importance of thorough history taking, including a three generation family history, and detailed clinical examination of children with fixed planovalgus feet and other family members to detect rare skeletal dysplasia conditions causing pain and deformity, and provides details of the spectrum of problems associated with SYM1B.

Leonidou A, Irving M, Holden S, Katchburian M. Recurrent missense mutation of *GDF5* (p.R438L) causes proximal symphalangism in a British family. *World J Orthop* 2016; 7(12): 839-842 Available from: URL: <http://www.wjgnet.com/2218-5836/full/v7/i12/839.htm> DOI: <http://dx.doi.org/10.5312/wjo.v7.i12.839>

INTRODUCTION

Proximal symphalangism (SYM1) is an autosomal dominant developmental disorder affecting joint formation^[1]. It is characterized by variable fusions of the proximal interphalangeal joints of the hands, typically of the ring and little finger, with the thumb typically being spared. SYM1 is frequently associated with coalition both of the tarsal bones and the ear ossicles, resulting in conductive hearing loss^[1]. This rare association was first reported by Cushing in 1916 and subsequently by Drinkwater in 1917 for the Brown and Talbot families respectively^[2,3].

Genetic mapping studies have shown that SYM1 shows genetic heterogeneity, with mutations in *NOG*, encoding *NOG*, and *GDF5*, encoding Growth Differentiation Factor 5 (1), causing SYM1A (OMIM 185800) and SYM1B respectively^[4-7].

Here we present a British caucasian family with SYM1B caused by an autosomal dominantly transmitted heterozygous mutation in *GDF5*.

CASE REPORT

A mother presented to our orthopaedic outpatient department with her three children, predominantly for feet related problems. During clinical examination inability to flex the proximal interphalangeal joint of the little fingers was observed and subsequent radiographs revealed symphalangism of the proximal and the middle phalanges. All children and the mother had multiple tarsal coalitions and hand involvement in the form of either brachydactyly or fusion of the proximal interphalangeal joint of the little fingers. The clinical findings of the affected members are demonstrated in Table 1. The father was not clinically affected. The pain and foot deformity significantly impacted upon activities of daily living, limiting exercise capacity and restricting hand use for writing, for example. Hearing was tested and no loss was observed in this family.

DNA sequencing of the eldest son identified a heterozygous missense mutation c.1313G>T in *GDF5*, which

causes the amino acid substitution arginine to leucine at codon 438 (R438L) which predicted to alter *GDF5* signalling and therefore disrupt its function. This was demonstrated to be dominantly transmitted from his affected mother. There were no mutations identified in the *NOG* gene.

DISCUSSION

SYM1 is part of a spectrum of disorders that cause joint fusion. At the most severe end of this spectrum is the multiple synostoses syndrome (SYNS1), which causes severe and widespread joint involvement including the hips, vertebrae and the elbows^[1,8].

Genetic mapping studies have shown that mutations in at least two genes underlie SYM1, *NOG* (SYM1A) and *GDF5* (SYM1B). *NOG* maps to chromosome 17 and encodes the protein Noggin (*NOG*), a bone morphogenic protein (BMP) inhibitor expressed at the sites of joint development^[1]. In the formation of the human skeleton BMPs induce mesenchymal cell proliferation and differentiation into chondroblasts, recruit chondrocytes and promote cartilage formation^[1,9]. *NOG* inhibits cartilage development, leading to separation of the cartilage and thus joint formation^[1,9]. *NOG* mutations lead to under expression of *NOG* resulting in high levels of BMPs, cartilage overgrowth and the absence of joint formation, which affects individuals with SYM1^[1,8-11]. *GDF5* encodes Growth and Differentiation factor 5, a protein belonging to the transforming growth factor beta superfamily which also has an important role in joint development. *GDF5* acts in the same way as BMPs that promote chondrocyte differentiation and cartilage formation^[1,4,5]. The *GDF5* gene is on chromosome 20 and has two coding exons. The c.1313G>T (R438L) mutations in *GDF5* has been previously detected in a family with SYM1^[5] and the causative mechanism is thought to be increased biological activity of *GDF5* leading to joint cartilage overgrowth and subsequent fusion^[1,4,5]. *GDF5* mutations associated with SYM1 and the allelic disorder SYNS1 are summarized in Table 2. The p.Arg438Leu substitution is a gain-of-function mutation known to be associated with proximal symphalangism^[4]. Compared to wild type *GDF5*, the resulting protein shows increased biological activity that alters receptor-binding affinity within the TGFB signalling pathway. Overexpression of *GDF5* disrupts normal joint formation and causes proximal symphalangism.

Hand involvement in patients with SYM1 prevents them from making a fist but does not usually cause significant impairment of functionality required for daily activities^[1]. In the reported family the cause for attendance at the orthopaedic outpatient department was pain and stiffness as a result of tarsal coalitions and fixed planovalgus feet, in general, and not primarily due to hand problems, though restriction was impacting upon their ability to write comfortably. The mother had multiple targeted injections and subsequently developed subtalar joint osteoarthritis and underwent subtalar joint fusion (Figure 1). The other family members have been treated conservatively with orthotics and surgery will be

Table 1 Clinical characteristics of the affected member of the reported family

Affected member (current age)	Hand abnormalities	Tarsal coalition	Conductive hearing loss	Other	Orthopaedic treatment required
Patient 1: Mother (40)	Brachydactyly, no symphalangism	Bilateral: Talonavicular coalition, calcaneocuboid coalition, middle and lateral cuneiform coalition	No	Pituitary adenoma – prolactinoma, platelet storage pool disorder	Yes for painful fixed valgus hindfoot, had targeted injections, developed subtalar osteoarthritis, underwent subtalar fusion
Patient 2: Son (18)	Symphalangism, bilateral little fingers proximal interphalangeal joints fusion ¹	Bilateral: Calcaneocuboid fusion, 3 rd metatarsal - lateral cuneiform, Right talonavicular coalition	No		Orthotics only
Patient 3: Son (15)	Brachydactyly, no symphalangism	Bilateral: Calcaneocuboid coalition, and medial cuneiform to third metatarsal coalition	No	Developmental delay, asthma, under investigation for Marfans	Orthotics only
Patient 4: Daughter (11)	Symphalangism, bilateral little fingers proximal interphalangeal joints fusion ¹	Bilateral: Calcaneocuboid and talonavicular coalition, and medial cuneiform to third metatarsal coalition	No	Developmental delay, asthma, mild platelet dysfunction	Orthotics only

¹For the symphalangism, bilateral little fingers proximal interphalangeal joints fusion of patients 2 and 4, please see the Figure 2.

Table 2 Identified mutations of *GDF5* in relation to proximal symphalangism and multiple synostoses syndrome 2

Ref.	Syndrome	Mutation identified	Effect on codon
Wang <i>et al</i> ^[6]	SYM1	1471 G - A	Glutaminic acid to lysine (E491K)
Yang <i>et al</i> ^[7]	SYM1	1118 T - G	Leukine to arginine (L373R)
Seemann <i>et al</i> ^[4]	SYM1	1632 G - T	Arginine to leucine (R438L)
Dawson <i>et al</i> ^[5]	SYNS2	1313 G - T	Arginine to leucine (R438L)

SYM1: Proximal symphalangism; SYNS2: Multiple synostoses syndrome 2.



Figure 1 Radiographs of patient 1 demonstrating multiple tarsal coalitions and right foot radiographs following subtalar joint fusion.

considered at skeletal maturity (Figure 2).

To the authors' knowledge, this is the first report of a British caucasian family with SYM1 associated with a *GDF5* mutation. This case report highlights the importance of taking thorough clinical history and performing detailed clinical examination of patient's

and their affected relatives in establishing a diagnosis in children presenting with fixed planovalgus feet with a view to planning appropriate management. Clinical data from this and other families with SYM1 caused suggest that foot pain is the main medical problem for affected individuals and that hand dysfunction is uncommon



Figure 2 Radiographs of patient 2 (left) and patient 4 (right) demonstrating little fingers symphalangism of proximal a middle phalanx. Patients are unable to flex the proximal interphalangeal joint of their little finger as opposed to the other fingers as demonstrated.

Joint dysfunction can usually be managed by non-operative methods, but may occasionally require surgical intervention.

COMMENTS

Case characteristics

Painful flat feet in a mother and her children.

Clinical diagnosis

Proximal symphalangism.

Laboratory diagnosis

DNA sequencing.

Imaging diagnosis

Radiographs of hands and feet confirming tarsal coalitions and symphalangism.

Treatment

Guided by symptoms and in the form of orthoses, injection therapy and subtalar joint fusion in the mother.

Experiences and lessons

Thorough clinical examination and history taking in order to correctly identify and treat skeletal dysplasias.

Peer-review

This is an interesting case report, highlighting the importance of thorough history

taking. It has great significance to clinical practice, so should be published.

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Glomus tumors of the fingers: Expression of vascular endothelial growth factor

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Abstract

Glomus tumors are uncommon, benign, small neurovascular neoplasms derived from glomus bodies in the reticular dermis. Glomus bodies are found throughout the body to regulate body temperature and skin circulation; however, they are concentrated in the fingers and the sole of the foot. The typical presentation is a solitary nodule in the subungual or periungual area of the distal phalanx. The primary treatment of choice is surgical removal. We investigated expression of vascular endothelial growth factor (VEGF) using immunohistochemistry in glomus tumors of the fingers. All five glomus tumor samples were positive for VEGF expression. VEGF immunoreactivity was largely localized to the cytoplasm of tumor cells, suggesting a contribution of VEGF to the vascularization of glomus tumors.

Key words: Glomus tumors; Fingers; Vascular endothelial growth factor

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Core tip: Glomus tumors are uncommon, benign, small neurovascular neoplasms derived from glomus bodies in the reticular dermis. The role of vascular endothelial growth factor has never been investigated in glomus tumors of the fingers. This case report demonstrated high

vascular endothelial growth factor (VEGF) expression in the glomus tumors of the fingers, suggesting a contribution of VEGF to the vascularization of glomus tumors.

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INTRODUCTION

Glomus tumor is a relatively rare benign perivascular painful neoplasm arising from the neuromyoarterial structure called a glomus body, a specialized arteriovenous anastomosis involved in thermoregulation^[1]. The diagnosis is virtually relied upon the basis of the clinical history and examination^[2]. The classic clinical triad includes paroxysmal shooting pain, localized tenderness, and hypersensitivity to cold. Patients usually present with a painful, exquisitely tender mass beneath the nail that is accompanied by a faint bluish discoloration. Except for the slight change in color of the nail overlying the tumor, the nail may appear normal. The differential diagnosis includes local infection, osteomyelitis, osteoid osteoma, inclusion cyst, and malignancy.

Vascular endothelial growth factor (VEGF) is a crucial stimulator of blood vessel growth and is one of the most potent growth factors of angiogenesis. Angiogenesis is a fundamental process for growth of new blood vessels from preexisting vasculature during fetal development and tissue repair; however, uncontrolled angiogenesis can contribute to a variety of disorders including neoplastic diseases. In the present study, we performed immunohistochemistry to evaluate the expression of VEGF in glomus tumor tissues.

CASE REPORT

Five patients with solitary glomus tumors of the fingers were surgically treated between 2010 and 2014 at the Department of Orthopaedics of King Chulalongkorn Memorial Hospital, Bangkok, Thailand. The histological diagnoses of each tumor were validated by an experienced pathologist.

The paraffin-embedded tissues were cut in 5 μ m thickness and processed for VEGF staining. Sections were deparaffinized and rehydrated in Tris-buffered saline. Endogenous peroxidase activity was blocked with 0.3% H₂O₂ for 10 min. For antigen retrieval, tissue sections were microwave heated in 10 mmol/L citrate buffer for 5 min. Nonspecific binding was blocked for 20 min with 3% normal horse serum (DAKO, Glostrup, Denmark), followed by incubation with primary antibody (rabbit polyclonal anti-human VEGF antibody 1:100; Santa Cruz Biotech, Santa Cruz, United States) in Tris-buffered

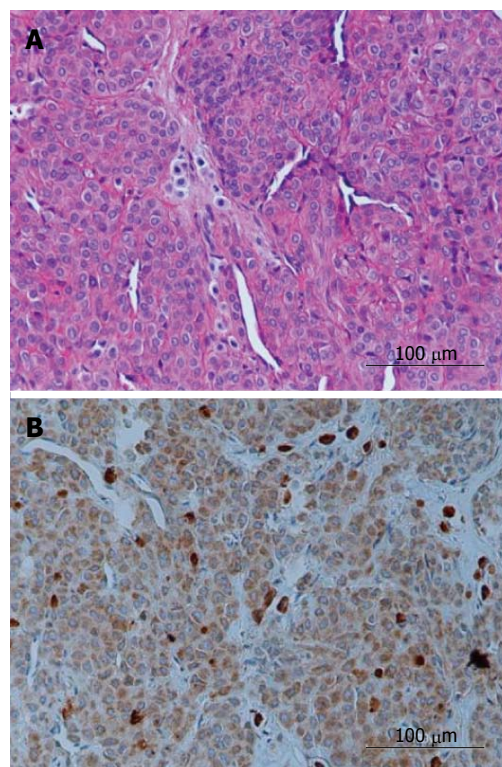


Figure 1 Histopathologic examination. A: The tumor is composed of multiple vascular channels lined by endothelial cells and aggregates of round cells with darkly staining round to ovoid nuclei and eosinophilic cytoplasm (hematoxylin and eosin stain, $\times 400$); B: The tumour cells are strongly positive for vascular endothelial growth factor (VEGF stain, $\times 400$). Strong cytoplasmic staining for VEGF in the tumor parenchyma. VEGF: Vascular endothelial growth factor.

saline containing 2% rabbit serum and 1% bovine serum albumin for two hours. Tissues were incubated with the same buffer without the antibody to serve as negative controls. Sections were subsequently stained with biotinylated goat anti-rabbit immunoglobulins (1:400; DAKO) and streptavidin/horseradish peroxidase complex (1:400; DAKO) and incubated at room temperature for 30 min. Reaction products were visualized using diaminobenzidine (Sigma, St. Louis, United States) as the chromogen. The sections were subsequently counterstained with Mayer's hematoxylin and mounted onto microscope slides using a permanent medium.

All five glomus tumor specimens derived from five female patients (age range, thirty-five to forty-eight years) and were located on the fingers. The entire tumors were completely excised surgically.

Representative sections evaluated for histological and VEGF immunostaining are demonstrated in Figure 1, and the grading is present in Table 1. Histopathologic examination demonstrated a well-defined mass to be a glomus tumor, which comprised uniform round or slightly polygonal cells with sharp cellular borders and eosinophilic cytoplasm. The cells are embedded in myxoid and collagenous stroma as solid sheets or thin layer around vascular spaces (Figure 1A). Positive VEGF staining was found in all specimens, with strong VEGF cytoplasmic staining in all five specimens (Figure 1B).

Table 1 Clinical data and scores of vascular endothelial growth factor immunohistological staining

Case	Age (years)	Clinical presentation	Finger	VEGF staining
1	35	Severe pain, point tenderness and bluish discoloration in subungual region	Left thumb	3+
2	38	Episodic pain, cold sensitivity and severe tenderness at the tip of digit	Left middle	2+
3	40	Excruciating pain, numbness and extreme tenderness at the tip of digit	Left small	4+
4	45	Severe pain, worse at night and bluish lesion in subungual base	Right index	2+
5	48	Paroxysmal pain, tingling, paresthesia and point tenderness at the tip of digit	Right ring	3+

VEGF: Vascular endothelial growth factor.

Immunohistochemical examination revealed that the tumor cells are strongly positive for vascular endothelial growth factor. VEGF immunoreactivity was largely localized to the cytoplasm of tumor cells (Figure 1B), suggesting a contribution of VEGF to the vascularization of glomus tumors. The patients' symptoms including severe pain, focal tenderness, and cold hypersensitivity greatly improved and resolved entirely within several weeks following surgical removal. At present, all patients have no pain at rest or at night two years after surgery.

DISCUSSION

Glomus tumors are uncommon, small, painful, and commonly benign hamartomas arising from the arterial end of the glomus body in the reticular dermis. The typical presentation is a solitary nodule in the subungual or periungual area of the distal phalanx. Patients typically present with a triad of symptoms including excruciating paroxysmal pain (worse at night), temperature sensitivity, and severe point tenderness. Direct pressure on the tumor with the head of a straight pin or the tip of a pencil leads to excruciating pain, while pressure applied slightly to one side of it elicits no pain. Immersing the involved hand or digit in cold water or ice cube also result in discomfort.

Glomus tumors usually are less than 1 cm in diameter, often being only a few millimeters in diameter, and may be visible through the overlying tissues as a blue or purple discoloration. They occur more often in the hand nail in 25%-65% of patients. Seventy-five percent are subungual, however, these may pose a difficult diagnosing these tumors^[3]. Although frequently found in the dermis, glomus tumors may occur in deep soft tissue or visceral sites throughout the body including lung, gastrointestinal, and liver^[4,5]. Hypertrophy of a glomus body, an innervated, coiled, arteriovenous dermal shunt that normally controls skin temperature, is evident this tumor. Glomus cells are specialized perivascular muscle cells that are round or oval and have a dense, granular cytoplasm. Nonmyelinated nerve fibers, which are intermixed with thick-walled capillaries, are responsible for the lancinating pain. The mechanism of pain in glomus tumors has not been clearly elucidated, but it may be associated with contraction of myofilaments in response to temperature changes, resulting in an increase in intracapsular pressure^[6]. Although the

precise pathogenesis of glomus tumors is unknown, it is postulated that they either represent hamartomas or reactive hypertrophy secondary to trauma^[6].

Glomangioma and glomangiomyoma are classic variants of the common feature of glomus tumors. The typical histological appearances of the glomus tumors comprise angiocentric uniform sheets of cells with oval nuclei, forming a perivascular collar around vessels. The three different tumor variants are differentiated by their histological characteristics. The common or solid form includes lobules, strands, and broad sheets of rounded, uniform glomus cells with indistinct capillaries in the walls of surrounding large blood vessels. Whereas glomangiomas exhibit prominent vascular structures with dilated veins surrounded by clusters of glomus cells, and glomangiomyomas consisted of prominent vascular and elongated, spindle smooth muscle cells^[7]. In malignant forms, glomangiosarcoma, pleomorphic tumor cells with marked nuclear atypia and frequent mitotic figures are found in variable numbers^[8]. Angiogenesis has been implicated in the progression from benign to malignant tumors. The contribution of angiogenesis and VEGF expression in glomus tumors as yet has not been completely elucidated. It is not yet clear whether there is any difference in VEGF expression between benign and malignant forms of glomus tumors.

In this study, VEGF was detectable in all specimens of glomus tumors. VEGF has been known as a potent angiogenic factor involved in neovascularization. It has been shown that VEGF are expressed in paragangliomas and may contribute to the extreme vascularity of these tumors^[9]. Hence, the elevated VEGF expression in the glomus tumor might play a paracrine role in the angiogenesis that vascularizes around the tumors by capillary sprouting from the adjacent vascular network. Angiogenesis may result in the reconstruction of nutrition for the expanding tumor and could enable further proliferation. However, prospective longitudinal studies with larger sample size are warranted to define the precise role of VEGF in glomus tumors.

These tumors can be removed with the patient under local anesthesia and should be accurately localized by marking the lesion just before operation. Removal of the portion of the nail plate over the area of tenderness and excision of the matrix that appears involved along with a margin of normal-appearing matrix is the treatment of choice. Magnification and high-intensity lighting

facilitate excision of these periungual and subungual masses. Meticulous and complete excision of the usually well-encapsulated lesions is curative, although rates of recurrence 4%-15% have been reported^[10]. Recurrence is attributed to incomplete excision, undetected multiple tumors, or development of a new tumor.

In conclusion, increased VEGF expression was observed in glomus tumors. VEGF could contribute to the process of promoting tumor angiogenesis and might be important in the pathogenesis of glomus tumors.

COMMENTS

Case characteristics

Five female patients (age range, thirty-five to forty-eight years) presented with exquisite pain, point tenderness, cold hypersensitivity on their fingers.

Clinical diagnosis

Severe paroxysmal pain, temperature sensitivity, and severe point tenderness in the finger tips, particularly in the subungual region.

Differential diagnosis

Local infection, osteomyelitis, osteoid osteoma, inclusion cyst, glomus tumor, glomangioma, glomangiomyoma, and malignancy.

Laboratory diagnosis

All labs were within normal limits.

Pathological diagnosis

Glomus tumor.

Treatment

Complete surgical excision of lesion.

Related reports

Glomus tumor is an uncommon benign perivascular painful neoplasm arising from the neuromyoarterial structure of the glomus body. Although frequently found in the dermis, glomus tumors may occur in deep soft tissue or visceral sites throughout the body.

Term explanation

Glomangioma and glomangiomyoma are classic variants of the common feature of glomus tumors. Glomangiomas exhibit prominent vascular structures with dilated veins surrounded by clusters of glomus cells, and glomangiomyomas consisted of prominent vascular and elongated, spindle smooth muscle cells.

Experiences and lessons

The entity of glomus tumor should not be confused with hemangioma or paraganglioma. When making a differential diagnosis of unexplained paroxysmal shooting pain, sensitivity to cold, and localized tenderness in the finger, glomus tumour must be taken into consideration. The standard treatment of choice is surgical removal. Recurrence of glomus tumor generally resulted from incomplete excision.

Peer-review

This is a report of vascular endothelial growth factor in glomus tumors of the fingers. The topic of this manuscript is interesting and it deserves serious consideration for publication. The high VEGF expression in the glomus tumors of the fingers has not been reported in other literatures.

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