Spike the PCHA! Overuse injury of the Posterior Circumflex Humeral Artery in elite volleyball
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CHAPTER 1

General introduction
Research questions
Outline of the thesis
1.1 - GENERAL INTRODUCTION

Volleyball was introduced more than 100 years ago by the American physical educator William Morgan1, and today enjoys one of the highest participation rates of any sport in the world. With the great success of the world competitions organised by the Fédération Internationale de Volleyball (FIVB), the number of active players in the world has grown exponentially recent decades (Figure 1).110 By most estimates, volleyball ranks second only to football (soccer) in terms of global popularity.100 One of the most appealing aspects of the sport is that it can be played indoors and outdoors, by the young and the old, by males and females, and by both the able-bodied and those with physical impairments.100 Volleyball is furthermore unique among team sports in that it has evolved into two distinct Olympic disciplines: a two-person per side outdoor game typically played on sand (beach volleyball), and an indoor version featuring six players on each team (indoor volleyball).

As with all sports, those who enjoy either of the two volleyball disciplines assume a certain risk of injury the moment they step on to the court.100 Volleyball, whether played indoors or on the beach, is a relatively safe sport in terms of overall injury rate8,9,122.

Figure 1110 Players (in millions) active in the world after volleyball introduction (1885). (a) In 1916 the Spalding Volleyball Guide, written by Robert C. Cubbon, estimated 200,000 players in the United States alone. (b) In 1990 a figure of 150 million players was estimated (170 countries). (c) In 1994 FIVB claimed 200 million players.1 (d) In 2004 volleyball had more than 500 million registered players worldwide (data FIVB, 2004). (e) Today there are 800 million players worldwide who play volleyball at least once a week (46 million of them in the United States).110
particularly compared to contact sports such as football (soccer) and field hockey. For instance, research conducted during the Athens 2004 Olympics confirms that volleyball enjoyed the lowest injury rate of any contested team sport. Published data suggest that the injury pattern is similar for men and women, and that volleyball athletes appear to be at greatest risk of acute injuries of the ankle (inversion sprains) and overuse injuries of the knee (predominantly patellar tendinopathy) and the shoulder (Figure 2). The most common overuse-related injuries of the shoulder girdle in volleyball players involve impingement, subscapular neuropathy, and functional instability.

Vascular injuries of the shoulder girdle in volleyball
Vascular injuries of the shoulder girdle in volleyball are rare and infrequently addressed in the sports medicine literature. Volleyball players are at risk of vascular overuse injuries in the dominant shoulder due to repetitive abduction and external rotation of the arm. Position-dependent compression of the subclavian and axillary vessels may result in functional limitation and a decline in overhead athletic performance as a result of heaviness, fatigue, paresthesias, and effort-related pain. When these symptoms are associated with pulse deficits, pallor, or differences in temperature in the dominant or spiking arm and hand, aneurysm formation with thromboembolism might be present. In volleyball, emboli in the spiking hand have been reported to originate from aneurysms in the hypothenar, forearm – and more proximal – in the ipsilateral shoulder.

Figure 2 A comparison of the acute injury patterns observed in two prospective epidemiological studies. The presented data represent the number of acute time-loss injuries in one season sustained by both male and female volleyball players during both training and competition (combined) while participating in a European national adult competitive amateur league.
Posterior Circumflex Humeral Artery (PCHA) Pathology

In 1993, Reekers\textsuperscript{95} was the first to suggest a causal relationship between traumatic aneurysm of the PCHA and volleyball. Fifteen years later, between November 2008 and November 2010, six volleyball players with ischemic digits of the dominant hand presented themselves in the Academic Medical Center (AMC) in Amsterdam. Following angiography of the hand and forearm, some of these players showed small microemboli in the digital arteries and were given thrombolytic therapy (Figure 3). Within weeks of returning to play, these players returned with identical complaints. Further evaluation, including angiography of the shoulder, showed an aneurysmatic dilatation of the PCHA with thrombus formation and emboli in the digital arteries of the ipsilateral limb (Figures 3 and 4). These players were treated by ligation of the PCHA to prevent further embolization and after rehabilitation returned to play at the highest level of competition within 3-4 months after surgery. All were elite male volleyball players active in the national top league and between 21 and 31 years of age.

This sudden increase in volleyball players with digital ischemia due to PCHA pathology was noted in 2010 by the physiotherapist of the Dutch national beach volleyball team and the former physician of the Dutch national indoor volleyball team. At that time, just five case reports had been published worldwide on volleyball players with finger ischemia due to arterial emboli originating from a thrombosed aneurysm in the PCHA in the dominant shoulder.\textsuperscript{68,95,96,115,126} Knowledge about this injury needs to be extended on an international scale considering the potential amputation of a finger as the devastating end result in a population of young, healthy and fit elite volleyball players. Therefore, from 2010 on, we set out to elucidate the unexplored entity of PCHA pathology among volleyball players.

\textbf{Figure 3\textsuperscript{99}} Digital Subtraction Angiography of the right hand of a 27-year-old volleyball player with ischemic symptoms of multiple digits. The arrows point to multiple abrupt stops in digital arteries, caused by microemboli
Pathogenesis
Several studies suggest that repetitive powerful overhead movements in volleyball, like spiking and serving, cause chronic vessel wall injury as a result of positional traction and compression of the proximal PCHA. This cumulative PCHA trauma can cause a continuum of pathology ranging from local intimal hyperplasia to vessel widening of <150% (dilatation) and >150% (aneurysm), and occlusion. Although small in size, turbulent flow in these aneurysms readily produces mural thrombus, which has a high propensity to embolize to the distal extremity, particularly during repetitive overhead movements. It is assumed that during the spiking and serving motion in volleyball, when the humeral head acts to compress the aneurysmal PCHA and the intraluminal thrombus like a tube of toothpaste, thrombi can be extruded from the aneurysmal artery branch into the axillary artery and embolize to the circulation of the forearm, hand, and digits.

Symptoms
In an early stage of disease, symptoms might only manifest after overhead movements in volleyball as a result of embolization into the digital circulation. This can lead to a wide range of symptoms in the spiking hand during or directly after volleyball. Similar symptoms will often be caused by, and attributed to, musculoskeletal injuries, and might therefore initially be perceived as minor, and thus ignored by the athlete. As a
result, athletes generally present themselves in an advanced stage of the disease with debilitating symptoms of ischemia in the spiking hand in daily life, like coldness, discoloration and paresthesia.\textsuperscript{68,95,96,115,126} These symptoms result in an inability to play volleyball and reduced daily quality of life, and may ultimately lead to necrosis and finger loss when trivialized. Therefore, awareness of these symptoms, with a timely detection is warranted. Which exact symptoms are associated with arterial emboli in the spiking hand originating from an aneurysmal and thrombosed PCHA in the dominant shoulder in volleyball players is unclear. Even so, the prevalence of these symptoms, related to digital ischemia and possibly due to vascular pathology in the shoulder, among elite indoor and beach volleyball players is unknown. These topics are addressed in Chapters 2, 4 and 5.

**Diagnostics and imaging**

Diagnosis of PCHA pathology is established based on history-taking, physical examination and diagnostic imaging, both non-invasive and invasive. Non-invasive testing, like digital photoplethysmography and vascular ultrasound (US), are used in the work-up towards invasive testing, i.e. digital subtraction angiography (DSA), the standard of reference or less invasive computed tomographic angiography (CTA) and magnetic resonance angiography (MRA). All three are associated with ionizing radiation or the use of potentially nephrotoxic contrast media. However, these modalities are currently required for diagnosis and treatment planning. An attractive modality for low threshold vascular imaging is US, which is readily available, applicable on-site, inexpensive, and patient friendly. In general, US is the first-line imaging modality for peripheral aneurysm assessment.\textsuperscript{42} It enables non-invasive measurement of vessel diameters and detection of intravascular thrombus.\textsuperscript{42} Currently there is no standardized vascular US protocol available for imaging of the PCHA.\textsuperscript{17} International standardization of PCHA imaging with US would assist in accurate assessment in both a clinical and screening setting, and for research purposes. Imaging of the PCHA with US is discussed in Chapters 6, 7 and 8. Imaging of the PCHA with MRA is described in Chapter 10.

**Treatment and rehabilitation**

Potential invasive treatment options include surgical ligation and endovascular coiling.\textsuperscript{3} Coil embolization is an option if the proximal PCHA is relatively long, unaffected, and contains no thrombi.\textsuperscript{3} Possible complications of coiling include the potential for further embolism and dislocation of coils or plugs.\textsuperscript{3} Ligation of the PCHA can be performed if an endovascular approach is not possible or when it is preferred by either the patient or surgeon. Endovascular treatment can result in an earlier return to the previous level of competition compared to surgical ligation.\textsuperscript{3} The rehabilitation programme might be supervised by a physiotherapist and consist of early mobilization without making the overhead motion, followed by a full active range of motion exercises approximately 6 weeks after surgery. A full return to the level of previous activity is realized within 3-4 months after surgery\textsuperscript{120}, with continuation of anti-thrombotic medication (aspirin 100 mg
daily) for an arbitrary total of 6 months. Since the effect of conservative management for PCHA pathology with symptomatic emboli in the spiking arm in volleyball players is unknown, it is elaborated on in Chapter 10.

**Prevention**

Knowledge about this vascular injury will raise awareness and enable recognition, which is an important factor in preventing serious ischemic complications. As stated above, invasive treatment options for PCHA aneurysms result in several months of revalidation and absence from sports activities. However, if PCHA pathology can be detected at an early stage, serious ischemic complications, irreversible tissue damage, and surgical ligation of the PCHA might be prevented. Since volleyball players are considered potentially at risk for developing critical digital ischemia, analysis of the presence of PCHA pathology, and associated risk factors, is warranted for prevention. Ultimately, as shown in Chapter 9, establishing risk profiles of individual athletes could support clinical management and optimize care. Moreover, since volleyball players become at risk for PCHA pathology when symptoms of digital ischemia arise, analysis of the presence of risk factors associated with these symptoms could serve as a first step in prevention. Therefore, Chapters 3 and 5 discuss risk factors associated with symptoms of digital ischemia in elite volleyball players.

In summary then, elite volleyball players are at risk of ischemic digits due to arterial emboli originating from an aneurysmal and thrombosed PCHA in the dominant shoulder. Knowledge about this injury needs to be extended on an international scale considering the potential amputation of a finger as the devastating end result in a population of young, healthy and fit elite volleyball players.
1.2 RESEARCH QUESTIONS

Based on the preceding arguments, the thesis is divided into three parts: I) symptomatology and associated risk factors; II) imaging; and III) clinical management, and involves the following research questions:

PART I - Symptomatology and associated risk factors
1. Which symptoms are most likely to be associated with PCHA pathology with distal embolization in the spiking hand in volleyball players? *(Chapter 2)*
2. Can the newly developed Shoulder PCHA pathology and digital Ischemia – Questionnaire (SPI-Q) be used for the reliable detection of these symptoms in elite volleyball players? *(Chapter 4)*
3. What is the prevalence of these symptoms in the spiking hand among elite indoor and beach volleyball players? *(Chapters 2 and 5)*
4. Which risk factors are associated with these symptoms among elite indoor and beach volleyball players? *(Chapters 3 and 5)*

PART II - Imaging
1. How can the proximal PCHA be assessed in a standardized way using vascular US, and can this be formulated in a protocol? *(Chapter 6)*
2. Can the newly developed Shoulder PCHA pathology and digital Ischemia – UltraSound (SPI-US) protocol be used for reliable diameter assessment of the proximal PCHA and DBA? *(Chapter 7)*
3. What is the prevalence of PCHA aneurysms in the dominant shoulder among international elite indoor and beach volleyball players? *(Chapter 8)*
4. Which characteristics, such as anatomy, branching pattern, course and diameter, distinguish the PCHA from the DBA? *(Chapter 8)*

PART III – Clinical management
1. What is the association between PCHA pathology in the dominant shoulder and self-reported symptoms of digital ischemia in the spiking hand in elite volleyball players? *(Chapter 9)*
2. Which risk factors are associated with PCHA pathology in the dominant shoulder in elite volleyball players, and is a dose-response relationship present? *(Chapter 9)*
3. Can risk profiles of individual athletes be recognized based on the combination of the presence of PCHA pathology and symptoms of digital ischemia, and can clinical management guidelines be formulated per profile? *(Chapter 9)*
4. Can conservative management for a PCHA aneurysm with symptomatic embolization in the spiking arm in volleyball players be considered as an alternative to invasive treatment modalities? *(Chapter 10)*
1.3 - OUTLINE OF THE THESIS

In volleyball, spiking is the act of scoring a point by slamming the ball over the net into the opposing court effectively. In this thesis, the first steps of elucidating the unexplored entity of Shoulder PCHA pathology and digital Ischemia in Known Elite volleyball players are made in order to provide an effective contribution to knowledge. Hence the title: SPIKE the PCHA. The thesis is divided into three parts: the first part covers symptomatology and associated risk factors, the second part focuses on imaging, and the third part concerns clinical management.

PART I – Symptomatology and associated risk factors
Chapter 2 describes which symptoms are most likely to be associated with PCHA pathology with distal embolization in volleyball players, and assesses the prevalence of these symptoms in the dominant limb among elite male indoor volleyball players in the Netherlands.

In Chapter 3 we assess which risk factors are associated with self-reported symptoms of digital ischemia among elite male indoor volleyball players in the Netherlands.

The test-retest reliability of the Shoulder PCHA pathology and digital Ischemia – Questionnaire (SPI-Q), which can be used for the detection of self-reported symptoms of digital ischemia in elite volleyball players, is determined in Chapter 4.

In Chapter 5 we assess the prevalence of self-reported symptoms of digital ischemia in the spiking hand, and associated risk factors, among international world-class beach volleyball players.

PART II – Imaging
In Chapter 6, we present a 4-step standardized vascular ultrasound (US) protocol for the assessment of the proximal PCHA: the SPI-US protocol (Shoulder PCHA pathology and digital Ischemia – UltraSound protocol).

The inter-observer reliability of the SPI-US protocol for the diameter assessment of proximal PCHA and DBA is assessed in Chapter 7.

In Chapter 8 the prevalence of PCHA aneurysms in the dominant shoulder of elite volleyball players is determined, and PCHA and DBA characteristics are described that can be used to accurately identify and assess the PCHA using the SPI-US protocol.
PART III – Clinical management

In Chapter 9 four risk profiles among elite volleyball players are presented based on the presence of US-detected PCHA pathology in the dominant shoulder, reporting of symptoms of digital ischemia in the spiking hand, and identified risk factors. Per profile, guidelines for clinical management are proposed to optimize care.

Chapter 10 describes the effect of conservative management for a PCHA aneurysm with symptomatic embolization in the spiking arm in a professional volleyball player with the use of novel Magnetic Resonance Angiography.