

Symptomatic os trigonum in national level javelin thrower: a case report

Spencer Bell, DC, MASc¹

Cameron Borody, DC, FRCCSS(C)²

Introduction: *Os trigonum syndrome is a relatively uncommon condition, resulting from compression of a congenital bony anomaly (os trigonum) and adjacent soft tissues during repetitive hyper-plantarflexion. This condition is currently well-described in ballet, soccer, and running athletes, but few cases exist describing os trigonum syndrome in overhead athletes.*

Case Presentation: *A 22-year-old national level male javelin athlete presented with a recalcitrant history of posterior ankle pain following a hyper-plantarflexion mechanism. Imaging demonstrated a symptomatic os trigonum and inflammation of surrounding soft tissues. Re-aggravation following a conservative trial of care led to orthopaedic referral. Surgical excision of the os trigonum was performed with an open posterolateral approach. The athlete returned to competition three months later with no recurrence of symptoms.*

Summary: *This case discusses the clinical presentation, imaging, and management of a*

Introduction : *Le syndrome de l'os trigone est une pathologie relativement peu fréquente causée par la compression d'une anomalie osseuse congénitale (os trigone) et des tissus mous adjacents durant les mouvements répétitifs en hyperflexion plantaire. Cette pathologie touche souvent les danseuses de ballet, les joueurs de soccer et les coureurs. Peu de cas sont enregistrés chez les athlètes pratiquant des sports comportant des mouvements au-dessus de la tête.*

Présentation du cas : *Un lanceur de javelot de 22 ans se plaignait d'une douleur postérieure de la cheville déclenchée par des mouvements répétitifs en hyperflexion plantaire. Les examens par imagerie montraient un os trigone symptomatique et une inflammation des tissus périphériques. Une aggravation après une tentative de traitement conservateur a mené à une demande d'une consultation en orthopédie. L'excision chirurgicale de l'os trigone a été réalisée à ciel ouvert par voie d'abord ouverte postérolatérale. L'athlète a repris la compétition au bout de trois mois; ses symptômes ne sont pas réapparus.*

Résumé : *Cette étude de cas présente le tableau clinique, les examens par imagerie et la prise en charge*

¹ Division of Graduate Studies, Sports Science, Canadian Memorial Chiropractic College

² Assistant Professor, Clinical Education, Canadian Memorial Chiropractic College

Corresponding Author:

Spencer Bell

Canadian Memorial Chiropractic College, 6100 Leslie Street, Toronto, ON, Canada, M2H 3J1

E-mail: sbell@cmcc.ca

T: 416-482-2340 ext. 501

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symptomatic os trigonum and related pathologies in a javelin thrower.

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KEY WORDS: chiropractic, os trigonum, posterior ankle impingement, plantarflexion, arthroscopic surgery, athletics, javelin

Introduction

Os trigonum syndrome is a painful, relatively uncommon condition that results from compression of a congenital bony anomaly – an os trigonum, found at the posterolateral aspect of the talus – and nearby soft tissue in the posterior tibio-calcaneal interval.¹ Os trigonum syndrome fits into a broader diagnostic umbrella of posterior ankle impingement syndrome (PAIS), a term which encompasses any condition characterized by posterior ankle pain exacerbated with plantarflexion of the ankle.^{2,3} While the presence of an os trigonum increases the risk of symptom development, a considerable disease list, which has been previously described elsewhere in great detail, contributes to PAIS.³⁻⁶ Some examples include osteophyte formation, thickened joint capsule and surrounding ligaments, additional accessory ossicles, fracture or syndesmotic injury, and accessory soft tissue components. As such, the syndrome is synonymous with several terms, including posterior talar compression syndrome, os trigonum syndrome, posterior ankle block, nutcracker-type impingement, and posterior tibiotalar impingement syndrome.⁵ More generally, the pathologic contributors can be categorized as either osseous or soft tissue, and, when affected, may all contribute to pain in hyper-plantarflexion.⁶

The presence of an os trigonum is a relatively common occurrence in the general population, with an estimated prevalence of nine to 25% in normal feet and ankles, often observed bilaterally.⁷⁻⁹ Typically, it is asymptomatic in the non-sporting population.^{3,7,10} First described in 1804 by Rosenmuller¹¹, an os trigonum was theorized to develop from a secondary ossification centre by Turner in 1882¹² which was confirmed by McDougall in 1955^{13,14}. The secondary ossification centre can be visualized on a lateral ankle radiograph between ages seven to 11 in females, and 11 to 13 in males; fusion occurs within one year after

d'un os trigone symptomatique et des pathologies apparentées observés chez un lanceur de javelot.

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MOTS CLÉS : chiropratique, os trigone, conflit postérieur de la cheville, flexion plantaire, chirurgie par arthroscopie, athlétisme, javelot

initial appearance.^{7,13-15} In the event of failed fusion, an os trigonum remains and is attached to the posterolateral talus via a fibrous or cartilaginous synchondrosis.^{7,14} The os trigonum is believed to be vulnerable to trauma, specifically in repeated plantar flexion of the ankle.¹⁴

The presence of an os trigonum in itself is not considered sufficient to produce PAIS; commonly, it is considered an incidental finding. Rather, it is the combination of pain in plantar flexion and exposure to repetitive loading in positions of hyper-plantarflexion that is believed to contribute to development of PAIS, regardless of bony anatomy.¹⁰ There is debate whether an acute incident may also be causative.^{4,16}

The close proximity of the flexor hallucis longus (FHL) tendon, which passes in the fibrous-osseous tunnel posterior to the medial malleolus and follows the groove between the medial and lateral posterior talar tubercles,¹ predisposes this tissue to stenosing tenosynovitis in the presence of an os trigonum – a common concomitant condition.^{5,17} Due to constant pressure exerted on the os trigonum by the FHL tendon^{1,5,18}, a chronic irritation of the FHL tendon inside its sheath can result in compression, synovitis, hypertrophy, nodules, and partial tearing^{5,17}.

PAIS has been well-described in ballet dancers, gymnasts, soccer players, and runners², but has not been well described in overhead athletes. The objective of this case report is to describe the presentation of a national level male javelin athlete with PAIS due to an os trigonum with concomitant FHL tenosynovitis. The case will also explore the prevalence, clinical presentation, and management strategies, as well as biomechanical considerations in the throwing athlete. The patient gave signed consent for release of his information to be included in this manuscript.

Case Presentation

History

A 22-year old male national level javelin thrower presented to a Sports Specialist chiropractor (Fellow of the Royal College of Chiropractic Sports Sciences, Canada) with an acute episode of chronic left posterior ankle pain. The pain began during a competition throw three months prior, where he reported over-extending his lead foot during the “block”, also known as the delivery step (Figure 1). This phase is where the javelin athlete ends their run-up and cross-over steps (including the penultimate) by planting their lead foot and blocking the body from moving forward, thus creating a strong pivot point for the throwing side of the body to rotate around.¹⁹ Offseason weight training and practice throws were not aggravating, but he continued to report intermittent pain and apprehension with competition level throws, where he describes extending his block foot further to gain an advantage.

At the time of assessment, pain intensity was rated as 3/10, but he noted that it could be 7/10 while throwing. The patient reported intermittent symptoms in the same region for approximately six years prior to presenting at the clinic. Symptoms typically came on in late spring when the outdoor throwing season begins in Canada, and symptoms dissipated in the fall when throwing season typically ends and he is able to rest for several weeks.



Figure 1.

The “block” phase at the end of the penultimate, immediately prior to throwing the javelin. The leading (left) foot is loaded in hyper-plantarflexion during this phase (arrowhead).

Previously, the patient had been diagnosed as an Achilles tendinopathy – a common misdiagnosis due to symptom location, as described in detail elsewhere²⁰ – which was treated conservatively by a therapist. Symptoms abated at that time, although this coincided with offseason rest, which may have contributed to symptom resolution.

Additional complaints included a chronic Grade 2 strain of the right external oblique near the pubic symphysis, suffered approximately six months prior to the time of initial assessment during a competition throw. This was treated conservatively by a therapist and was beginning to resolve at the time of assessment. The patient is right-handed and otherwise had no major prior injuries.

Physical Examination

Upon observation, there was no obvious signs of swelling, discolouration, or other visual abnormalities. Palpation revealed tenderness over the posterolateral ankle, slightly anterior to the Achilles tendon, as well as in the flexor compartment on the posteromedial ankle. Pain in both regions was increased with passive plantar flexion of the left ankle as well as with passive dorsiflexion of the great toe. Passive motion of the great toe caused a squeaking sound from the posteromedial ankle, which could be felt on palpation as a vibration over the flexor hallucis longus tendon at the medial ankle. Motion palpation revealed hypomobility in the cuboid and navicular bones, and restriction in subtalar eversion. There was a 20% decrease in active dorsiflexion of the left ankle in comparison to the right ankle as measured with the standing knee to wall test. Walking, jogging, lunging, squatting, weightlifting, and plyometric training did not exacerbate symptoms. Toe walking and standing inversion were mildly uncomfortable. Single leg squat revealed significant difficulty balancing on the left side. Muscle testing and neurologic examination was within normal limits.

Diagnosis and Management

A lateral radiograph of the left ankle demonstrated an os trigonum (Figure 2A). Based on a combination of clinical symptoms, mechanism of injury, and radiographic findings, a working diagnosis was provided of PAIS (secondary to an os trigonum) with associated tenosynovitis of the flexor hallucis longus tendon. A 10-week trial of conservative care was implemented, including manipulation and mobilization of the foot and ankle, and Active Release

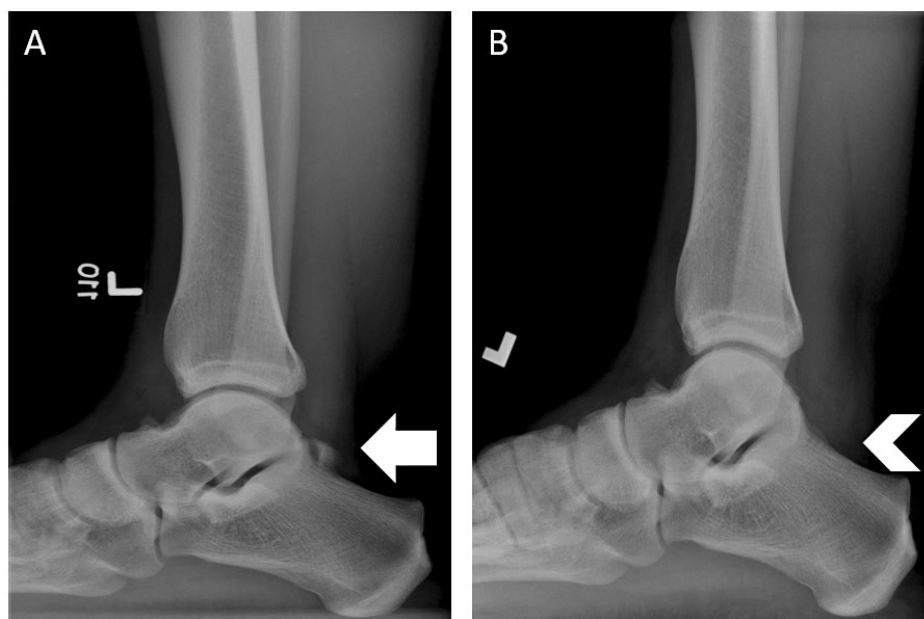


Figure 2.
Radiograph of athlete's left ankle pre and post surgery. (A) Lateral ankle radiograph demonstrating presence of os trigonum (arrow). (B) Post-surgical lateral ankle radiograph demonstrating the successful resection of the os trigonum (arrow head).

Techniques® to address soft tissue. The treating chiropractor and coach collaborated to integrate a rehabilitation program into the athlete's current training regimen, which was modified to avoid aggravating positions of excessive plantar flexion, thus minimizing disruption to his training schedule. Rehabilitation was geared towards improving mobility, intrinsic strength, and balance of the foot, as well as improving strength and coordination of the lower kinetic chain. At the end of the 10-week trial of care, the patient's condition stabilized, ankle dorsiflexion was symmetrical, and he was able to return to full training volume and competition-level throws with no pain. However, during a high-level competition eight weeks later, the patient re-aggravated the condition with the same mechanism.

At this point, the patient was referred to a sports medicine physician for co-management, who prescribed an MRI (Figure 3). The MRI identified marrow edema in both the os trigonum and adjacent talus extending into the body. There was fluid present at the synchondrosis and moderate fluid around the os trigonum extending into the posterior ankle recess. High signal was noted at the talar attachment of the posterior talofibular ligament (PTFL) and posterior tibiotalar ligament. There was also mild soft tissue edema and fluid present in the flexor hallucis lon-

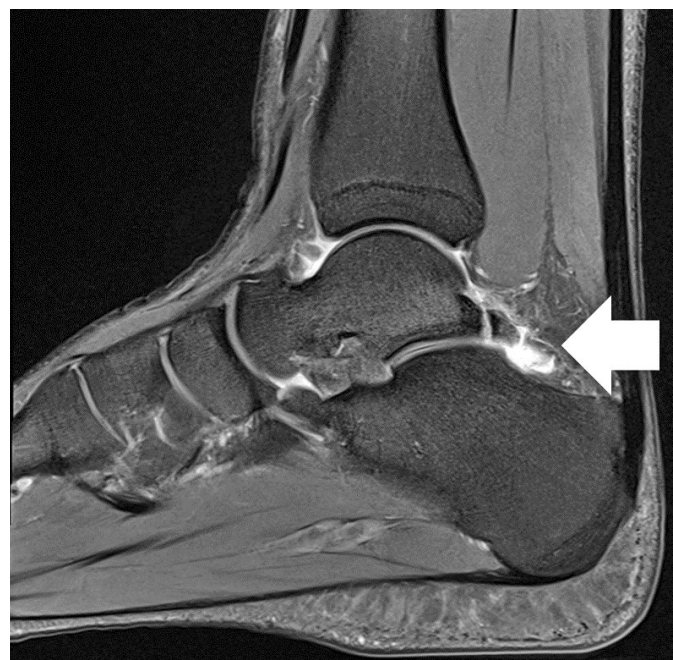


Figure 3.
MRI imaging of athlete's left ankle prior to surgery. Proton density fat suppression sagittal MRI slice demonstrating marrow edema in the os trigonum extending into the talar body and posterior synovial recess (arrow).

gus, tibialis posterior, and flexor digitorum longus tendon sheaths.

Due to the recurrent nature of symptoms and failure of conservative care, the patient was referred for orthopaedic consultation. Surgical excision of the os trigonum and subtalar synovectomy was recommended and successfully performed using an open posterolateral approach (Figure 2B). Following a standard post-surgical plan of management to control inflammation and restore range of motion, the athlete participated in a rehabilitation program designed to re-establish strength, stability, and coordination through the lower kinetic chain, trunk, and upper limbs. The rehabilitation program was influenced by concepts from dynamic neuromuscular stabilization (Figure 4) and emphasized the derivation of foot support in a tripod fashion (1st and 5th metatarsal heads and the calcaneus) to maximize arch support.²¹ The program was global, with an emphasis on establishing control and strength through the foot and was designed to coordinate the integration of intra-abdominal pressure regulation with the dynamic and stabilizing functions of neighbouring joints and extremities to optimize movement competency and throwing efficiency. The athlete returned to full competition three months later with no pain and had no recurrent issues at one-year follow-up.

Discussion

PAIS Management

The first clinical report of osseous impingement of the posterior ankle was in 1982.²² The overall prevalence of PAIS remains unknown,³ due to a paucity of published literature on the subject, and the wide number of conditions that may contribute to its development.^{2,3} Albisetti reported a 6.5% prevalence of PAIS in 186 trained ballet dancers over a one year period²³, although this may be as high as 30% or greater in the same population²⁴, and as high as 60% in a wider spectrum of athletics²⁵. Ribbans *et al.* presented unpublished data from 18 teams under the English Cricket Board in 2001-02, showing PAIS to be the second-leading cause of injury in the foot and ankle region in this population – more than lateral ankle sprains and Achilles disorders combined.³ This was reported to most commonly affect the front foot of fast bowlers, although there is little available literature to support these observations in cricket athletes.



Figure 4.

Example of rehabilitation exercise based on dynamic neuromuscular stabilization principles, designed to maintain the arch structure of the foot in a centered tripod support.²¹

Typically, conservative measures are first considered for PAIS management, which are shown to be effective in the general population, although due to heterogeneity and a lack of understanding in causative mechanisms, treatment is seen to vary considerably.^{2,3,23,26} Mouhsine *et al.* reported 16 of 19 athletes responded effectively to corticosteroid injection in a case series and were returned to sport with no further issues at a follow-up time of two years.¹⁶ Surgical intervention was successfully performed on the three recalcitrant cases, and athletes returned to sports within seven to nine months from the date of initial injury. Similarly, Robinson *et al.* presented a case series of 10 soccer athletes with subacute PAIS, following an inver-

sion mechanism of injury, in which two of these athletes were found to have an os trigonum, and one subsequently underwent surgery for removal of the bony anomaly.²⁷ Hedrick *et al.* report a 60% success rate with NSAIDs, steroid injections, cast immobilization and rehabilitation.²⁵ However, the short-term follow-up and lack of prospective studies limits the conclusions drawn surrounding conservative management of PAIS, particularly as recurrence of symptoms is quite common. Based on all available evidence, despite empirical recommendation for conservative treatment as a first option for treatment, there is little substance to provide evidence-based recommendations on the choice of non-surgical interventions, or prognostic factors to inform the patient on their condition.³

In the presence of a symptomatic os trigonum, it has been suggested that surgical intervention may be necessary for longstanding relief of symptoms⁴, although recommendations for this approach are equally debatable. A comprehensive review of available literature by Ribbons *et al.* demonstrates a predominance of retrospective Level IV and V evidence and significant heterogeneity in reporting and outcome measures, limiting conclusions that can be made.³ However, based on their review of 26 papers (384 open surgical procedures, 521 arthro-endoscopic procedures), surgical interventions overall were seen to have a self-reported 67-100% effectiveness rate in returning the athlete to sport, with low complication rates of 4.8% (arthro-endoscopic), 3.9% (posteromedial), and 14.7% (posterolateral), respectively, many of which were temporary in nature.³

There is debate as to the most appropriate surgical approach, although there appears to be no clear advantage based on outcome.^{1,3,4} Although a slightly faster return-to-play was noted with the arthro-endoscopic approach over the open approach³, it is suggested that this approach may have greater risk for injury to cartilage due to the small space¹. Overall, complication rates are very low with surgical intervention for PAIS, the most reported of which involve injury to the sural and tibial nerves, many of them transient and short-term.^{1,3} Regardless of intervention, most athletes tend to return to their sport of choice, with many authors highlighting the importance of a dedicated therapist to facilitate the successful return to competition.²⁸ It should be noted that, due to the low quality of evidence, these findings should be interpreted with caution.

Lack of return to play guidelines for PAIS make it difficult to accurately determine prognosis for an individual athlete. Senecal *et al.* present a case report of a middle-aged recreational hockey player, suggesting the utility of a progressive return to play program adopted from that following ankle sprains.²⁶ Rogers also proposed a five step post-surgical rehabilitation protocol for horizontal jump athletes,⁴ while Coetzee *et al.* presented a rehabilitation program designed specifically for ballet dancers, with the “Pointe Functional Tests” – comprised of the topple test, airplane test, and single-leg sauté test – as the final step before returning to relevé.²⁸ All programs follow logical, progressive steps towards recovery, with good self-reported results, although none have been explored further in prospective randomized control trials to assess efficacy.

Mechanism of Injury

Currently, there is discussion as to the development of PAIS. The first, and most commonly described, is through repetitive hyper-plantarflexion in loaded movements. A 2016 study from Russell *et al.* used high resolution MRI to assess 6 ballet dancers in a non-weightbearing en pointe position.²⁹ Every participant demonstrated convergence of the posterior edge of the distal tibia with the posterior talus and superior calcaneus; providing further visual confirmation of the “nutcracker” mechanism commonly described in previous literature.² Interestingly, in this position of extreme plantarflexion, the authors described incongruency of the talocrural joint in all athletes, noting one third of the articular surface of the tibial plafond resting on the posterior talus in this position, with anterior translation of the talus due to compression of the bone at the posterior aspect. The en-pointe and demi-pointe positions seen in ballet dancers have been proposed as a potential contributor to the high incidence of PAIS in this population, particularly as this highly repetitive forced plantar flexion is practiced during skeletal maturation.^{6,14}

In a biomechanical study of soccer players, the degree of plantar flexion during ball strike exceeded that which was reproducible with passive clinical assessment, suggesting a consistent, repeatable compression of the posterior tibiotalar structures.³⁰ Rogers describes the mechanism of horizontal jumping athletes, who transition their running momentum into take-off foot in a rapid (40-70ms) compressive-based plantar flexion movement,

during which time upwards of 10-15 times body weight is applied through the lead leg in maximal plantarflexion.⁴ Furthermore, Rogers describes increased plantar flexion and braking load in the lead leg the further the lead leg is in front of the body's centre of mass.⁴ This is thought to be mechanistically analogous to the forces experienced by a javelin thrower during the final delivery step and matches the mechanism of injury for the athlete in this case report, although there is no literature to support this supposition.

PAIS may also develop as the result of an acute injury. Mouhsine presented a case series of 19 athletes with PAIS, noting that 8 athletes presented with persistent pain following an ankle inversion sprain mechanism and a failed standard course of treatment for lateral ankle sprains.¹⁶ Additional mechanisms of acute injury are postulated, including a single plantar flexion event and forced dorsiflexion leading to avulsion of the posterior talofibular ligament which can attach to the os trigonum.³¹

In general, whether acute or chronic, the available literature describes a consistent, logical mechanism of injury for impingement of osseous and soft tissue structures in the posterior ankle, particularly in the presence of an os trigonum or an elongated lateral tubercle.³² However, anatomical, biomechanical, and capacity-based risk factors need to be determined to identify those at greatest risk of developing PAIS. In the absence of sound, evidence-based recommendations, intimate knowledge of a sport's physical demands becomes imperative for the treating clinician to understand. A team-based approach is of paramount importance, with coaches, therapists, and surgeons communicating freely to identify relevant potential risk factors that may affect prognosis, particularly positions of extreme plantar flexion associated with sport. This knowledge is important to consider in clinical management of patients with PAIS.

Global Biomechanical Considerations

Due to the chronicity of PAIS in many athletes, it is essential to monitor the athlete for compensatory movement strategies that may affect health status and performance. This case presents a national level javelin thrower with recalcitrant PAIS and a subacute external oblique strain, potentially representing an injury higher in the kinetic chain, that may be influenced by compensatory movement strategies. There is growing evidence that painful conditions in the lower limb can affect neurological pat-

ternerng in the hip, pelvis, trunk, and the upper limb which holds potential to affect performance and increase injury risk in the overhead athlete. A review by Steinberg *et al.* into the impact of painful lower leg conditions on hip muscle performance demonstrated decreased strength and endurance, and delayed onset and offset of hip musculature in various movement patterns.³³ This co-dependent relationship between segments of the lower kinetic chain has been explored previously in other conditions including patellofemoral pain syndrome.³⁴⁻³⁵

Arguably of greater consequence in overhead athletes are the compensatory ramifications experienced higher in the kinetic chain. In this particular case, it could be suggested that altered activation patterns at the hip and pelvis may alter frontal plane mechanics and coordination of trunk rotation, leading to greater lateral trunk lean and loading in the abdominal wall – as demonstrated by an chronic external oblique strain in this athlete, as well as the throwing arm.³⁶ A study of 99 college-aged baseball pitchers by Solomito *et al.* demonstrated a 4.8% increase in varus moment of the elbow and 3.2% increase in glenohumeral internal rotation moment for every 10° increase in trunk lean.³⁷ These findings were echoed by Oyama *et al.* in high school pitchers.³⁸ Furthermore, altered timing and magnitude of trunk rotation, a ramification of altered pelvic and hip control³⁶ has been demonstrated to significantly increase external rotation angles and proximal forces experienced at the glenohumeral joint^{36,39}. These combined findings are important, as the load placed on the UCL during pitching is already close to matching the ultimate moment of failure observed in cadaveric UCL studies.^{37,40} Additional load through the UCL and rotator cuff may have an added biomechanical cost in the throwing athlete, with only negligible performance gains.^{36,37,39}

In a comprehensive review of throwing mechanics, Chu *et al.* highlights the necessary integrative nature of linked segments throughout the body in performing a thoroughly complex activity such as throwing which incorporates the entire kinetic chain.⁴¹ The authors suggest that deficiencies in any of these areas may have a detrimental effect on performance and injury rates in the throwing population, thus endorsing the necessity of a thorough clinical and functional evaluation of the leg, hip, core, scapula, and shoulder for the overhead athlete. Ultimately, while there is no direct correlation established between injury of the lower quarter and upper extremity loads experi-

enced in the throwing athlete, this information suggests the importance of assessing global biomechanical function of the throwing athlete, particularly in the presence of painful lower quarter conditions, to screen for aberrant movement patterns and the potential for increased loading throughout the kinetic chain.

Limitations

This is a case report, limiting the findings to a single case. It is not prudent to generalize the findings of this study to the general public, or to other patients. Further observation and larger prospective trials are required to assess individual contributors to PAIS, and to identify prognostic factors and the impact of conservative and surgical interventions on PAIS patients.

Summary

PAIS has a plethora of causes. While conservative treatment is encouraged as the primary intervention, surgical intervention may be required in the presence of osseous anomalies such as os trigonum for long-term relief of symptoms, particularly in elite athletes. This case describes a 22-year-old national level male javelin athlete with a 3-month history of posterior ankle pain following a hyper-plantarflexion mechanism during a competition-level javelin throw. Radiographs and MRI demonstrated the presence of an os trigonum with inflammation of surrounding soft tissues matching the clinical presentation. After a 10-week trial of conservative care was unsuccessful, surgical intervention was successfully performed using an open posterolateral approach. The athlete returned to javelin competition 3 months later with no recurrence of symptoms. The generalizability of the approach in this athlete is limited, as this represents a single case. Future research should focus on prospective studies to identify key prognostic indicators and standardize treatment methodologies, and a strong effort should be made to improve reporting in published studies. The reader is reminded of the potential for global biomechanical ramifications throughout the kinetic chain in response to painful lower limb conditions, and the importance of observation and full kinetic chain assessment in the overhead athlete before returning them to sport.

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