Prevalence and Anatomical Distribution of Bone Stress Injuries in the Elite Para Athlete

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Abstract: Bone stress injury is a common overuse injury in athletes. Risk factors for bone stress injury in athletes include the female athlete triad (triad); this has not been evaluated in para athletes. The aim of this study was to identify risk factors, prevalence, and anatomical distribution of bone stress injury in para athletes. A cross-sectional online survey on health characteristics and previous fractures including bone stress injury was completed by para athletes training for the 2016 or 2018 Paralympic Games. Two hundred sixty para athletes completed the survey (659 invited, response rate = 40%). Half reported previous fracture, and bone stress injury was reported in 9.2% of all athletes. Twenty-four athletes (11 men and 13 women) sustained one or more bone stress injury, including 13 athletes with two bone stress injuries. No risk factors of the triad, disability type, or duration of disability were associated with bone stress injury. Injuries were most common in the metatarsals ($n = 8$) and hand/wrist ($n = 7$). In an elite para athlete population, locations for bone stress injury included both the upper and lower limbs. Clinically, para athletes presenting with pain localized to bone require further workup to evaluate for bone stress injury particularly for pain in both upper and lower limbs. Further research is required to identify risk factors for bone stress injury in para athletes.

Key Words: Stress Fracture, Para Athlete, Female Athlete Triad, Relative Energy Deficiency in Sport, Adaptive Sports, Paralympic Athlete

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Bone stress injury (BSI) is an overuse injury common in athletes. Previous reports suggest that women may be at higher risk for BSI than men.[1] The pathophysiology of BSI relates to cyclic overload to bone that results in microtrauma and can progress to a stress fracture or full fracture.[2] Untreated, this injury could result in the need for surgery and long-term functional impairment. Even when the diagnosis of BSI is made, the injury may limit an athlete being able to effectively train or compete at all levels of sports including in the Olympics.[3] Bone stress injuries were 2.2% of all injuries sustained during the Rio de Janeiro 2016 Summer Olympics and were most common in track and field athletes (44% of all athletes with this form of injury).[3] A BSI may occur in an athlete with normal bone who exceed the loading capacity of bone, although impaired bone quality may further increase susceptibility to injury.

Athletes with disability, also referred to as para athletes, are a population at risk for impaired bone health resulting from the combination of impaired mobility and behaviors associated with sports participation. The female athlete triad (triad)[4] is the interrelationship of low energy availability (EA), menstrual dysfunction, and impaired bone density.[4] Although the true prevalence of the triad is unknown in para athletes,[5] a recent report in 260 athletes preparing for the 2016 or 2018 Paralympic Games identified a high proportion of athletes with risk factors for the triad.[6] Of all athletes surveyed, 65% of all athletes reported were attempting to lose weight or change body composition to enhance performance, 32% had elevated scores on the Eating Disorder Examination Questionnaire pathological behavior subscale, 44% of women reported menstrual dysfunction, and 9.2% reported previous BSIs. The International Olympic Committee has adopted new terminology of Relative Energy Deficiency in Sport (RED-S), describing the effects of low EA beyond menstrual dysfunction and impaired bone health to include the broader effects on physiological and performance domains in both men and women, including those with disability.[7] The 2018 updated International Olympic Committee RED-S statement calls for further work to understand the influence of low EA on para athletes including methods to promote skeletal health in this population.[8]

Paralympic sports have seen immense growth in total participation since the first official Paralympics in 1960.[9] The limited studies evaluating para athletes have identified impaired bone health and low EA in athletes with spinal cord injury (SCI).[10–13] To date, no study has measured the influence of triad risk factors to the development of BSI, and the anatomical distribution of BSI has not been described in para athletes. Therefore, the primary aim of this study is to evaluate the association of triad risk factors with BSI in an elite population of para athletes. We hypothesized that BSI would be more common in para athletes with triad risk factors, impaired weight bearing, and longer duration of disability. A secondary aim was to describe the anatomical location of BSI in this population. We hypothesized that BSI would occur in both upper and lower limbs given increased reliance on the upper limbs to participate in many para sports.

METHODS

The research protocol and study design have been previously reported.[6] Briefly, US para athletes training to qualify

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for the 2016 Rio Summer Paralympic games or 2018 Pyeongchang Winter Paralympic games were invited using e-mail to complete a research study. The study design was cross-sectional and consisted of completing an online survey on health and injury. The survey was hosted by Research Electronic Data Capture on Partners HealthCare Research Computing, Enterprise Research Infrastructure & Services (ERIS) group. Research Electronic Data Capture is a secure web application for data acquisition and management.[14] The online survey tool enters the data directly into a database that can be extracted for analyses. The Partners HealthCare Institutional Review Board approved this research protocol. Electronic informed consent was obtained by all participants before completion of the survey. This report meets the STROBE guidelines for report of human subject research.

The survey contained demographic and health questions including age, sex, race/ethnicity, primary sport, disability type, ambulatory status, and primary mode of mobility (ambulatory or wheelchair use). Details on disability included age of onset and primary medical diagnosis. Sport type was separated into sports that emphasized leanness using modified criteria as previously published.[6,15]

Health questions included health behaviors related to the triad and RED-S. Questions included queries related to previous or current diagnosis of disordered eating/eating disorder and self-reported height and weight (to calculate body mass index [BMI]). We did not measure low EA directly. Instead, the Eating Disorder Examination Questionnaire subscales of pathologic behaviors and dietary restraint were each used because these have been shown to be associated with low bone mineral density (BMD) and BSI in athletes.[16,17] In addition, each participant was asked to select the best descriptor for current weight: underweight, ideal weight, or overweight (broken into categories of slightly overweight 5–10 lb, moderately overweight 10–20 lb, or very overweight >20 lb).

Each participant was asked about previous diagnosis of low bone density using dual-energy x-ray absorptiometry (DXA, including osteopenia or osteoporosis). Family history of low bone density and fracture was also queried.

Menstrual history was obtained in each female athlete, including age of menarche, number of menses in past 12 mos, previous oligomenorrhea (defined as 6–9 periods for 12 mos), or amenorrhea (<6 periods for 12 mos). In addition, each female participant reported use of hormonal therapy including oral contraceptive pills.

Each athlete was instructed to report previous diagnosis of a stress reaction or stress fracture. For each injury, additional requested details included date of injury, use of imaging (x-ray, magnetic resonance imaging, or computed tomography), and whether injury was sports related. Inclusion of a BSI (defined as either stress reaction or stress fracture) was determined after review by two study authors (AST, EGM) to ensure that the injury was in an anatomically plausible location and the injury occurred after the date of disability.

We summarized the athletes' demographic characteristics and anatomical site of BSI separated by sex and described differences between sex and within those with injury by age, BMI, ethnicity/race, and disability type. Because risk factors for BSI and low BMD are not well-described in the para athlete population, exploratory risk factors of interest included primary disability and related characteristics (including age of onset and duration of disability) and triad risk factors including diagnosis of disordered eating/eating disorder, elevated dietary restraint, low BMI, low BMD, previous BSI, and in women, age of menarche, and history of secondary oligo/amenorrhea. Mean and SD were used to describe continuous variables, and number and percentage were used to describe categorical variables. We assessed the associations between sex and history of BSI using χ^2 test. We then performed logistic regressions to assess association between history of BSI and each risk factor including triad risk factors, disability type, and duration of disability. We conducted logistic regressions separately by sex. All analyses were conducted in SAS v9.4 (SAS Inc, Carry, NC).

RESULTS

Subjects

Of 659 athletes invited to participate, 264 completed the survey (response rate = 40%). Four athletes were excluded from analysis because of incomplete survey responses. The resulting 260 participants in the final analysis were primarily men (n = 150), self-identified as white, and reported normal BMI (Table 1). The most common primary disability was SCI (30.4%) and lower limb amputation (25.8%).

Fracture and BSI

Overall, half of the total population reported history of fracture. Most individuals with central neurological injury, musculoskeletal disorder, visual impairment, and SCI reported previous fracture (Fig. 1). A total of 53 subjects reported previous BSI (defined as response of "yes" on survey to previous stress reaction or stress fracture). Each response was reviewed to determine whether these met study criteria for inclusion. This resulted in 24 athletes classified as having sustained one or more BSI (Table 2). Of the 24 athletes, 13 sustained one BSI and 11 sustained 2 BSIs.

Risk Factors for BSI

Univariate analysis did not identify any risk factor for BSI within the population studied. There was no difference in presence of injury between women and men (5.0% vs 4.2%, P = 0.22). Odd ratios exceeding 1.0 included participation in leanness sport, low BMI, low BMD, primary amenorrhea, and disability since birth; no values reached statistical significance and confidence intervals were wide (all P > 0.10).

Anatomical Distribution of BSI

Of 35 BSI sustained, most were in the lower limbs including spine and pelvis (n = 22, 63%), and 37% (n = 13) were reported in the upper limbs. The most common location for BSI was in the limbs including metatarsals (22.8%, n = 8), followed by hand and wrist (20%, n = 7), ribs (11.4%, n = 4), tibia (11.4%, n = 4), ankle (8.5%, n = 3), spine (5.7%, n = 2), pelvis (5.7%, n = 2), humerus (5.7%, n = 2), and one in the femur, knee, and sesamoid of the foot.

By total number of athletes with one or more BSI, the primary disability represented within the cohort was visual impairment (n = 9), followed by SCI (n = 5), cerebral palsy

TABLE 1. Demographics of athlete population

Demographic	Mean ± SD (Range)
Age, yr	31.7 ± 11.5
Height, m	1.7 ± 0.1
Weight, kg	70.6 ± 19.0
BMI, kg/m^2*	23.9 ± 5.0
Race	*n* (%)
White	213 (81.9)
African American	20 (7.7)
Asian	10 (3.8)
More than one race	10 (3.8)
Other	6 (2.3)
Did not report	1 (0.4)
Disability classification	*n* (%)
SCI	79 (30.4)
Lower limb amputee	67 (25.8)
Neurological injury	25 (9.6)
Visual impairment	24 (9.2)
Cerebral palsy	23 (8.8)
Other	13 (5.0)
Upper limb amputee	10 (3.8)
Musculoskeletal disorder	9 (3.5)
Arthrogryposis	5 (1.9)

*4 data points for height and/or weight were excluded because of outlier analysis suggesting possible error in survey response.

(*n* = 3), neurological injury (*n* = 2), and one in each classification of arthrogryposis, musculoskeletal disorder, lower limb amputation, upper limb amputation, and other. Accounting for differences in disability type, the highest prevalence of BSI was in athletes with visual impairment (9/24 or 37.5%), other classification and arthrogryposis (each 1/5, 20%), and cerebral palsy (3/23, 13%). Two BSI were observed in para athletes with visual impairment (*n* = 4), SCI (*n* = 2), cerebral palsy (*n* = 2), and one in arthrogryposis, neurological injury, and other classification.

DISCUSSION

The primary objective of this investigation was to report the prevalence and anatomical distribution of BSI in elite para athletes. Of 260 athletes, 9.2% (*n* = 24) reported history of one or more BSI from sport. The anatomical locations of injury primarily included the limbs of the foot and wrist/hand. Previous studies of BSI have reported primarily lower limb injuries in populations of military and athletes with no disability.[1] The large number of upper limb injuries in our population was not surprising and may be postulated to result from greater demands in use of upper limbs to participate in many para sports.

Using conventional risk factors of the triad and markers of low EA, we did not identify risk factors for BSI. There are multiple explanations for our findings. Our population had a small sample size that may have limited power to detect differences. In addition, the cross-sectional design may have limited detection of risk factors for BSI. Although the triad was first described over two decades ago,[18] the current published guidelines for risk assessment are based primarily on studies in young, exercising women without disability.[4] Although the triad and the expanded syndrome of RED-S represent important advances in understanding consequences of low EA in athletes,[4,8] further research specific to the para athlete may be required to understand health consequences in this athlete population.

Of athletes with BSI, the highest prevalence was in those with visual impairment. Notably, 37.5% of all para athletes with visual impairment sustained BSI. Of the nine athletes with two BSI, four were para athletes with visual impairment. Previous studies have not characterized bone health in the visually impaired athlete population. In contrast, the type of bone injury varied substantially in athletes with SCI. Although a smaller proportion of SCI athletes had BSI (5 of 79 athletes, 6.3%), a

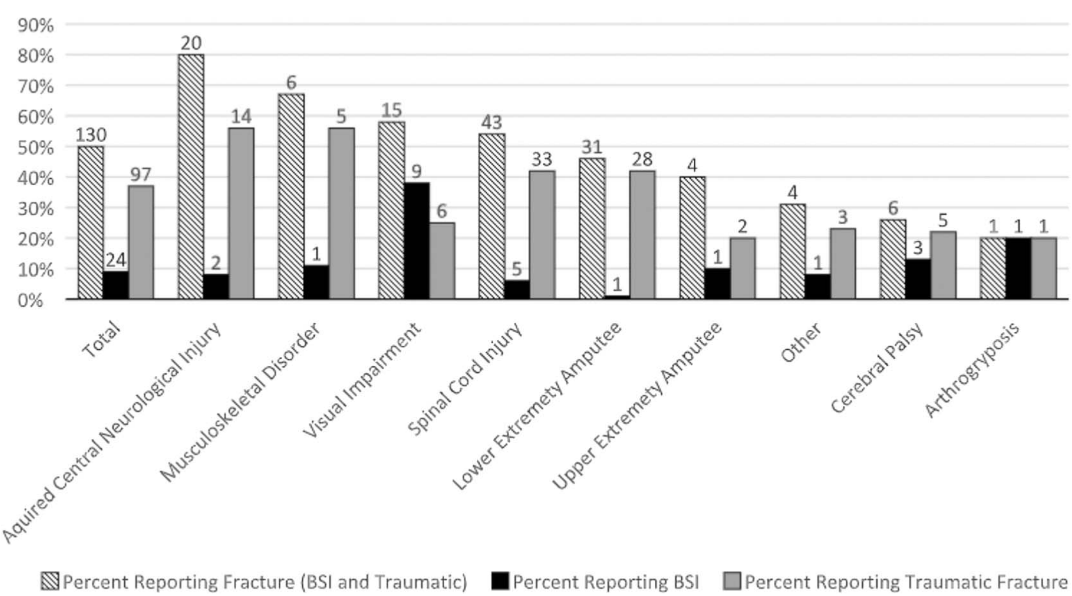

FIGURE 1. The total prevalence of bone injury, identifying percentage and number who sustained traumatic fracture and BSI within each disability category.

TABLE 2. Para athletes characteristics for individuals with BSI

Primary Disability	Sex	Sport Type	Primary Mode of Mobility	Location
Visual impairment	M	Athletics	Ambulatory with no assistive device	Ankle
Visual impairment	M	Athletics	Ambulatory with no assistive device	Pelvis
Visual impairment	F	Goalball	Ambulatory with no assistive device	Arm
Visual impairment	M	Track and field	Ambulatory with no assistive device	Tibia
Visual impairment	M	Cycling	Ambulatory with no assistive device	Ribs
Visual impairment	M	Cycling	Ambulatory with no assistive device	Right arm
Visual impairment	F	Swimming	Ambulatory with crutches, cane, or walker	Wrist
Visual impairment	M	Athletics	Ambulatory with no assistive device	Shins
Visual impairment	M	Triathlon	Ambulatory with no assistive device	Fifth metatarsal
Visual impairment	M	Triathlon	Ambulatory with no assistive device	Third metatarsal
Visual impairment	F	Track and field	Ambulatory with no assistive device	Inner sesamoids foot
Visual impairment	F	Track and field	Ambulatory with no assistive device	Pubic symphysis
Visual impairment	M	Goalball	Ambulatory with crutches, cane, or walker	Left ankle
SCI	F	Track and field	Manual wheelchair use	Femur
SCI	F	Swimming	Ambulatory with no assistive device	Metatarsal
SCI	F	Track and field	Manual wheelchair use	Spine
SCI	F	Track and field	Manual wheelchair use	Spine
SCI	F	Track and field	Manual wheelchair use	Wrist
SCI	M	Wheelchair basketball	Manual wheelchair use	Hand
SCI	M	Wheelchair basketball	Manual wheelchair use	Hand
Cerebral palsy	F	Wheelchair fencing	Ambulatory with orthotics or braces	Knee
Cerebral palsy	F	Wheelchair fencing	Ambulatory with orthotics or braces	Tibia
Cerebral palsy	F	Track and field	Ambulatory with orthotics or braces	Left thumb
Cerebral palsy	M	Soccer (Football) 7-a-side	Ambulatory with no assistive device	Metatarsal
Cerebral palsy	M	Soccer (football) 7-a-side	Ambulatory with no assistive device	Metatarsal
Neurological injury	M	Soccer (football) 7-a-side	Ambulatory with no assistive device	Ankle bone
Neurological injury	F	Track and field	Ambulatory with orthotics or braces	Second metatarsal
Neurological injury	F	Track and field	Ambulatory with orthotics or braces	Third metatarsal
Arthrogryposis	M	Track and field	Manual wheelchair use	Wrist
Arthrogryposis	M	Track and field	Manual wheelchair use	Wrist
Other	F	Rowing	Manual wheelchair use	Ribs
Other	F	Rowing	Manual wheelchair use	Ribs
Upper limb amputee	F	Swimming	Ambulatory with no assistive device	Fifth metatarsal
Lower limb amputee	F	Cycling	Ambulatory with prosthesis	Tibia
Musculoskeletal disorder	M	Rowing	Ambulatory with no assistive device	Rib

majority had history of traumatic fracture. Caution should be taken interpreting these findings given small sample size; however, it is likely that disability type and form of athletic competition may each contribute to risk for injury. Athletes with SCI (and therefore disuse osteoporosis of the lower limbs) may be at higher risk for traumatic fracture, whereas para athletes with visual impairment who maintain weight bearing status are at higher risk for overuse BSI. A majority of all para athletes reported history of previous fracture, suggesting para athletes require further investigations to understand methods to modulate risk factors for injury.

The most common sport type for para athletes with BSI was track and field. This is interesting given that the sport of track and field had highest number of athletes with this form of injury at the 2016 Summer Olympics.[3] Further studies with larger populations of adaptive sport athletes may help clarify which sports are most strongly associated to this form of injury.

In summary, the current report highlights that BSI should be on the differential diagnosis for a para athlete who presents with injury and bone pain, including pain in either the upper or lower limb for athletes of both sexes. Similar to reports in able-bodied athletes, lower limb injuries may be more frequent in athletes and those who participate in the sport of track and field. Concerns for impaired bone quality should prompt efforts to promote methods to optimize bone health by ensuring adequate EA and other nutritional requirements are met, including calcium and vitamin D. Screening criteria for obtaining a DXA and completing a workup for bone health have not been published for the para athlete. We would suggest screening DXA for any athlete who sustains a BSI, with a focus on identifying athletes requiring further intervention to optimize skeletal health and reducing risk for future injury.

REFERENCES

1. Wentz L, Liu PY, Haymes E, et al: Females have a greater incidence of stress fractures than males in both military and athletic populations: a systemic review. *Mil Med* 2011;176:420–30
2. Warden SJ, Davis IS, Fredericson M: Management and prevention of bone stress injuries in long-distance runners. *J Ortho Sports Physio Ther* 2014;44:749–65

3. Hayashi D, Jarraya M, Engebretsen L, et al: Epidemiology of imaging-detected bone stress injuries in athletes participating in the Rio de Janeiro 2016 Summer Olympics. *Br J Sports Med* 2018;52:470–4
4. De Souza MJ, Nattiv A, Joy E, et al: 2014 Female Athlete Triad Coalition consensus statement on treatment and return to play of the female athlete triad: 1st International Conference held in San Francisco, CA, May 2012, and 2nd International Conference held in Indianapolis, IN, May 2013. *Clin J Sport Med* 2014;24:96–119
5. Blauwet CA, Brook EM, Tenforde AS, et al: Low energy availability, menstrual dysfunction, and low bone mineral density in individuals with a disability: implications for the para athlete population. *Sports Med* 2017;47:1697–708
6. Brook EM, Tenforde AS, Broad EM, et al: Low energy availability, menstrual dysfunction, and impaired bone health: a survey of elite para athletes. *Scand J Med Sci Sports* 2019;29:678–85
7. Mountjoy M, Sundgot-Borgen J, Burke L, et al: The IOC consensus statement: beyond the female athlete triad—Relative Energy Deficiency in Sport (RED-S). *Br J Sports Med* 2014;48:491–7
8. Mountjoy M, Sundgot-Borgen JK, Burke LM, et al: IOC consensus statement on relative energy deficiency in sport (RED-S): 2018 update. *Br J Sports Med* 2018;52:687–97
9. Gold JR, Gold MM: Access for all: the rise of the Paralympic Games. *J R Soc Promot Health* 2007;127:133–41
10. Krempien JL, Barr SI: Eating attitudes and behaviours in elite Canadian athletes with a spinal cord injury. *Eat Behav* 2012;13:36–41
11. Krempien JL, Barr SI: Risk of nutrient inadequacies in elite Canadian athletes with spinal cord injury. *Int J Sport Nutr Exerc Metab* 2011;21:417–25
12. Miyahara K, Wang DH, Mori K, et al: Effect of sports activity on bone mineral density in wheelchair athletes. *J Bone Miner Metab* 2008;26:101–6
13. Goktepe AS, Yilmaz B, Alaca R, et al: Bone density loss after spinal cord injury: elite paraplegic basketball players vs. paraplegic sedentary persons. *Am J Phys Med Rehabil* 2004;83:279–83
14. Harris PA, Taylor R, Thielke R, et al: Research electronic data capture (REDCap)—a metadata-driven methodology and workflow process for providing translational research informatics support. *J Biomed Inform* 2009;42:377–81
15. Torstveit MK, Sundgot-Borgen J: Participation in leanness sports but not training volume is associated with menstrual dysfunction: a national survey of 1276 elite athletes and controls. *Br J Sports Med* 2005;39:141–7
16. Barrack MT, Rauh MJ, Barkai HS, et al: Dietary restraint and low bone mass in female adolescent endurance runners. *Am J Clin Nutr* 2008;87:36–43
17. Barrack MT, Gibbs JC, De Souza MJ, et al: Higher incidence of bone stress injuries with increasing female athlete triad-related risk factors: a prospective multisite study of exercising girls and women. *Am J Sports Med* 2014;42:949–58
18. Yeager KK, Agostini R, Nattiv A, et al: The female athlete triad: disordered eating, amenorrhea, osteoporosis. *Med Sci Sports Exerc* 1993;25:775–7