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REVIEW ARTICLE

Paget-Schroetter syndrome in athletes: a comprehensive and systematic review

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Background: Paget-Schroetter syndrome (PSS) is a rare condition of axillosubclavian vein thrombosis often seen in athletes with a history of repetitive external rotation and abduction of the shoulder. The purpose of this review was to analyze the literature and characterize PSS in the athletic population, including risk of PSS by sport. We also provide a comprehensive review of PSS to inform clinicians on the pathophysiology, detection, and management of the condition.
Methods: Four databases were reviewed to identify cases of PSS occurring in athletes. Data on patient demographics, reported sport, diagnosis, treatment, management, return to sport, and complications were extracted and analyzed by 2 independent reviewers.
Results: Of the 123 cases of PSS identified, baseball and weight lifting had the highest incidence (26.8% and 19%, respectively), followed by swimming, football, and basketball. The average return to sport was 4.7 months, and 26.7% of subjects reported complications, most commonly pulmonary embolism.
Conclusion: In athletes presenting with upper extremity pain and swelling with a history of playing baseball or weight lifting, PSS should be higher on a clinicians differential diagnosis list. Swimmers, football, and basketball players are less likely to present with PSS but are still more likely than other types of athletes to develop the condition. Clinician awareness of PSS in athletes is critical to avoid delays in treatment and misdiagnosis, and to allow for a timely return to sport with minimal complications.
Level of evidence: Level IV; Systematic Review
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Keywords: Paget-Schroetter; subclavian vein; effort thrombosis; exercise; athlete; blood clot

Paget-Schroetter syndrome (PSS), also known as "effort thrombosis," was first described by Sir James Paget in 1875.[30] PSS is defined as axillosubclavian vein thrombosis that is most commonly seen in athletes or individuals with a history of significant repetitive overhead motion, specifically shoulder abduction and external rotation.[15,20,30] It is a rare phenomenon, with incidence of approximately 1-2 per 100,000 cases (1%-4%) of venous thrombotic events (VTE) reported annually in the United States.[30] However, between 10% and 20% of upper extremity VTE are said to be attributable to PSS.[15] It is suggested that external compression of the subclavian vein within the thoracic outlet combined with repetitive overhead motion results in fibrous deposition, scar formation, and subsequent intimal hyperplasia and endothelial injury,[15,20,30] a key component of Virchow's triad, ultimately leading to venous thrombosis. PSS is thought to typically occur in younger, active patients without a significant history of thrombotic risk factors, including genetic deficiencies of proteins C and S, antithrombin III, or acquired factors such as smoking and

Institutional review board approval was not required for this systematic review.

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obesity; however, this remains of active debate in the literature.

Anatomically, the thoracic outlet is defined by the clavicle (superior border), first rib (inferior border), costoclavicular ligament (medial border), and anterior scalene muscle (posterolateral border).[20] Structures that travel through the thoracic outlet include the subclavian vein that courses between the clavicle and first rib, the subclavian artery, and the lower trunk of the brachial plexus.[15] Thoracic outlet syndrome occurs when at least one of these structures is symptomatically compressed. If the subclavian vein is compressed, a significantly higher risk of thrombosis is created.[30] Bony abnormalities such as a history of clavicle fracture or cervical rib, more lateral insertions of the costoclavicular ligament, or muscular hypertrophy of the subclavius, anterior scalene, or even pectoralis minor have been reported as findings in patients who develop PSS.[15,20] These abnormalities are thought to be the source of venous compression, and when they are exacerbated by repetitive overhead motion, endothelial injury can occur, increasing the risk of venous thrombosis. This endothelial injury then activates the coagulation cascade, creating a thrombogenic environment inside the vessel that predisposes to clot formation.[15] Intraoperatively, studies have observed that the loose connective tissue normally surrounding the vessels is replaced by a dense collagen scar, resulting in a less mobile vein that can be damaged easily when compressed or stretched.[30] It is thought that this scar formation is the result of repeated extrinsic insult to vessels and may further limit the vessel's capability for laminar flow.

Most cases of PSS (60%-85%) present within 24 hours of vigorous physical activity with acute pain, swelling, discoloration, ''heaviness'' of the affected limb, and sometimes Urschel's sign, which is the presence of dilated superficial collateral veins found over the upper arm, anterior shoulder, base of the neck, and upper chest.[30] A comprehensive physical examination including visualization and palpation of the upper extremity, spine, chest, shoulder girdle, and supraclavicular fossa should be conducted, along with auscultation of the supraclavicular fossa, which may reveal a bruit indicative of obstruction.[15] Provocative maneuver tests for thoracic outlet syndrome are not routinely recommended in diagnosing PSS, as they have a high false-positive rate for detecting vascular compression and are more likely to detect nerve compression.[15] Duplex ultrasound is recommended by the American College of Radiology as the gold standard and best initial test for diagnosis, with a sensitivity of 78%-100% and specificity of 82%-100%.[15,30] It is also important to conduct a thorough history inquiry about the presence of any thrombophilic risk factors and to keep other thrombophilic states high on the list of differential diagnoses. There is a lack of prospective randomized control trials concerning the treatment of PSS, but there is agreement in the literature that the most effective treatment includes immediate anticoagulation and thrombolysis for PSS alone and subsequent surgical decompression via first rib resection within 14 days for PSS with evidence of extrinsic compression. After 14 days, treatment is less likely to be successful and affected veins are more likely to remain occluded, possibly requiring venous reconstruction.[20,30]

The complications of PSS can lead to considerable morbidity and mortality. Re-thrombosis occurs in one-third of patients without timely treatment and post-thrombotic syndrome (PTS) is a potential complication of all VTE that presents in 7%-46% of patients with upper extremity deep vein thrombosis (DVT).[20] PTS is a chronic and potentially debilitating condition seen after VTE that is characterized by pain, heaviness, and swelling of the affected limb.[3] Pulmonary embolism (PE), which can be fatal without treatment, has also been reported to occur in 20%-30% of patients presenting with PSS.[20] Considering the serious impact of these complications, it is paramount that the diagnosis of PSS be made quickly and accurately, demonstrating the need for a high index of suspicion for active patients who present with unexplained upper extremity symptoms. With rapid diagnosis and proper management, most studies report upward of 90%-95% success rates with treatment of PSS and nearly 100% of athletes return to sport within 1 year.[15,20]

Although PSS is relatively rare, it can be extremely detrimental to quality of life if not managed promptly. PSS primarily affects athletes, yet there is a considerable lack of research focusing strictly on PSS in this population, as most studies report combined occupational risks and focus on other populations. There is a general consensus in the literature that sports requiring repetitive overhead motions place athletes at a higher risk for developing PSS, but the incidence of PSS in each sport individually has not been extensively studied.

The aim of this systematic review is to examine the literature to determine which sports pose the most significant chance for development of PSS. We plan to report the incidence of PSS found in each type of sport in the literature and increase clinician awareness of PSS to expedite prompt recognition of the condition when caring for athletes, to avoid complications associated with misdiagnosis.

Methods

Two independent reviewers performed a literature search of 4 databases: PubMed, Cumulative Index of Nursing and Allied Literature (CINAHL), Cochrane, and EMBASE. Use of the search terms ''Paget Schroetter Syndrome,'' ''Effort Thrombosis in Athletes,'' and '''Upper Extremity Deep Vein Thrombosis' AND 'Athletes''' yielded a total of 2160 articles. Duplicate results were removed, and the remaining articles were screened by each reviewer for relevance by title, abstract, and then full text. Discordant items were handled by a third

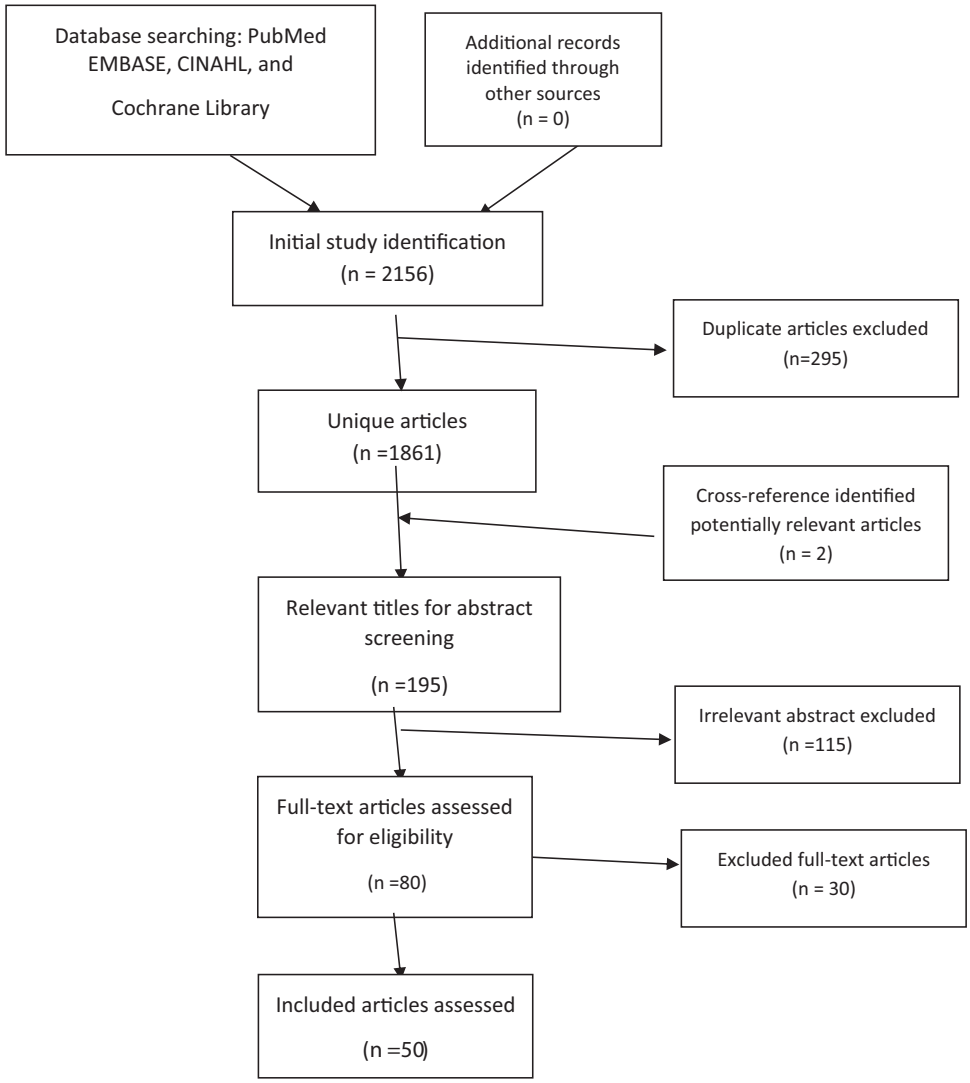

Figure 1 PRISMA flow chart. Overview of literature search and article inclusion process according to PRISMA guidelines. *PRISMA*, Preferred Reporting Items for Systematic Reviews and Meta-Analyses.

independent reviewer who made the final decision for inclusion. Full text articles written in English, Spanish, French, or Portuguese and published between 1960 and 2019 were included. All types of articles were eligible for inclusion if they specified at least 1 case of an Olympic sport–related cause of PSS, as recognized by the International Olympic Committee. In addition, American football was included because of its popularity and prevalence as a sport that requires repetitive overhand throwing motions. This review was registered in PROSPERO and followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines throughout the research process.

Data extraction was performed in duplicate by the 2 reviewers independently, and accuracy was crosschecked by the third reviewer. Data were stored in organized spreadsheets accessible to all members of the research team, who individually approved each step of this review. Information extracted included patient age, sex, hand dominance, location of thrombus, sport played, any sport-specific details (position, level of play, changes in training), past medical history, treatment course, complications, outcomes, and return to sport (RTS). A proportional incidence ratio was calculated by determining the total number of cases of PSS and dividing the number of occurrences in each sport by that total number.

Results

The database search yielded 2160 articles that were assessed for eligibility (Fig. 1). We identified 50 reports[2-4,6,8-14,16-19,21-25,27-29,31-34,37,39-47,49-61] consisting of 123 Olympic sport–related cases of PSS. The average age of patients in our review was 23.99 ±7.87 years, with a range of 16-56 years. The majority of individuals (80.5%) were male, and 55.3% of cases occurred in the dominant arm of the athlete. The most common site for thrombus formation was the subclavian vein (53.7%), followed by both axillary and subclavian (29.3%), and then the axillary

vein alone (11.4%). The average time from symptom onset to diagnosis was 13.4 days. Past medical history was reported in 46.3% of articles reviewed, and included tobacco use in 3.2% of patients,[44,56,59,61] oral contraceptive use in 3.2%,[3,21,32,61] family history of thrombosis in 2.4%,[18,61] and prior history of thoracic outlet syndrome in 1.6%.[10,58] Athletes had no relevant past medical history in 32.5% of cases (Table I).

Of the 123 cases of PSS identified in the literature, the most common sports reported were baseball[2,4,6,13,18,23,27,37,58,59] (26.8%), weight lifting[10,16,19,21,29,31,33,40,41,44-46,54-56,60,61] (19.5%), and swimming[14,37,39,53,61] (13%). American football was associated with the development of thrombosis in 9.8%[6,9,22,37,50-52] of cases, basketball in 5.7%,[16,37,61] and volleyball in 3.3%.[6,8,25,37] The remaining sports identified made up less than 3% of cases (Fig. 2). Of the 33 cases associated with baseball, 70% of the athletes were pitchers.[2,13,23,58,59] Level of play was reported for 19 of the baseball players: 3 were high school athletes,[37] 7 played at the collegiate level,[13,27,37,58] and 9 were listed as either "professional" players or were participating in Major League Baseball.[13,37,39]

Twenty articles[2,4,6,9,14,16-18,21,22,25,27,37,39,42,44,50-52,59] reported on RTS for 72 athletes. The average RTS for this cohort was 4.78 ± 3.11 months (Fig. 3, *a*), and of the 4 most prevalent sports, football was associated with the longest RTS time, followed by baseball (Fig. 3, *b*). Of note, 2 cases[6,59] identified in our review did not return to sport at all. Many were reported as missing an entire season, or returning to play the following season,[13,58] but did not quantify the number of months so were unable to be included in our analysis. The remaining articles did not comment on RTS.

Complications associated with PSS occurred in 26.7% of the cases identified. A total of 34.1% of cases reported no complications, whereas the remaining 41.5% of studies did not specify whether or not a complication occurred. Pulmonary embolism was the most frequent complication reported, with an incidence of 8.1%.[4,13,19,21,23,24,27,31,49,59] Re-thrombosis occurred in 5.7% of cases,[2,9,16,27,37,40,51,61] neuropathy in 3.3%,[3,16,39] and PTS was reported in 1.6%.[21,32] "Persistent pain and discomfort" were also listed as complications in 1.6%[51,61] (Table I). Surgical complications such as neuropathy (3.3%), surgical site hematoma (1.6%), hemothorax (0.8%), and hemoperitoneum (0.8%) were also reported in those cases undergoing operative management.

Treatment in the included studies consisted of anticoagulation, balloon angioplasty or catheter-directed thrombolysis, and surgical decompression in most cases. The vast majority of cases identified by our review were treated operatively (77.2%). First rib resection together with catheter-directed thrombolysis and pharmacologic anticoagulation was performed in 60.1% of cases, whereas 5.7% of cases were treated with balloon angioplasty or catheter-directed thrombolysis and pharmacologic anticoagulation, without first rib resection.[11,14,24,34,43,45,56] An additional 17% of athletes underwent surgical treatment, but the exact procedure was not specified in the article.[53] Conservative treatment with pharmacologic anticoagulation alone was reported in 13% of cases.[12,19,21,23,25,29,32,41,42,47,49,50,52,55,57,59] Treatment mechanism was unspecified in 4.1% of cases.[33,61]

Table I Athlete demographics: description of included case series

	Years (± SD)
Age	23.99 ± 7.87
Sex, %	
Male	80.5
Female	11.4
Location of thrombus, %	
Subclavian vein	53.7
Axillary and subclavian veins	29.3
Axillary vein	11.4
Axillary, subclavian, and brachial veins	2.4
Axillary, subclavian, brachial, cephalic, and basilic veins	1.6
Axillary, subclavian, and basilar veins	0.8
Subclavian and brachiocephalic veins	0.8
Dominant arm, %	
Yes	55.3
No	10.6
Not specified	34.1
PMH, %	
None	32.5 (40)
Tobacco use	3.2 (4)
OCP use	3.2 (4)
Family hx of thrombosis	2.4 (2)
Hx of thoracic outlet syndrome	1.6 (2)
Hx of shoulder dislocation	1.6 (2)
Hx of clavicle fracture	0.8
Hx of DVT	0.8
Anticardiolipin antibody	0.8
Factor V Leiden deficiency	0.8
Not specified	53.7 (66)
Complications, %	
None	34.1 (42)
PE	8.1 (10)
Re-thrombosis	5.7 (7)
Neuropathy	3.3 (4)
Persistent pain	1.6 (2)
Hematoma	1.6 (2)
Hemothorax	1.6 (2)
Post-thrombotic syndrome	1.6 (2)
Pneumothorax	0.8 (1)
Hemoperitoneum	0.8 (1)
Pleural effusion	0.8 (1)
Depression	0.8 (1)
Not specified	41.5 (51)

PMH, past medical history; *OCP*, oral contraceptive; *Hx*, history; *DVT*, deep vein thrombosis; *PE*, pulmonary embolism; *SD*, standard deviation.

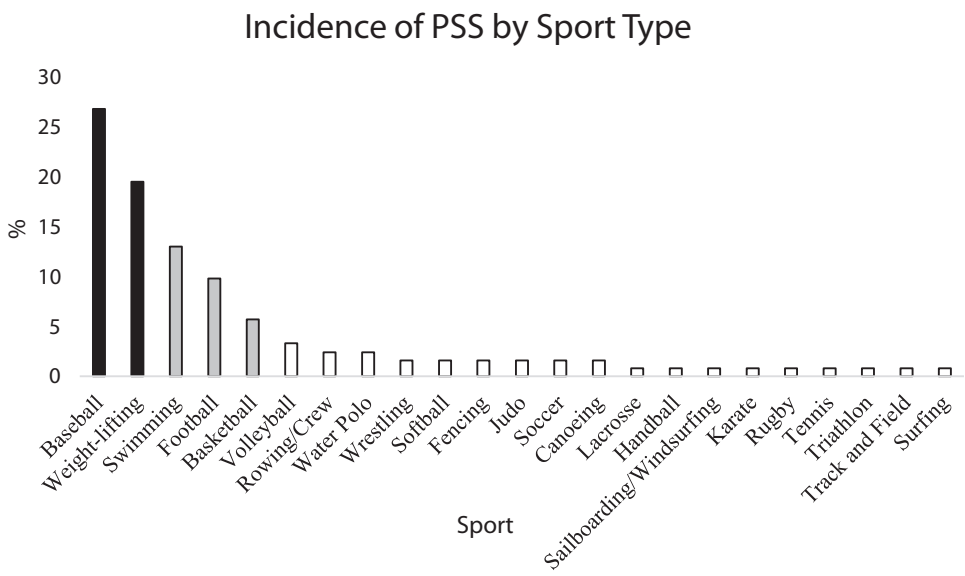

Figure 2 Incidence of PSS by sport type. Baseball was reported in the highest number of PSS cases, followed by weight lifting, swimming, football, and basketball. All other sports were reported in less than 5% of cases. *PSS*, Paget-Schroetter syndrome.

Discussion

PSS is a rare phenomenon of axillosubclavian vein thrombosis mostly seen in athletes and widely thought to be associated with extrinsic compression of the axillosubclavian venous region within the thoracic outlet. Repetitive shoulder abduction and external rotation is considered to be the most important risk factor for the development of this condition, and some sports require this positioning more than others. Our literature review yielded several studies that suggest that some sports are more likely to be associated with PSS,[20,30,38] but none had actual published data on the incidence of PSS by sport. The purpose of this review was to examine the incidence of PSS in each sport and provide a comprehensive review of PSS to inform clinicians on the details of this diagnosis.

Of the cases identified in the literature, baseball and weight lifting were the most common sports associated with the development of PSS. With incidences of 26.8% and 19.5%, respectively, these are still relatively low percentages, but were the highest of any sport reported. Swimming (13%), American football (9.8%), and basketball (5.7) were the next most common sports, and their overall incidence is low as well. PSS is a rare condition, only representing 1%-4% of VTE in the United States each year, and is often misdiagnosed as clinicians do not suspect a diagnosis of thrombosis in young, healthy, athletic populations. When an athlete who participates in sports such as baseball or weight lifting presents with pain, swelling, heaviness, or erythema of the arm, it is imperative that PSS be placed on the differential list by the clinician. It should be noted that all of the sports identified in this review require some degree of repetitive overhead motion, and each was identified as the cause of at least 1 case of PSS, putting them at a higher risk at baseline compared with sports that do not require any overhead motion. These ratios show baseball as the most common sport associated with PSS at 26.8%; interestingly, this is lower than some prior studies. A 2008 review showed that 14 of 32 (43.8%) athletic cases off PSS were associated with baseball.[37] This study included results over a period of several years at 1 institution, rather than all available literature, and may represent an overestimation of the true incidence in baseball due to region-unique involvement in different sports, as well as the setting of the institution itself.

Slight increases in rates of PSS were found with age and level of play, especially in baseball. Three cases of PSS were identified in high school baseball pitchers, 7 cases in college, and 9 at the professional level. This increase may be attributable to the increased years of use and accumulation of external compression in the thoracic outlet. Surgical examination of PSS patients has demonstrated both internal and external venous fibrosis, generally thought to be a result of chronic compression.[30] Older patients or higher-level athletes who train more frequently are thus expected to show more significant fibrosis than their younger counterparts. Roughly 80.5% of cases were in males, which may be related to disproportionate gender involvement in studied sports. The majority of cases (55.3%) also occurred in the dominant arm of the athlete, especially in throwing sports. The dominant or throwing arm is placed into the vulnerable abduction and external rotation position much more frequently than the nondominant arm in sports that require the throwing motion (baseball, football, softball), resulting in higher amounts of external compression compared with the nondominant arm. Although it is interesting that only 55% of PSS cases

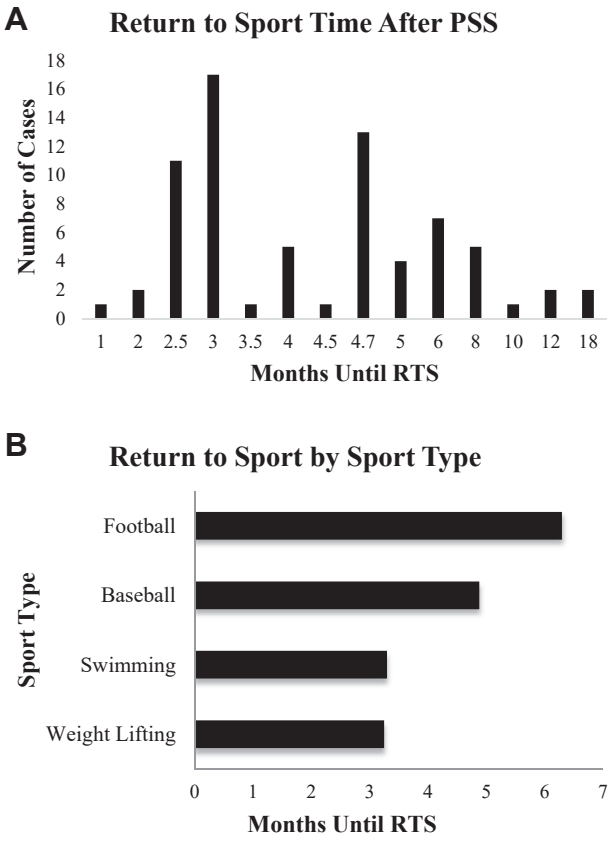

Figure 3 Return to sport after PSS. (**A**) The average RTS for an athlete diagnosed with PSS occurred at 4.78 ± 3.11 months, with majority of cases returning at 3 months. The minimum return to sport was 1 month, and 2 athletes did not return to sport at all. (**B**) Time to RTS stratified by sport type for the most prevalent sports. Football players returned at an average of 6.33 months, with baseball at 4.88 months, swimming at 3.3 months, and weight lifting at 3.25 months. *PSS*, Paget-Schroetter syndrome; *RTS*, return to sport.

occurred on the dominant arm, this is likely due to a reporting bias. Many (34.1%) of the studies analyzed did not specify whether or not the dominant arm was affected.[10,11,14,19,22,24,25,28,29,31,33,34,41,42,44,45,47,49,50,53-57,61] It is plausible that in sports that have a clear dominant arm such as baseball or football, the authors found it relevant to report which side the thrombus occurred on; however, in sports that require equal, bilateral use of each arm (swimming, weight lifting), the hand dominance of the athlete may not have seemed as important to report.

The average time from symptom onset until the diagnosis of PSS was 13.4 days. Prior studies have suggested that 14 days are considered the cutoff for optimized outcomes.[20] Of note, 15 cases[2,8,9,12,19,21,24,27,31,32,39,44,55] were initially misdiagnosed as conditions other than PSS, such as muscle strains, tendinitis, tendon ruptures, or cellulitis. It seems logical to suggest that athletes may have returned to sport more quickly or experienced fewer complications if their misdiagnosis had been avoided. It is worth mentioning that 54.5% of studies did not mention the time interval between symptom onset to diagnosis.

Once the diagnosis of upper extremity DVT has been made, it is critical to determine the correct etiology of each patient's thrombophilia, as hereditary and acquired thrombophilia can coexist and may affect management. The most common causes of thrombophilia reported with effort thrombosis include Factor V Leiden, Protein C/S deficiency, antithrombin III deficiency, plasminogen activator inhibitor 1, and anticardiolipin antibodies. Current studies have varying estimates on the prevalence of concomitant hypercoagulability in patients with effort thrombosis. Some suggest that as many as 67% of patients had concomitant hypercoagulability, with 90% of postoperative complications attributable to thrombophilia.[5] Other studies have demonstrated the prevalence as low as 25%, with no statistically significant difference in complications between patients affected by associated thrombophilia, provided they received lifelong anticoagulation.[36] In our review, only 2.44% of athletes reported a personal history of DVT or hypercoagulable state; however, considering that over half of the included studies did not mention past medical history, the true incidence might be higher. Patients with spontaneous upper extremity should be assessed for these common causes of thrombophilia to minimize their risk of postoperative complications and long-term thrombosis.

The treatment of PSS generally involves thrombolysis and subsequent surgical thoracic outlet decompression via first rib resection,[1] as seen in 60.1% of the studies included in this review. Removal of the first rib negates the typical source of external compression, as the inferior border of the thoracic outlet is no longer present. Without this restrictive border, the vasculature remains decompressed even with nearby muscle hypertrophy. Surgical decompression with first rib resection and anterior scalenectomy in combination with pharmacologic anticoagulation is typically considered to be the most effective treatment for PSS, but the procedure does not come without risks. Complications such as pneumothorax, bleeding, and arterial or nerve injuries acquired during surgery must be taken into consideration when determining a treatment plan for these athletes. In our review, most articles did not specify whether complications occurred as a result of the surgery or the diagnosis itself, but 4 cases of neuropathy, 2 hematoma's, 2 hemothorax's, and 1 case of hemoperitoneum and pleural effusion were reported and can be presumed to be surgical.

Pharmacologic treatments for PSS include anticoagulation, systemic fibrinolysis, and catheter-directed thrombolysis. Although most patients in our review were managed surgically (77.2%), 13% were treated conservatively with anticoagulation alone. Studies have shown that the treatment of PSS with anticoagulation alone leads to a

higher incidence of residual symptoms, disability, and recurrent thrombosis.[1,30] However, first rib resection is a surgical procedure with risks of its own, and it is unclear if the reason for conservative management was based on patient preference or medical indication.

Systemic fibrinolysis has been shown to be more efficacious than anticoagulation alone at achieving venous patency; however, complications such as intracranial hemorrhage are of concern.[1] Conversely, catheter-directed thrombolysis with alteplase or reteplase is more frequently used due to its local fibrinolytic effects with minimal systemic bleeding risk.[1] Some studies suggest that catheter directed thrombolysis (CDT) is most therapeutic when used for clots formed within the last 14 hours.[1,30] Strong, stable fibrin clots that are relatively resistant to proteolytic cleavage are formed when fibrin covalently links with activated Factor XIII, a likely explanation for the proposed decrease in efficacy of thrombolysis after 14 hours.

A feared complication of PSS is PE, as it can be fatal or otherwise detrimental to a patient's quality of life. The aggregate incidence of PE in the analyzed studies was 8.1%, much less than the rate of upper extremity pulmonary thromboembolism reported at 20%-30% by Hangge et al.[20] There are no data to suggest that pulmonary embolism occurred at a different rate or via a different mechanism in PSS than in other cases of upper extremity DVT. PTS, another feared complication of PSS, only occurred in 2 cases that we reviewed but "residual pain and discomfort," 2 major symptoms of PTS, were listed as complications in 2 additional cases.[51,61] Surprisingly, complications were not specified in 41.5% of analyzed studies, making it challenging to provide a comprehensive statement regarding other complications that may have occurred but were not reported.

RTS is a significant concern in high-level athletes due to its potential implications in performance, which can ultimately compromise scholarships, scouting opportunities, and income at the professional level. RTS can be influenced by a variety of factors, including the sport itself, whether the dominant limb was affected, whether treatment was invasive or not, or by other patient-specific factors. To our knowledge, there has not been a review or meta-analysis to date with pooled data on RTS after a diagnosis of PSS. The literature has reported that most athletes return to sport within 1 year of diagnosis,[6,20] and our data support that finding. One athlete returned in as little as 1 month,[25] some took up to 18 months, and 2 never returned at all.[6,59] Many reports stated the athlete was unable to complete the remainder of the season and was able to return the following season, but did not give an actual time value. Our data suggest that RTS ranges from 1 to 18 months, but more specific studies are needed to draw further conclusions on RTS after PSS, as 41.5% of the articles analyzed in this review made no mention of it.

Athletes are generally a healthy population, and because exercise is typically considered protective against VTE, it is common for clinicians to suspect other diagnoses as more likely when athletes present with upper extremity pain and edema.[48] An athlete's physical conditioning and tendency to overcompensate for pain also makes it easier to miss a diagnosis of DVT.[26,48] Although exercise is considered preventative, there are some studies that suggest that high-intensity exercise may actually increase thrombogenic risk.[47] Factors proposed to predispose athletes to thrombosis include dehydration, which can lead to hemoconcentration, trauma, and subsequent immobility as a result of injury, long duration travel to competition, and oral contraceptive use.[48] Endothelial injury can occur via physical strain or trauma, and some postulate that the bradycardia seen in athletes with a high level of cardiopulmonary fitness may even contribute to venous stasis.[26]

Some studies also explain the physiological findings attributing exercise as a thrombogenic risk factor. Lee and Lip[35] reported seeing an increase in tissue plasminogen activator (tPA) that was directly proportional to exercise intensity when they studied the difference between acute and habitual exercise habits. The authors mentioned that short-term, acute, maximal exercise increased platelet aggregation and platelet counts, and that high levels of epinephrine were found to increase platelet aggregation as well.[35] Chicharro et al[7] reported seeing these changes at exercise intensities above the anaerobic threshold. Interestingly, the greatest incidence of PSS in the studies included in our review was found amongst pitchers, who typically are not considered to be beyond anaerobic threshold while pitching. Although these data support the notion that exercise may contribute to hypercoagulability metabolically, we suspect that the hypercoagulability associated with PSS is more mechanical given our findings.

The limitations of this study are consistent with those intrinsic to a systematic review. Systematic reviews rely on available literature, and with the rarity of a diagnosis such as PSS, the quality of the literature was poor. Because this review included mostly case reports and case studies, it was difficult to critically appraise the quality of the studies with appropriate tools. As with most, this review is subject to publication bias, as the only cases included were those that were published in the 4 databases searched. There are likely more cases of PSS related to sports that have occurred but have not been published in the literature at this time, particularly given the high rate of misdiagnosis of PSS. However, our selection bias was minimal as we included every available report that specified an Olympic sport–related cause of PSS. We did not exclude based on age, sex, or level of play, and our search included articles published in 4 languages in 4 large databases, from inception to present.

Conclusion

Athletes presenting with upper extremity pain and swelling with a history of playing baseball or weight

lifting should be considered high risk for the development of PSS. A history of participating in swimming, American football, and basketball should also trigger clinician thoughts of a diagnosis of PSS. Clinician awareness of PSS in athletes is critical to avoid delays in treatment and misdiagnosis, and to allow for a timely RTS with minimal complications.

Disclaimer

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