

A systematic review of prevalence and risk factors associated with playing-related musculoskeletal disorders in pianists

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Background	Playing-related musculoskeletal disorders (PRMDs) are a recognized problem amongst instrumental musicians. Although pianists are prominent in data regarding prevalence of PRMDs, there is poor understanding of piano-specific risk factors associated with PRMDs.
Aim	To synthesize published literature on the prevalence of and risk factors associated with PRMDs in pianists.
Methods	Thirty-eight databases were searched. Eligible studies were those investigating prevalence of or risk factors associated with PRMDs in pianists, using an appropriate methodology according to a hierarchy of evidence. Information regarding study population, operational definition of PRMD, risk factors investigated, statistical tests used and outcomes was extracted and narratively synthesized for all eligible papers above an arbitrarily chosen quality score.
Results	The literature search identified 482 citations. Fifty-two papers were ranked in a hierarchy of evidence; 12 were eligible for evaluation using a quality assessment tool. Common methodological limitations included sampling/measurement biases, inadequate reporting of reliability/validity of outcome measures, lack of operational definition of PRMD and no statistical significance testing. Prevalence rates for PRMDs in pianists varied widely (26–93%). Four authors demonstrated statistically significant risk factors; however, no authors combined a clear operational definition of PRMD with statistically established risk factors. There was no consensus between authors regarding risk factors.
Conclusions	Current evidence does not provide sufficient information regarding prevalence of and risk factors associated with PRMDs in pianists. Future studies should provide an operational definition of PRMD, use valid, reliable measurement tools, utilize a prospective cohort study design and perform appropriate statistical tests.
Key words	Literature review; occupational epidemiology; occupational injury; prevalence; risk factors.

Introduction

Over 473 000 children aged 5–14 play a musical instrument in Australia [1]. The piano is one of the most popular instruments learned at all levels of music tuition; piano teachers comprise ~70% of the teachers in the largest directory of private music teachers in Australia [2]. At elite (tertiary and professional) levels of performance, playing the piano is analogous to athletic performance due to the intense level of demand and practice, emphasis on speed and accuracy and stress of competi-

tion at this level [3,4]. These high physical loads predispose elite-level pianists to musculoskeletal disorders, as reflected by research which has shown piano to be associated with high rates of upper-extremity injuries in university-level performers [5].

Numerous terms have been used to describe musicians' musculoskeletal disorders, including 'overuse syndrome' [6–8], 'repetitive strain injury' [9,10] and 'cumulative trauma disorder' [11–13]. Lack of consensus regarding terminology has led to confusion in this field [14–16]. Authors argue against the use of the above terms because they imply a specific etiology that cannot necessarily be supported scientifically [14–19]. Hence the term 'work-related musculoskeletal disorder' has been recommended [17,18]. The playing of a musical instrument has been cited as an example of 'work' in reference to

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this term [17]. As ‘playing’ is the ‘work’ of musicians, ‘playing-related musculoskeletal disorder’ (PRMD) is an appropriate music-specific derivative of work-related musculoskeletal disorder.

Qualitative research by Zaza, Charles and Muszynski [20] has derived the following operational definition of PRMD:

“...pain, weakness, lack of control, numbness, tingling, or other symptoms that interfere with your ability to play your instrument at the level you are accustomed to”

This operational definition of PRMD was validated as an outcome measure in a risk factor study of musicians [21]. The term ‘PRMD’ has been used in a previous systematic review [22]. PRMD will be used in this systematic review as an umbrella term that encompasses the terms outlined above and contained in Figure 1.

Although there are isolated historical examples of scientific investigation of PRMDs [23–25], the major growth of performing arts medicine as a speciality field has taken place in the last 25 years, with the formation of specialist networks and conferences in the early 1980s, and a peer-reviewed journal, *Medical Problems of*

Performing Artists, in 1986. Despite the establishment of specialist music medicine clinics such as the Miller Institute for Performing Artists [26] in several major cities, there is a lack of research regarding the factors that increase the likelihood of developing a PRMD.

When investigating risk factors for PRMDs, epidemiological research focusing on specific instrumental groups is necessary because the physical demands (and therefore the risks) of playing different instruments are highly variable [27]. Early research regarding injury rates and postulated risk factors associated with PRMDs has been conducted on mixed instrumental cohorts such as symphony orchestras [28] (13 out of 2212 subjects were pianists/keyboardsists), secondary school students [29] (number of pianists not stated), tertiary populations [5] (138 pianists/513 subjects) and mixed orchestra/music school populations [30] (89 pianists/658 subjects).

A common finding in these studies was that playing-related disorders were more prevalent in pianists, guitarists and string players than in woodwind players [5,31]. Because only a small percentage of the subjects in these studies were pianists, a clear picture of the prevalence of and risk factors associated with PRMDs in pianists is difficult to ascertain. Further epidemiological research regarding PRMDs that focuses on pianists is needed [32]. Specifically, there is a need to determine the prevalence of PRMDs in pianists and the risk factors that may predispose pianists to PRMDs.

An understanding of risk factors for PRMDs forms a foundation for the development of strategies designed to prevent such disorders. In a health-care climate where funding and cost savings are paramount, there is an emphasis in all areas of medicine on occupational injury prevention [33]. This priority is shared by authors in the field of performing arts medicine [34–36]. Although pianists are identified in epidemiological injury data on mixed instrumental cohorts, a systematic review of existing epidemiological literature focusing on PRMDs in pianists has not been undertaken.

An understanding of current knowledge regarding PRMDs specific to pianists could guide further primary research, aid clinical management of pianists and facilitate injury prevention strategies. Therefore the aims of this study were to synthesize published literature on the prevalence of and risk factors associated with PRMDs in pianists.

Methods

To synthesize previous research findings, a systematic review was undertaken. Systematic reviews are distinguished from narrative literature reviews by use of a focused research question, an explicit search strategy and a system to evaluate the quality of the evidence. This minimizes the potential bias associated with narrative

Search Strings:

[Instrumentalist* or Music or Musician* or Performer* or Performing Artist* or

Pianist* or Keyboard* or Piano or Piano Keyboard*]

AND

[Cumulative Trauma Disorder* or Disability or Disabilities or Musculoskeletal

Disease* or Musculoskeletal Disorder* or Occupational Disease* or Occupational

Disorder* or Overuse or Overuse Syndrome* or Pain or Repetitive Motion Disorder*

or RSI or Repetition Strain Injury or Repetition Strain Injuries or Repetitive Strain

Injury or Repetitive Strain Injuries]

OR

["Sprains and Strains"]

AND

[Causality or Cohort Studies or Cross-Sectional Studies or Epidemiology or

Epidemiologic Factor* or Follow-up Studies or Incidence or Incidence Studies or

Prevalence or Prevalence Studies or Prospective Studies or Risk* or Risk Factor* or

Survey*]

Abridged search strings (for databases that did not use medical subject headings or had a limited number of allowable search terms):

[piano* or pianist*]

AND

[pain or disorder* or injur* or musculoskeletal]

Figure 1. Search strings.

reviews, which rely more heavily on subjective evaluation [37–39].

A total of 38 medical and arts databases were searched (Table 1). Terms such as RSI and overuse syndrome were mapped to Subject Headings in MEDLINE to identify the broad range of terminology used to describe PRMDs, epidemiological terms and pianists. The resulting search terms were combined as outlined in Figure 1. A modified, shorter search combination was used for databases that did not use Subject Headings or had a limited number of allowable search terms. The major journal, *Medical Problems of Performing Artists* and two other journals (*The International Journal of Education in the Arts*, *International Journal of Arts Medicine*) were searched manually because they were not indexed in the databases searched at the time of the review. A manual search was also used for a specialized bibliography (the ‘Sailor’ bibliography), two indices of conference proceedings’ (one index of the Performing Arts Medicine Association conferences, and one index containing various other performing arts medicine conferences), nine websites and articles in the authors’ pre-existing collection. Reference lists of papers sourced were scanned to identify other relevant papers. All available years were searched for each database. Non-English-language papers were included in the review.

Two reviewers independently assessed each paper for eligibility into the study. Eligible papers were studies in which (i) the primary aim was to investigate prevalence of, or risk factors associated with, PRMDs specifically in pianists; (ii) the study utilized an appropriate epidemiological methodology (cross-sectional, case-control, cohort study) to gather information on prevalence and/or risk factors [40].

Study quality was evaluated by two methods. First, study design was evaluated by using The University of Sheffield Hierarchy of Evidence [41]. Use of a hierarchy to rank studies provides a broad indication of methodological strength [42]. The chosen hierarchy differentiates between different types of epidemiological studies, unlike other hierarchies that emphasize intervention studies such as randomized controlled trials [43,44].

Second, all eligible papers were rated using a psychometrically sound quality assessment tool, the Critical Review Form—Quantitative studies [45]. This tool assesses methodological rigor and bias within each study via dichotomous (yes/no) and descriptive items. An arbitrary quality score was obtained by allocating 1 for yes and 0 for no in 15 dichotomous elements, with higher scores reflecting higher methodological quality.

The following information was extracted from eligible papers:

- Study population (type of pianist, population size, response rate, gender, age, number of years played piano).

Table 1. Databases searched

Database type	Database name	Citations
Medical	1. Medline	60
	2. Pre-Medline	0
	3. Cinahl	6
	4. PubMed	105
	5. ISI (Institute for Scientific Information) Current Contents	15
	6. ISI Web Of Science	15
	7. Proquest 5000	4
	8. Psych Info	5
	9. Expanded Academic ASAP	6
	10. AMED (Allied and Complementary Database)	2
	11. Biological Abstracts	15
	12. Biomedcentral	1
	13. Meditext, Austhealth, Ausport	2
	14. Evidence Based Medicine Combined Databases (OVID): Cochrane, ACP journal club, DARE (Database of Abstracts of Reviews of Effectiveness)	0
	15. TRIP (Turning Research Into Practice) database	0
Arts/Music	16. ERIC (Educational Resources Information Center)	1
	17. Humanities Index	0
	18. Music Index Online	10
	19. PCI (Periodical Contents Index) Web	2
	20. RILM (Repertoire International de Literature Musicale)	40
	21. SAILOR (Bibliography based at Maryland's libraries)	93
Websites	22. AMANZ (Arts medicine Aotearoa NZ)	0
	23. BPMAT (British Performing Arts Medicine Trust)	0
	24. CNHA (Canadian Network for Health in the Performing Arts)	6
	25. CAIRSS (Computer-Assisted Information Retrieval System)	10
	26. DGfMM (German Association on Physiology of Music and Performing Arts Medicine)	0
	27. International Arts Medicine Association	0
	28. International Foundation for Performing Arts Medicine	3
	29. International Journal of Education and the Arts	0
	30. Ithaca College Performing Arts Medicine Page	9
	31. MUSICA (Music and Science Information Computer Archive)	2
	32. Musicians and Injuries website	16
	33. Performing Arts Medicine Association website	0
Other	34. Med Probl Perform Art Journal	21
	35. International Journal of Arts Medicine	7
	36. Performing Arts Medicine Association Conference Proceedings	8
	37. Conference Proceedings (other)	2
	38. Existing Journal Collection and Bibliographies of Citations	16
Total		482

- Operational definition of injury used to establish 'cases'.
- Risk factors investigated (including validity/reliability of chosen outcome measures).
- Statistical tests used to establish risk factors.
- Outcomes (prevalence/incidence, significant risk factors).

The information was synthesized, in a narrative format, for all papers with an arbitrary quality score of 7 or more out of a maximum of 15.

Results

A total of 482 citations were retrieved. Citations and papers were assessed for eligibility in several stages as outlined in Figure 2. The most frequent reasons for exclusion of papers were paper not piano specific ($n = 256$), paper primarily an intervention study ($n = 56$), paper primarily a measurement study ($n = 37$) and paper primarily an investigation of piano teaching and learning practices ($n = 32$). Two papers ranked 6 (case reports), and nine papers ranked 7 (expert opinion) on the University of Sheffield Hierarchy of Evidence [41] were eliminated because they did not utilize a recognized epidemiological study design for the establishment of risk factors [40,46]. Although they were not scored using the quality assessment tool, summary information from these papers was compared with the results of the scored papers. The 12 papers scored for methodological quality comprised 1 cohort study [47], 1 case-control study [7] and 10 cross-sectional surveys [9,32,48–55]. One of these papers, originally published in French, was translated into English [53].

Quality scores for all papers are summarized in Table 2. Papers generally scored well in the areas of clearly stating study objectives, reviewing background literature, appropriate choice of study design and consideration of clinical implications. Methodological limitations identified in a majority of papers were presence of sampling and measurement biases, no justification of sample size, lack of description of informed consent procedures,

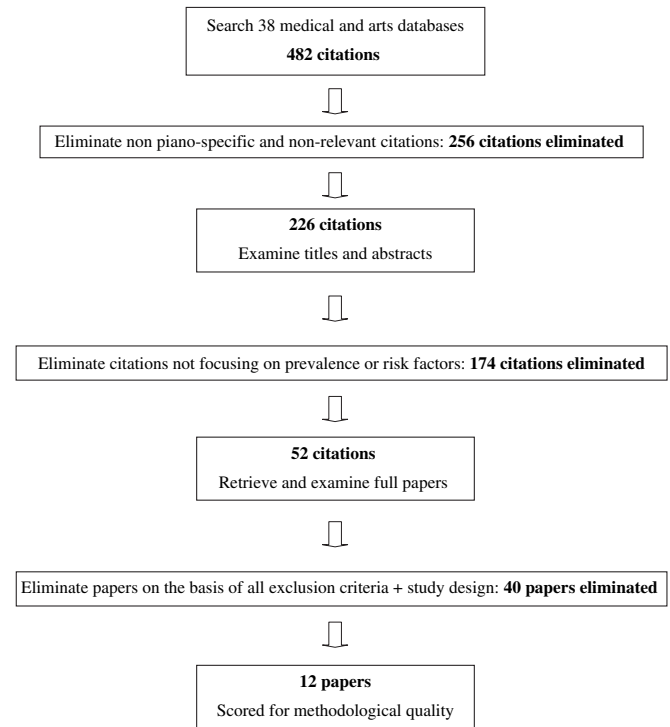


Figure 2. Selection of papers for review.

Table 2. Score summary

Reference	Hierarchy of evidence	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	Score
Pfalzer and Walker [47]	3: cohort study	1	0	1	0	0	0	0	0	0	1	1	1	1	0		7
De Smet, Ghyselen and Lysens [7]	4: c-control study	1	1	1	0	0	0	0	0	0	1	0	1	1	0		7
Farias, Ordóñez, Rosety-Rodríguez <i>et al.</i> [9]	5: c-sectional survey	1	1	0	0	0	0	0	0	0	0	1	0	1	1	0	5
Grieco, Occhipinti, Colombini <i>et al.</i> [48]	5	1	1	1	0	0	1	0	0	0	0	1	0	0	1	0	6
Blackie, Stone and Tiernan [49]	5	1	1	1	0	0	1	1	0	0	0	1	0	0	1	0	7
Pak and Chesky [32]	5	1	1	1	0	0	1	0	0	0	1	1	1	1	1	1	10
Revak [50]	5	1	1	1	0	0	0	0	0	0	0	1	0	1	1	1	7
Shields and Dockrell [51]	5	1	1	1	0	0	1	1	0	0	1	1	1	1	1	1	11
Yee, Harburn and Kramer [52]	5	1	1	1	0	0	1	1	1	1	1	1	1	1	1	1	13
Van Reeth, Chamagne, Cazalis <i>et al.</i> [53]	5	1	1	1	0	0	1	0	0	0	0	1	0	0	1	1	7
Sakai [54]	5	1	0	1	0	0	0	0	0	0	0	1	0	0	1	0	4
Sakai [55]	5	1	1	1	0	0	0	0	0	0	0	1	0	0	1	0	5
Totals		12	10	11	0	0	6	3	1	1	5	11	5	7	12	5	$X=7.4$

Key to scoring (Law *et al.* [45]): all items 1 for yes, 0 for no except biases (0 for yes, 1 for no). 1 = Was the study purpose stated clearly? 2 = Was relevant background literature reviewed? 3 = Was the design appropriate for the study question? 4 = Were there any biases present? 5 = Was sample size justified? 6 = Was the sample described in detail? 7 = Was informed consent obtained? (if not described, assume no); 8 = Were the outcome measures valid? (if all not described, assume no); 9 = Were the outcome measures reliable? (if all not described, assume no); 10 = Results were reported in terms of statistical significance? 11 = Clinical importance was reported? 12 = Were the analysis methods appropriate? 13 = Conclusions were appropriate given the study methods? 14 = Are there implications for clinical practice given the results of the study? 15 = Were limitations of the study acknowledged and described by the authors?

Table 3. Study design, population and prevalence/incidence rates

Reference	Quality score/15	Study design	Population: <i>n</i> type of pianist (RR)	Sample demographics: gender, F (female), M (male), age range and (mean); <i>n</i> (years playing) range (mean)	Operational definition of injury used to identify cases?	Results: % prevalence/incidence 'descriptor'; other results
Pfalzer and Walker [47]	7	Prospective 3-year cohort	200 professional pianists/piano teachers (61%)	Not given	No	50% incidence 'upper quadrant injuries'
De Smet, Ghyselen and Lysens [7]	7	Case-control	132 pianists (level and RR not stated)	Study: 33 F, 33 M, 18-32 (22.6) Control: 33 F, 33 M, 18-32 (24.3)	No	42.5% prevalence overuse injuries Significant difference in injury rate between study and control group; ($P < 0.001$, chi-square)
Farias, Ordonez, Rosety-Rodriguez <i>et al.</i> [9]	5	Study of anthropometric hand characteristics	341 professional/student pianists (RR not stated)	191 F, 150 M, 8-70 (no mean)	No	65.1% prevalence RSI; 151 F, 71 M
Grieco, Occhipinti, Colombini <i>et al.</i> [48]	6	Survey + EMG analysis	117 tertiary piano students (RR 'about 75%')	63 F, 54 M, 8 to >25 (no mean)	No	62% prevalence 'complaints'; 37% 'serious' according to symptom severity
Blackie, Stone and Tiernan [49]	7	Survey	16 tertiary piano students (64%)	12 F, 4 M, 18-24 (no mean); <i>n</i> (years playing) 4-20 (13.5)	No	93% prevalence 'injuries'; 62% more than one 'injury'
Pak and Chesky [32]	10	Survey (web based)	455 pianists (level and RR not stated)	243 F, 205 M, 10 to >60 (no mean)	No	58.7% prevalence pain; 29.5% 'severe' according to self-rating of pain levels (pain scale not described)
Revak [50]	7	Survey	71 tertiary/graduate pianists (18 graduate) (31%)	48 F, 23 M	No	42% prevalence (quoted as incidence)
Shields and Dockrell [51]	11	Survey	159 tertiary pianists (87%)	127 F, 32 M, 17-58 (20.5)	'Any problem caused by playing the piano which prevented piano playing for a period of 48 hours or longer'	25.8% lifetime prevalence of injury according to stated operational definition
Yee, Harburn and Kramer [52]	13	Survey + video analysis	33 tertiary pianists (RR not stated)	33 F, no age range (21.2); no <i>n</i> (years playing) range (14.3)	No (SF-36 used but scores were within normal range)	91% 'a history of musculoskeletal symptoms'
Van Reeth, Chamagne, Cazalis <i>et al.</i> [53]	7	Survey	44 pianists 'of different levels' (28 professional) (13%)	24 F, 20 M, 16-76 (37); 1-60 (25)	No	59% reported at least one symptom
Sakai [54]	4	Survey (one question asked to a series of patients)	40 tertiary and professional pianists (RR not stated)	36 F, 4 M, 16-53 (23.5)	Hand pain solely from playing-related overuse	Not applicable; (only cases were studied)
Sakai [55]	5	Survey (two questions asked to a series of patients)	200 tertiary and professional pianists (RR not stated)	165 F, 35 M, 18-66 (26.3)	Hand pain solely from playing-related overuse	Not applicable; (only cases were studied)

EMG = electromyography; RR = response rate.

Table 4. Risk factors associated with PRMDs

Reference	Quality score/15	Hypothesized risk factors	Validity/reliability of measures	Statistical tests (significance level)	Results: significant risk factors
Pfalzer and Walker [47]	7	Previous bilateral injury Previous neck injury Previous shoulder injury Previous elbow injury Previous wrist injury Previous hand injury	Not stated	Chi square Logistic regression $P < 0.05$	Previous bilateral injury ($P = 0.012$) Previous neck injury ($P = 0.001$) Previous shoulder injury (0.001) Previous elbow injury (0.04)
De Smet, Ghyselen and Lysens [7]	7	Hand size Hypermobility (mod. Beighton's) Starting age Duration and intensity of practice Playing a second or third instrument Warming up Relaxation Stretching afterwards	Not stated	Chi square Student's test $P < 0.05$	Hand size (P -value not given; based upon one of four measures of hand size; unclear whether males only or males and females)
Farias, Ordonez, Rosety-Rodriguez <i>et al.</i> [9]	5	Hand size	Not stated	Descriptive statistics	No risk factors statistically established
Grieco, Occhipinti, Colombini <i>et al.</i> [48]	6	Hand morphotype Practice time	Not stated	Descriptive statistics	No risk factors statistically established
Blackie, Stone and Tiernan [49]	7	Breaks/length of breaks EMG activity (6 subjects) Practice time	Not stated	Descriptive statistics	No risk factors statistically established ($r = 0.7906$ for correlation between time to onset of pain and duration of practice)
Pak and Chesky [32]	10	Level of injury prevention education Musician type Playing time Gender	Not stated	Correlation Pearson Chi square Linear by linear association Chi square $P < 0.05$	Age inversely associated with prevalence ($P = 0.003$) F gender (0.001) F gender for severe problem (0.025)
Revak [50]	7	Age (Playing the piano) Change in practice routine Change in technique Change in training Health factor/other activity (Unclear if based upon participant responses or asked to participants)	Not stated	Descriptive statistics	No risk factors statistically established

Shields and Dockrell [51]	11	Gender Responses to perceived risk factors analyzed: Practice time Posture Overuse Techniques, e.g. octaves (related to hand size) Fatigue Stress Other	Not stated	Chi squared	No risk factors statistically established
Yee, Harburn and Kramer [52]	13	Postural stressors (APPRI)	Test-retest reliability study on APPRI	Independent <i>t</i> -tests	UMBA and SOPA-A contributed significantly to SF-36 physical score, but combined with APPRI only accounted for 29% of the variance in the score
		Stresses of pain and discomfort (UMBA)	Other measures referenced	Pearson product-moment calculations	Number of years playing correlated with SF-36 physical score ($r = 0.45, P < 0.01$)
		Mediators of pain and discomfort (SOPA-R) Age Height Weight		Multiple linear regression Step-down regression ($P < 0.05$) Most tests compared junior (1 st year) with senior (other years) pianists, not affected with non-affected	
Sakai [54]	4	Years played piano Techniques	Not stated	Descriptive statistics	No risk factors statistically established
Sakai [55]	5	Techniques	Not stated	Descriptive statistics	No risk factors statistically established

APPRI = Adapted Postural and Repetitive Risk-factors Index; UMBA = Upper Body Musculoskeletal Assessment; SOPA-R = Survey of Pain Attitudes-Revised; SF-36 = Short-Form Health Survey.

inadequate reliability and validity (or reporting of reliability/validity) of outcome measures, lack of statistical significance testing, inappropriate analysis methods and inadequate acknowledgment of study limitations.

Table 3 summarizes information regarding study characteristics and prevalence/incidence results. Response rates for the studies were highly variable, with a range of 13–87%. Only one author provided an adequate operational definition of PRMD for identifying cases. Shields and Dockrell [51] defined a PRMD as ‘any problem caused by playing the piano which prevented piano playing for a period of 48 hours or longer’. Two other authors provided an operational definition of PRMD, but in both cases the descriptions given were non-specific or ill-defined, and were applied *post hoc* to assess the severity of symptoms rather than being used to identify cases [48,50]. Many authors used general terms such as ‘injury’ [47], ‘overuse injury’ [7], ‘repetitive strain injury (RSI)’ [9] and ‘pain’ [32] to describe PRMDs, but did not define these terms or use them to identify cases.

Eligibility criteria were described by only three authors [32,49,51]. Knowledge of eligibility criteria allows judgments about possible sampling biases to be made [45]. A wide range in prevalence rates of 26% [51] to 93% [49] was observed. The measurement bias resulting from lack of operational definitions and/or eligibility criteria, coupled with considerable variability in population characteristics and methodologies between the studies, mitigated against pooling of results using meta-analysis. Therefore a narrative synthesis of information regarding prevalence and risk factors was undertaken.

Table 4 summarizes risk factors investigated and significant findings. Only four authors demonstrated statistically significant risk factors, although many risk factors such as hand size, posture, practice habits, gender, age, change in practice routine and technique were discussed in the literature. Statistically significant risk factors were previous upper quadrant (bilateral, neck, shoulder or elbow) injury [47], small hand size [7], increasing age and female gender [32] and subjective measures of stress and a pain mediator (pain control) [52]. None of these risk factors were found to be significant in more than one study. The mean quality score of these papers was 9 (range = 7–13).

In the non-scored papers, hand size and anatomy (interconnections between tendons, hand shape), posture, technique and playing habits (warm-up, overuse/fatigue, scheduling, choice of repertoire and seating) were the most frequently discussed risk factors [11,12,27,56–63]. Of these, small hand size is the only risk factor that was found to be statistically associated with PRMDs in the 12 papers that were scored for methodological quality [7]. Six of the 11 non-scored papers outlined an operational definition of PRMD. Four authors defined the term overuse syndrome and this was used to describe PRMDs [11,12,27,63]. Two

authors defined ‘dystonia’, a disorder of motor control [56,61]. These definitions were less specific than those used by Shields and Dockrell [51]. Authors who did not define PRMDs used the term overuse syndrome [59], and other terms such as ‘tendinitis’ [57], ‘repetitive strain injury (RSI)’ [57,59], ‘physical injuries’ [59] or a general description of physical symptoms [58,60] to describe PRMDs. The wide variability in definition of PRMD in both scored and non-scored papers indicates a lack of consensus regarding the operational definition of PRMD.

Discussion

This is the first known systematic review of epidemiological literature regarding PRMDs in pianists. It is possible that not all evidence regarding prevalence of and risk factors associated with PRMDs was gathered in this review. However given the large number (38) and breadth of databases searched (medicine and arts/music), it is likely that a large percentage of relevant papers have been sourced. Because the quality scoring system equally weighted each item in this systematic review, items pertaining to bias and psychometric properties were given the same weight as arguably less important characteristics such as stating the purpose clearly. This potentially biases the total score, for example by scoring papers with different shortcomings equally [64]. However, this is a limitation of most critical appraisal tools, which do not weight items such as study description differently to items pertaining to bias and psychometric properties [65]. The cut-off score of 7 or above for synthesis was chosen arbitrarily in this review. It is standard practice to only include evidence of adequate methodological quality in a synthesis [44].

This review has identified several methodological limitations in the literature focusing on pianists. Because only one author used an operational definition of PRMD to identify cases [51], it is not possible to establish whether studies are measuring the same severity of PRMD. Zaza [22] conducted a systematic review of literature focusing on studies investigating incidence and prevalence of PRMDs in classical musicians. Ten of the 18 studies that were critically evaluated were ineligible for data synthesis, predominantly due to a lack of operational definition of PRMD.

In the papers eligible for data synthesis in Zaza’s review [22], the range of prevalence estimates was 39–87% when broad operational definitions of PRMD were used to establish prevalence, but reduced to 39–47% when definitions that excluded mild, transient complaints were used. The wide prevalence range (26–93%) in the studies evaluated in the current review, which focuses on pianists, suggests that a similar measurement bias is present.

Relative risk, odds ratios and differences in proportions are accepted statistical tests of association [40]. Five

authors used no statistical tests in their studies. This creates difficulties when interpreting the results of the studies regarding risk factors, as causation in these studies is inferred, rather than formally tested. The review by Zaza [22] identified a similar lack of statistical significance testing. In the current systematic review, four authors found statistically significant risk factors for PRMD [7,32,47,52]. These risk factors were previous upper quadrant (bilateral, neck, shoulder or elbow) injury, small hand size, increasing age and female gender and subjective measures of stress and a pain mediator (pain control). However three of the four authors did not state a response rate and the other response rate (61%) was poor [47]. Sampling biases such as poor response rates and lack of nonrespondent analysis can contribute to systematic error [46]. Moreover there was no consistency between the risk factors established in these papers.

In a narrative review of literature regarding PRMDs conducted by Bejjani *et al.* [66], potential risk factors were identified as changes in playing schedule, posture, technique, body habits and joint laxity. None of these risk factors are consistent with those established in the current systematic review.

However, Bejjani *et al.* [66] acknowledged that these postulated risk factors were supported largely by anecdotal evidence or clinical experience, rather than statistical association. Comparisons between the review by Bejjani *et al.* [66] and the current review are also limited because the previous authors conducted a narrative (not systematic) review and sourced papers from mixed instrumental cohorts. Moreover this review [66] was published well before most of the studies in the current systematic review were undertaken.

Very few single studies (as opposed to reviews) in performing arts medicine have statistically evaluated interactions between risk factors; they have generally employed simpler survey methodologies. In a comprehensive survey of 281 classical musicians (including, but not limited to, pianists) undertaken by Zaza and Farewell [21], female gender, playing a stringed instrument and number of years a musician has played were associated with higher PRMD risk. Several factors such as performing a musical warm-up and taking breaks were protective of a first-episode but not a recurrent PRMD. Other factors such as postural, workplace and scheduling changes were postulated for further investigation. Only one of these factors, female gender, is consistent with the statistically established risk factors identified in the literature in this systematic review.

No author combined a clear operational definition of PRMD with statistically established risk factors. The sourced studies either quantified the prevalence of a clearly defined disorder, or established associations between risk factors and disorders that were defined too broadly or not at all.

Other common methodological limitations identified included poor descriptions of study samples, which

create difficulties in comparing the results of different studies, and use of outcome measures with no justification of their reliability or validity. Measurement biases arising from the use of unreliable or poorly validated outcome measures can profoundly affect results [67]. Zaza's review [22] identified similar methodological limitations such as low response rates (<50%), unsystematic data collection and erroneous reporting of prevalence as incidence.

The establishment of a causal relationship between a risk factor and an outcome (in this case a PRMD) flows from a detailed evaluation of existing evidence with particular reference to internal validity, generalizability and comparison with other evidence [40]. The methodological limitations outlined make any conclusions regarding associations between risk factors and PRMDs in pianists difficult based on the results of this systematic review. Additionally, only two of the sourced papers utilized study designs that are considered desirable for establishing causation [40]. One of these was a cohort study [47], and one was a case-control study [7]. The remaining studies were cross-sectional surveys, which have the potential for other biases such as survivor bias [64].

Conclusion

This systematic review highlighted several methodological limitations in current studies investigating prevalence of and risk factors associated with PRMDs in pianists. Future studies should consider how cases of PRMD are defined, use valid and reliable measurement tools, utilize a recognized study design for establishing causation and perform appropriate statistical tests of association.

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Conflicts of interest

None declared.

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