ABSTRACT

Title of Thesis: A SYSTEMATIC REVIEW AND META-ANALYSIS OF PLAYING-RELATED MUSCULOSKELETAL DISORDERS IN MUSICIANS RELATING TO BIOLOGICAL SEX AND INSTRUMENT PLAYED

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Playing-related musculoskeletal disorders (PRMDs) are occupational health concerns that cause pain, discomfort, or disability for musicians. We utilized a systematic review and meta-analysis to analyze disparities in prevalence based on sex and instrument, and assess whether instrument choice was associated with sex-based prevalence disparities. Six databases were searched for studies containing prevalence rates for males or females, or specific instruments. Studies were evaluated for methodological quality and data was statistically evaluated in a meta-analysis. Our initial search yielded 5961 articles, of which 23 articles met quality assessment criteria and were included in quantitative analyses. Sex-based disparities were observed such that being female increased one’s odds of having a PRMD by 88% when compared to males. Limited available studies with relevant data prevented analysis of the interaction between instrument and sex.

A SYSTEMATIC REVIEW AND META-ANALYSIS OF PLAYING-RELATED MUSCULOSKELETAL DISORDERS IN MUSICIANS RELATING TO BIOLOGICAL SEX AND INSTRUMENT PLAYED

by

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Thesis submitted in partial fulfillment of the requirements
of the Gemstone Honors Program,
University of Maryland,
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Acknowledgements

We have many people to thank for helping us through our research process, especially as we transitioned our project to a virtual environment. First, we would like to thank our mentor, Dr. Shim, for his guidance and support throughout these past four years. We would also like to thank Dr. Skendall, Dr. Lovell, Dr. Coale, and the entire Gemstone staff for their direction and assistance. To our librarians, Nedelina Tchangalova and Stephen Henry, for answering questions, providing feedback, and advising us. To Dr. Bastepe-Gray, for helping facilitate a new path for our research and always being available to help. To Elizabeth Bell for sharing her expertise with the Vicon 3D motion capture system and assisting us with our statistical analyses, Dr. Kwon for his assistance with the SmartGuitar modifications, and Dr. Yang for her advice on meta-analysis. Lastly, to Humza Yahya, for his encouragement, advice, and enthusiasm for our research and for making late night meetings fun.

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Table 1
Eligibility Criteria

Inclusion Criteria	Exclusion Criteria
Published in peer-reviewed academic journal	Theses, dissertations, editorials, or letters
Written in English	Case study design
Study population composed of instrumental musicians	Study population composed solely of vocalists
Included subgroup prevalence/incidence rate of PRMDs for males, females, instrument group, or instrument	Systematic review articles
Received 6-8 points on the Loney Scale (Loney et al., 1998)	

Table 2
Quality Assessment Criteria: The Loney Scale

Loney Scale Question	Scoring Criteria
1. Are the study design and sampling method appropriate for the research question?	Random or whole population sampling method and described study population (1 point)
2. Is the sampling frame appropriate?	Sampling frame does not include risk of bias, no case control studies (1 point)
3. Is the sample size adequate?	Sample size > 1, no case studies (1 point)
4. Are objective, suitable and standard criteria used for measurement of the health outcome?	Measurements are validated questionnaires or the reliability and validity of measures are included (1 point)
5. Is the health outcome measured in an unbiased fashion?	Included description of how bias was reduced (1 point)
6. Is the response rate adequate? Are the refusers described?	Response rate of ≥70% and described refusers unless response rate of ≥85% (1 point)
7. Are the estimates of prevalence or incidence given with confidence intervals and in detail by subgroup, if appropriate?	Included an estimate of prevalence or incidence with appropriate sex or instrument subgroups (1 point)
8. Are the study subjects and the setting described in detail and similar to those of interest to you?	Included description of subjects and described PRMDs in relation to instrument or sex (1 point)
Total Possible Score	8 (*6 to include*)

Based on the Loney Scale (Loney et al., 1998)

Table 3

Quality Assessment: Loney Scale Scores

Author & Year	Loney Scale Score
Paarup et al. (2011)	8
Bruno et al. (2008)	7
Kenny & Ackermann (2015)	7
Ranelli et al. (2008)	7
Bragge et al. (2008)	6.5
Gohl et al. (2006)	6.5
Kennedy et al. (2006)	6.5
Kochem & Silva (2017)	6.5
Lima et al. (2015)	6.5
Logue et al.(2005)	6.5
Mehrparvar et al. (2012)	6.5
Morse et al. (2000)	6.5
Allsop & Ackland (2010)	6
Barton et al. (2008)	6
Brandfonbrener (1989)	6
Hagberg et al. (2005)	6
Hagglund & Jacobs (1996)	6
Joseph et al. (2020)	6
Larsson et al. (1993)	6
Nemoto & Arino (2007)	6
Pan et al. (1999)	6
Roach et al. (1994)	6
Vinci et al. (2015)	6
Ackermann et al. (2012)	5
Ackermann et al. (2014)	5
Brusky (2009)	5
Eller et al. (1992)	5

Engquist et al. (2004)	5
Fjellman-Wiklund & Chesky (2006)	5
Frizziero et al. (2018)	5
Gómez-Rodríguez et al. (2020)	5
Hartsell & Tata (1991)	5
Ioannou & Altenmüller (2015)	5
Kok et al. (2018)	5
Lockwood (1988)	5
Robitaille et al. (2015)	5
Rodríguez-Romero et al. (2016)	5
Sadeghi et al. (2004)	5
Savino et al. (2013)	5
Stanek (2019)	5
Stanhope et al. (2020)	5
Thrasher & Chesky (2001)	5
Viljamaa et al. (2017)	5
Woldendorp et al. (2016)	5
Zaza (1992)	5
Zaza & Farewell (1997)	5
Heikkilä et al. (2012)	4.5
Amorim & Jorge (2016)	4.5
Arnason et al. (2014)	4.5
Brandfonbrener (2009)	4.5
Brusky (2010)	4.5
Daenen et al. (2010)	4.5
Fishbein et al. (1988)	4.5
Fry (1987)	4.5
Furuya et al. (2006)	4.5
Gembris et al. (2018)	4.5

Kaneko et al. (2005)	4.5
Kovero & Könönen (1995)	4.5
Kreutz et al. (2008)	4.5
Lombart et al. (1995)	4.5
Lonsdale & Boon (2016)	4.5
Nawrocka et al. (2014)	4.5
Pampel et al. (2014)	4.5
Revak (1989)	4.5
Rickert et al. (2012)	4.5
Robitaille et al. (2018)	4.5
Sandell et al. (2009)	4.5
Selms et al. (2020)	4.5
Shaffer et al. (2013)	4.5
Shoup (1995)	4.5
Sousa et al. (2016)	4.5
Steinmetz et al. (2014)	4.5
Steinmetz et al. (2014)	4.5
Storms et al. (2016)	4.5
Yee et al. (2002)	4.5
Abréu-Ramos & Micheo (2007)	4
Ackermann & Adams (2003)	4
Beckett et al. (2015)	4
Britsch (2005)	4
De Smet et al. (1998)	4
Ginsborg et al. (2009)	4
Johansson & Theorell (2003)	4
Lonsdale et al. (2014)	4
Mathews & Mathews (1993)	4
Paarup et al. (2012)	4

Porter et al. (2018)	4
Rigg et al. (2003)	4
Schwab & Schultze-Florey (2004)	4
Semmler (1998)	4
Shanoff et al. (2019)	4
Steinmetz et al. (2015)	4
Wallace et al. (2016)	4
Yasuda et al. (2016)	4
Ackermann et al. (2011)	3.5
Ajidahun et al. (2017)	3.5
Bowie et al. (2000)	3.5
Caldron et al. (1986)	3.5
Chan et al. (2000)	3.5
Crnivec (2004)	3.5
Cygańska et al. (2020)	3.5
Doose et al. (2005)	3.5
Farias et al. (2002)	3.5
Fry & Rowley (1989)	3.5
Fry et al. (1988)	3.5
Heming (2004)	3.5
Kok et al. (2015)	3.5
Ling et al. (2018)	3.5
Ranelli et al. (2011)	3.5
Rodríguez-Lozano et al. (2010)	3.5
Roset-Llobet et al. (2000)	3.5
Shields & Dockrell (2000)	3.5
Sousa et al. (2017)	3.5
Stanek et al. (2017)	3.5
Boyle (2012)	3

Brown (1997)	3
Davies & Mangion (2002)	3
Devroop et al. (2002)	3
Fotiadis et al. (2013)	3
Grieco et al. (1989)	3
Kaufman-Cohen et al. (2018)	3
Kok et al. (2013)	3
O'Neill L et al. (2001)	3
Pak & Chesky (2001)	3
Papandreou & Vervainioti (2010)	3
Steinmetz et al. (2006)	3
Thrasher & Chesky (1998)	3
Yoshimura et al. (2008)	3
Adam et al. (2018)	2.5
Woldendorp et al. (2018)	2.5
Yoshimura et al. (2006)	2.5
Kenny et al. (2018)	2
Manchester (1988)	2

Table 4

Overall Prevalence Estimates of PRMDs from 23 High-Quality Studies

			95% Confidence Interval	
Author (Year)	**Type of Instrument**	**Prevalence of PRMDs**	**Lower Bound**	**Upper Bound**
Allsop & Ackland (2010)		0.42	0.38	0.46
Barton et al. (2008)		0.65	0.55	0.74
Bragge et al. (2008)	Piano	0.68	0.58	0.78
Brandfonbrener (1989)		0.29	0.27	0.31
Bruno et al. (2008)	Piano	0.38	0.32	0.45
Gohl et al. (2006)	Piano	0.16	-0.01	0.32
Hagberg et al. (2005)		0.24	0.19	0.29
Hagberg et al. (2005)	Piano	0.32	0.17	0.46
Hagberg et al. (2005)	Strings	0.26	0.19	0.33
Hagberg et al. (2005)	Woodwind	0.19	0.12	0.26
Hagglund & Jacobs (1996)		0.62	0.48	0.76
Joseph et al. (2020)	Percussion	0.69	0.44	0.94
Kennedy et al. (2006)	Strings	0.25	0.08	0.42
Kenny & Ackermann (2015)		0.85	0.81	0.88
Kochem & Silva (2017)	Strings	0.87	0.80	0.93
Larsson et al. (1993)		0.37	0.33	0.40
Lima et al. (2015)	Strings	0.78	0.59	0.97
Logue et al. (2005)	Strings	0.43	0.17	0.69
Mehrparvar et al. (2012)		0.44	0.39	0.50
	Percussion	0.68	0.57	0.79
	Piano	0.32	0.15	0.49
	Strings	0.44	0.37	0.50
	Woodwind	0.44	0.21	0.67
Morse et al. (2000)		0.29	0.23	0.36
	Brass	0.25	-0.05	0.55
	Percussion	0.50	0.10	0.90
	Piano	0.33	0.23	0.44
	Strings	0.31	0.19	0.42
	Woodwind	0.17	0.02	0.33
Nemoto & Arino (2007)	Woodwind	0.30	0.24	0.36
Paarup et al. (2011)		0.89	0.85	0.92
Pan et al. (1999)		0.64	0.49	0.79
Pan et al. (1999)	Percussion	1.00	1.00	1.00
Ranelli et al. (2008)		0.67	0.63	0.70
Roach et al. (1994)		0.68	0.58	0.78
Vinci et al. (2015)	Strings	0.26	0.16	0.37

Note. Unless otherwise specified, prevalence rates represent multiple or unspecified instrument subgroups.

Table 5

Prevalence and Odds Ratios of PRMDs Based on Sex

		95% CI			95% CI			95% CI		
Author (Year)	**Female Prevalence**	**Lower Bound**	**Upper Bound**	**Male Prevalence**	**Lower Bound**	**Upper Bound**	**Odds Ratio**	**Lower Bound**	**Upper Bound**	
Barton et al. (2008)	0.82	0.71	0.93	0.50	0.36	0.64	1.07	0.82	1.40	
Brandfonbrener (1989)	0.29	0.27	0.32	0.28	0.23	0.33	1.88	0.54	6.48	
Bruno et al. (2008)	0.42	0.33	0.51	0.33	0.23	0.44	2.24	1.25	4.02	
Hagglund & Jacobs (1996)	0.68	0.51	0.85	0.53	0.29	0.77	2.04	1.48	2.81	
Kenny & Ackermann (2015)	0.90	0.85	0.94	0.79	0.73	0.85	6.58	2.28	18.96	
Larsson et al. (1993)	0.46	0.40	0.51	0.29	0.24	0.34	1.16	0.24	5.58	
Paarup et al (2011)	0.97	0.94	1.00	0.83	0.78	0.88	1.73	0.70	4.28	
Pan et al. (1999)	0.67	0.36	0.97	0.63	0.46	0.81	1.66	1.12	2.47	
Ranelli et al. (2008)	0.70	0.65	0.74	0.62	0.57	0.68	1.08	0.42	2.78	
Roach et al. (1994)	0.73	0.61	0.86	0.62	0.46	0.77	1.50	0.42	5.32	
						Pooled Effect:	**1.8802**	**1.285**	**2.7511**	***p* = 0.0045**

Table 6

Prevalence and Odds Ratio of PRMDs Based on Sex Within Instrument Groups

			95% CI			95% CI			95% CI	
Author (Year)	**Type of Instrument**	**Female Prevalence**	**Lower Bound**	**Upper Bound**	**Male Prevalence**	**Lower Bound**	**Upper Bound**	**Odds Ratio**	**Lower Bound**	**Upper Bound**
Allsop & Ackland (2010)	Piano	0.46	0.41	0.51	0.34	0.26	0.41	4.63	1.81	11.81
Bragge et al. (2008)	Piano	0.68	0.56	0.80	0.67	0.50	0.84	1.44	0.79	2.62
Vinci et al. (2015)	Strings	0.29	0.16	0.41	0.21	0.03	0.39	1.88	0.98	3.58
Nemoto & Arino (2007)	Woodwind	0.41	0.28	0.55	0.27	0.21	0.34	1.38	1.01	1.89

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Figure 1
PRISMA 2009 Flow Diagram

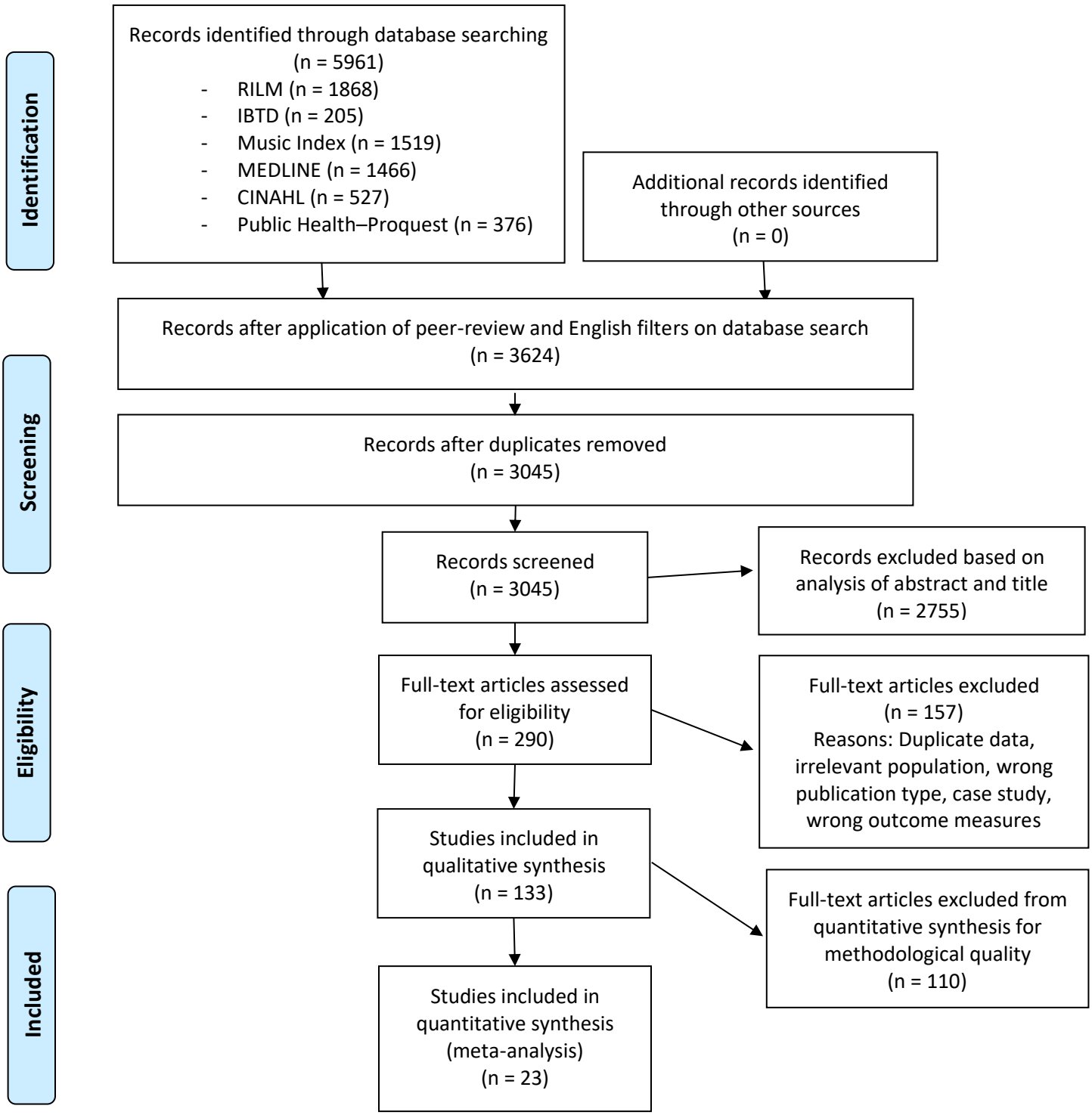


Figure 2

Prevalence of Playing-Related Musculoskeletal Disorders Across 23 Studies

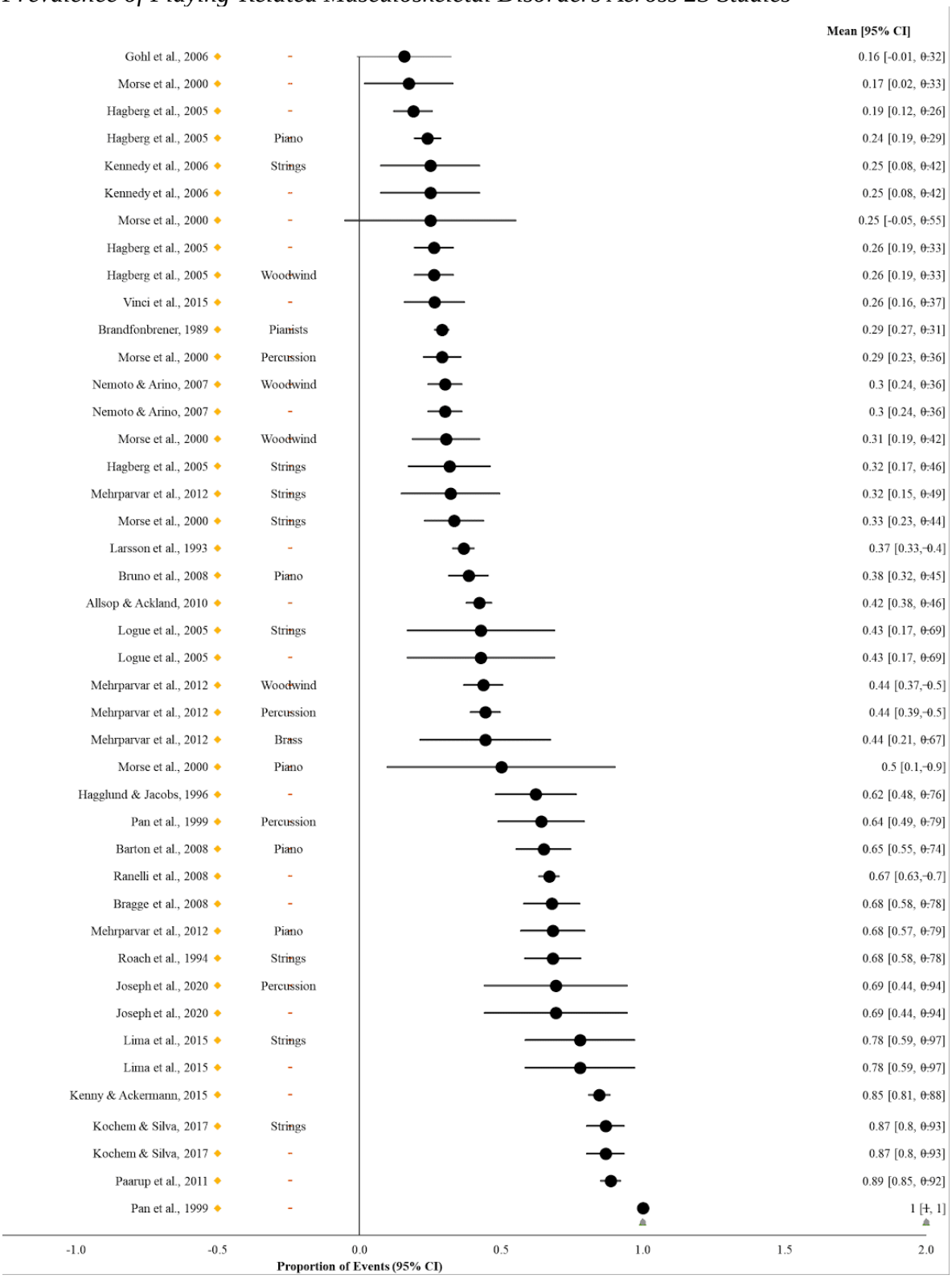
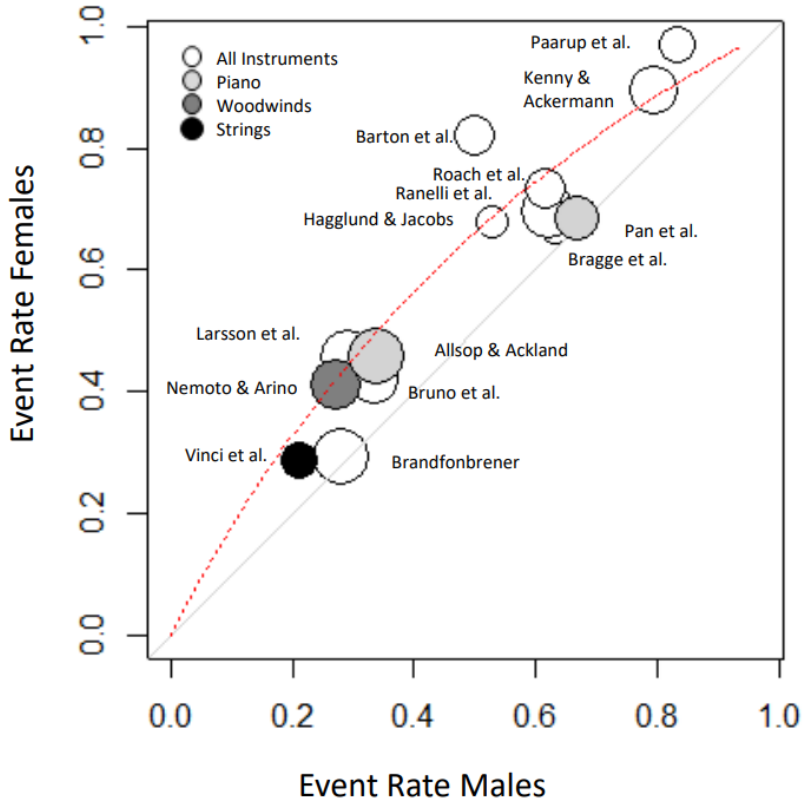


Figure 3

L'Abbé Plot of Relative Risk of PRMDs for Females Compared to Males



Note. The size of the circle indicates the sample size of the study. The instrument population of the study is indicated by color, with the majority of the studies including a combination of musicians from multiple instrument groups. The gray line indicates a one-to-one ratio for prevalence of PRMDs between males and females. The dotted red line indicates the trend of the 23 studies included in the meta-analysis.

Chapter 1: Introduction

Playing-Related Musculoskeletal Disorders (PRMDs)

Playing-related musculoskeletal disorders (PRMDs) are conditions musicians can develop from playing an instrument that negatively impact their health and professional careers. PRMDs have been found to impact between 62-93% of musicians in their lifetime (Kok et al., 2016). Symptoms associated with PRMDs often lead musicians to be unable to play their instrument or impair musicians' performances (Zaza et al., 1998). This inability to play may result in risk of job loss and financial strain, making fear of unemployment one of the most significant stressors affecting musicians with PRMDs (Zaza et al., 1998). Severity of PRMD pain has also been found to be associated with depressive symptoms in orchestral instrumentalists, indicating the importance of improving understanding of these conditions to reduce the mental, physical, and financial burdens of PRMDs on musicians (Kenny & Ackermann, 2015).

A critical health disparity between biological males and females has been reported in which PRMDs are more common in females than males. Multiple systematic reviews including a variety of instrumentalists across a large age-range support that females exhibit higher rates of PRMD symptoms than males (Baadjou et al., 2016; Kok et al., 2016; Kochem & Silva, 2018). Factors influencing this occupational health disparity require further investigation and may include physiological as well as cultural influences. An understanding of both the physiological and cultural factors

underlying this disparity is imperative to inform the development of prevention and intervention efforts to address PRMDs and overcome this critical health disparity.

Team METR: Original Research Study

Prior to the outbreak of COVID-19, Team METR proposed to complete a primary research study to investigate the biomechanical correlates of PRMDs in guitarists, such as finger joint forces/torques and muscular co-contraction, and reveal the underlying mechanisms of the sex-based disparity in prevalence of PRMDs. The proposed novel research would have utilized an innovative guitar-like apparatus (SmartGuitar) developed through collaboration between the Kinesiology department at the University of Maryland (UMD) and the Peabody Institute at the Johns Hopkins University and refined by Team METR. Since the guitar is a popularly played instrument, the proposed study would have focused on guitarists to maximize the impact of its insights into detrimental, but preventable, risk factors of PRMDs. We proposed to recruit participants with and without a history of PRMDs composed of equal numbers of males and females to investigate group differences in selected biomechanical correlates of playing guitar between males and females and participants with and without PRMDs. Participants would have visited the UMD Neuromechanics Research Core and performed multiple trials of short excerpts standardized for speed and loudness on the SmartGuitar. The excerpts were selected from the commonly encountered movement patterns on the guitar. Simultaneous force, kinematic, and muscular activation data would have been collected using force sensors embedded in the SmartGuitar, infrared motion capture cameras, and electromyography.

The proposed study was intended to investigate the underlying biomechanical mechanisms of the development of PRMDs and explore physiological correlates of the greater prevalence of PRMDs in females than males. The identification of these mechanisms by future researchers will be instrumental in developing early prevention efforts to reduce the prevalence of PRMDs; improve the physical health, occupational security, and quality of life of guitarists; promote evidence-based instrument pedagogy regarding PRMD prevention; and offer solid preliminary data for future research on causality between the identified biomechanical markers and playing related musculoskeletal and motor control disorders in a longitudinal study.

Adjustment Following Outbreak of COVID-19

Following the outbreak of COVID-19 and the subsequent closure of research activities, Team METR revised our plan for the completion of our thesis project. Rather than complete the proposed primary research study, for which we had received IRB approval and had been approaching recruitment and data collection in March of 2020, we decided to complete a meta-analysis and systematic review on the associated factor of instrument choice that may relate to the observed sex-based disparity in prevalence of PRMDs. This meta-analysis was intended to inform future researchers investigating the sex-based disparity in prevalence of PRMDs by highlighting the role of instrument choice in this disparity and synthesizing existing knowledge on this topic.

Chapter 2: Systematic Review and Meta-Analysis Article Manuscript

Abstract

Playing-related musculoskeletal disorders (PRMDs) are occupational health concerns that have been shown to affect many musicians and other performing artists by causing discomfort and, in some cases, chronic pain or disability. A systematic review and meta-analysis were conducted to address whether prevalence rates of PRMDs differed based on sex and instrument, and whether instrument choice was associated with previously reported disparities in prevalence between males and females. Six literature databases were searched for cross-sectional studies, longitudinal studies, and randomized control trials for which a baseline prevalence or incidence rate could be determined for males or females, or for specific instruments. Studies were evaluated by two reviewers for methodological quality using the Loney Scale, and data was extracted, summarized, and statistically evaluated in a meta-analysis. We identified 5961 articles in our initial search, 133 of which met criteria for methodological assessment. Only 23 articles received a "good" score on the Loney Scale, suggesting strong methodological quality, and were included in quantitative analyses. Sex-based disparities were observed in the literature, such that being female increased one's odds of having a PRMD by 88% when compared to males. Only four articles included sex-based PRMD prevalence estimates within

instrument groups. Therefore, no analyses were conducted to assess the interaction between instrument and sex on prevalence.

Introduction

Playing-related musculoskeletal disorders (PRMDs) are occupational health disorders which affect musicians and other performing artists, causing discomfort and, in some cases, chronic pain or disability (Zaza, 1997). PRMDs are extremely common, affecting between 62-93% of musicians in their lifetime (Kok et al., 2016). Further, the Zaza et al. (1998) review determined that in studies assessing point prevalence rates of PRMDs, an estimated 34-62% of secondary school musicians and 39-87% of adult musicians were affected by PRMDs, signifying a critical need for additional research on PRMDs.

Biological Sex as an Associated Factor of PRMDs

While these prevalence rates are striking, disparities have been observed in the impact of PRMDs and it has been reported that PRMD prevalence rates differ based on a variety of factors. Perhaps the most well-documented disparity in prevalence of PRMDs is based on the sex of the instrumentalist. Multiple previous reviews and studies have shown evidence of an unexplained disparity between male and female musicians, such that female musicians are much more likely to develop PRMDs than male musicians (Kochem & Silva, 2018; Kok et al., 2016). Despite the considerable evidence suggesting a sex-based disparity in prevalence of PRMDs, there is very little research exploring the underlying factors associated with this disparity. One suggestion is that there are physiological differences between males and females that

make females more likely to develop PRMDs. For instance, a review conducted by Côté (2012) found evidence suggesting a sex-based difference in muscular coordination and movement strategies which could potentially contribute to sex-based differences in occupational health disorders. An additional study utilizing electromyography (EMG) during repetitive motion tasks reported significant sex-based differences in the patterns of muscular variability and activity that contributed to higher endurance, suggesting that different injury mechanisms may contribute to the development of musculoskeletal disorders between males and females (Fedorowich et al., 2013). Sex-based differences in physiological predispositions to injury have also been observed in musicians. For instance, in a study assessing postural stabilization systems in musicians, it was found that female musicians displayed a higher frequency of dysfunction of the scapular stabilizers than male musicians, and that significant sex-based differences were present for the development of musculoskeletal disorders (Steinmetz et al., 2010).

Instrument Played as an Associated Factor of PRMDs

However, one potential reason for this sex-based disparity that is not well-researched is the possibility of gender-based instrument choice. The location and prevalence of pain due to PRMDs have been suggested to differ based on the type of instrument played. A review assessing the prevalence of PRMDs in string musicians reported extremely high 12-month prevalence rates (64.1-90%) as well as lifetime prevalence rates (77-89.5%), but also reported that the location of pain varied by type of instrument played, such that violists and violinists were more likely to develop pain

of the left elbow, left shoulder, left hand, and neck, while bassists and cellists were more likely to develop pain of the right elbow, right shoulder, right hand, and lumbar spine (Kochem & Silva, 2018). Additionally, a systematic review conducted by Zieba et al. (2019) reported that instrumentalists who played wind instruments, the violin, and the viola experienced the greatest frequency of PRMD pain. An additional review found that brass instrumentalists reported the lowest prevalence rates of PRMDs (Kok et al., 2016). It has been suggested that the instrument-based disparity in prevalence rates of PRMDs, and location of pain associated with PRMDs, results from the differing physiological demands of playing certain types of instruments (Kochem & Silva, 2018).

Gender-Based Instrument Choice

Given that prevalence rates of PRMDs vary between instrumental groups, gender-based instrument choice provides an alternative explanation for the disparities observed in PRMD prevalence based on sex (Kochem & Silva, 2018; Kok et al., 2016; Zieba et al., 2019). Several studies have investigated gender-based instrument choice, reporting that many instruments are played more frequently by either males or females, in part due to stereotypes surrounding these instruments (Harrison & O'Neill, 2000; Wrape et al., 2016). There exists a complex relationship between gender and musical instruments that has historically resulted in both the gendering of musical instruments and the establishment of gender roles among instrumentalists (Doubleday, 2008). One study found that gender roles significantly affected college students' perceptions of females and males who played either male-dominant or

female-dominant instruments (Cramer et al., 2002). Specifically, instrumentalists playing “masculine” instruments were perceived as possessing more masculine traits while instrumentalists playing “feminine” instruments were perceived as possessing more feminine traits (Cramer et al., 2002). Not only are instrumentalists perceived differently based on the instrument they play, instruments themselves are often perceived as being masculine or feminine (Abeles & Porter, 1978). A study conducted on adult non-musicians found that the flute, violin, and clarinet are considered feminine instruments and the trumpet, trombone, and drums are considered masculine (Abeles & Porter, 1978). Further, undergraduate college students rated the string bass, trumpet, and tuba as masculine and the flute, harp, and piccolo as feminine (Groswold & Chroback, 1981). One study suggests that preferences for instruments based on gender begin early in life but develop earlier and with less variance for boys than girls (Abeles & Porter, 1978). These gendered perceptions of instruments have been found to be moderately stable in more recent studies (Abeles, 2009; Wych, 2012). In support of the early development of these stereotypes, Wrape et al. (2016) found that most instruments were rated relatively consistently as “boy” or “girl” instruments by middle school band students. In particular, the flute and clarinet were consistently rated as “girl” instruments, while the tuba was consistently rated as a “boy” instrument (Wrape et al., 2016).

Gender-based stereotypes appear to influence the instruments musicians choose to play. As reported in Hallam et al. (2008), girl students played the flute, harp, clarinet, violin, oboe, and piccolo at disproportionately high rates while boys composed high proportions of those playing the electric and bass guitar, kit drums, trombone, tuba,

and tabla. Experience influences the development of these stereotypes and their role in instrument choice. Wrape et al (2016) reported that less experienced middle schoolers exhibited the least stereotypical choices; however, sex-based perceptions of instruments changed and solidified with greater experience and skill-level. Additionally, an experiment found that children's initial interest in and stereotyped beliefs regarding instruments were generally consistent with stereotypes reported in previous literature (Harrison & O'Neill, 2000). However, these beliefs changed following concerts in which children were exposed to either gender-consistent or gender-inconsistent instrumentalists (Harrison & O'Neill, 2000). In particular, the gender-inconsistent concert intervention was associated with changes to children's preferences for learning previously highly rated instruments that had been stereotyped as "girl" or "boy" instruments (Harrison & O'Neill, 2000). Thus, experience appears to play a role in the solidification and development of gender-based stereotypes regarding instrument choice.

Current Study

It is unclear if gender-based instrument choice plays a role in the reported sex-based disparity in prevalence of PRMDs. However, previous studies have shown that gender-based instrument choice produces differing rates of females and males between instrument groups (Hallam et al., 2008), and the prevalence of PRMDs varies based on instrument (Kok et al., 2016; Zieba et al., 2019). This meta-analysis will investigate the relationship between sex, instrument, and the prevalence of PRMDs by compiling the findings of previous studies on the sex-based and

instrument-based prevalence and incidence rates of PRMDs. It is predicted that there will be an association between instrument and gender on PRMD prevalence such that instruments for which higher rates of PRMDs are reported will overlap with the instruments for which females compose a larger proportion of instrumentalists than males. This interaction would suggest a relationship between instrument choice and the reported sex-based disparity in rates of PRMDs, although the direction of the relationship would need to be clarified by future research.

Methods

A systematic review and meta-analysis were conducted to synthesize the findings of previous research on instrument and sex-based differences in prevalence of playing-related musculoskeletal disorders (PRMDs) in musicians.

Eligibility Criteria

To be included for analysis, articles had to meet the following criteria: (1) the article was published in a peer-reviewed academic journal; (2) the article was written in English; and (3) the study population was instrumental musicians. Only articles that included prevalence or incidence rates of playing-related musculoskeletal disorders (PRMDs) separately for males and females or for specific instrument groups or instruments were included. Articles for which a prevalence or incidence rate could not be calculated for a specific sex, instrument group, or instrument were excluded. Case studies were excluded from analysis. Articles denoting dissertation papers, systematic review articles, editorials, and letters were excluded from analysis. Articles in which the sample was solely composed of vocalists were also excluded.

Only articles that received scores of 6 to 8 points on the Loney Scale were included for quantitative analysis (Loney et al., 1998). These eligibility criteria are outlined in Table 1.

Search Strategy

A literature search was conducted between 10 August 2020 - 13 August 2020 using the following databases: Répertoire International de Littérature Musicale (RILM) - Abstracts of Music Literature, Music Index, MEDLINE, CINAHL, International Biography of Theatre and Dance (IBTD), and Public Health - Proquest. Two sets of search terms were included in the search: **population terms** (e.g. "music", "instrumentalist", and "musical instrument") AND **health issue terms** (e.g. "musculoskeletal", "PRMD", "overuse", "repetitive motion injury", "occupational disorder", and "cumulative trauma disorder"). These terms were expanded upon using the N5 near operator and asterisk (*) search symbols. The complete search strategy can be found in Appendix A. Citations were tracked and duplicates were removed using Zotero, an online citation management tool developed by the Corporation for Digital Scholarship, based in the United States (https://www.zotero.org/). Articles were included or excluded based on our eligibility criteria using Rayyan. Rayyan Systems Inc. is an online software tool based in the United States for conducting collaborative systematic reviews (https://www.rayyan.ai/) (Ouzzani et al., 2016).

Study Selection

The literature search resulted in 5961 published articles. The application of search filters to include only peer-reviewed journal articles written in English resulted in

3045 relevant articles. 579 duplicates were removed. Two independent reviewers initially screened the title and abstract of articles for eligibility. All disagreements were resolved by group consensus. This screening identified 290 eligible articles. The full text of these 290 articles were then reviewed for eligibility by two reviewers and disagreements were again resolved by group consensus. This screening led to the exclusion of 157 articles, resulting in the identification of 133 eligible articles for inclusion in this paper. Following quality assessment, only 23 articles received at least 6 out of 8 possible points on The Loney Scale and were included in this meta-analysis (Loney et al., 1998).

Quality Assessment

The Loney Scale was utilized to assess the methodological rigor and quality of articles for inclusion (Loney et al., 1998). The Loney Scale establishes a guideline on assessing the quality of studies that estimate prevalence and/or incidence rates (Pereira et al., 2011). This scale was chosen because other systematic reviews in the field of occupational health disorders used the Loney Scale to critically appraise articles that report the prevalence of PRMDs (Kochem and Silva, 2018; Kok et al., 2016). The Loney Scale is composed of eight questions for which scoring criteria were selected prior to evaluation, as listed in Table 2 (Loney et al., 1998). All evaluations were conducted by two independent reviewers and group consensus resolved all conflicts for inclusion in the meta-analysis. Conflicts that did not affect inclusion were resolved by averaging the two scores. Consistent with previous reviews on PRMDs, articles were considered of poor quality if they received between 0 and 2 points, regular quality if they received between 3 and 5, and good quality if

they received between 6 and 8 (Kochem & Silva, 2018). Only articles of good methodological quality (6-8 points) were included in the meta-analysis.

Data Extraction

For each included article, overall sample size, sample size for subgroups of interest (female, male, and/or instrument group), response rate, methodological descriptions, and prevalence or incidence rates for subgroups of interest were extracted when appropriate. Relevant information on the article's author, title, and year published were also extracted.

Statistical Method

A Random-Effects meta-analysis of the event rate data between male and female groups was performed in R software using the R package meta (Balduzzi et al., 2019). A Random-Effects model was selected to account for the large heterogeneity in the populations of included articles. Odds Ratios (OR) and 95% confidence intervals were computed using the Mantel-Haenszel method for each individual study. The overall odds ratio, 95% confidence interval, and p-value were computed for groups of studies that presented sex-specific event rates for musicians (with $\alpha = 0.05$).

Results

There were three central hypotheses to this study. The first hypothesis was that females would be significantly more likely to report having a PRMD. The second hypothesis was that PRMD prevalence would differ significantly between instrument

groups. The final hypothesis was that there would be an interaction between instrument group and sex such that instruments found to show a greater prevalence of PRMDs would also be composed of a larger proportion of females. In order to test these hypotheses, a systematic review was conducted, yielding data from 133 peer-reviewed studies on PRMDs. Of the 133 articles examined for methodological rigor and quality, only 23 received a score between 6 and 8 on the Loney Scale, identifying the study's methodology as high quality (Loney et al., 1998; see Table 2). Loney Scale scores for each individual article can be found in Table 3.

Overall Prevalence of PRMDs

The overall prevalence rates of PRMDs as reported by the 23 high quality studies included for quantitative analysis are outlined in Table 4. The forest plot in Figure 2 depicts these prevalence rates. There was considerable variability in the total prevalence reported across the 23 studies, ranging from 16% (Gohl et al., 2006) to 89% (Paarup et al., 2011).

Sex-Based Disparity in Prevalence

To test the hypothesis that females would show a greater risk of PRMDs than males, the Mantel-Haenszel meta-analytical method was used with a Hartung-Knapp adjustment for random effects to determine the pooled odds ratio (OR) for the likelihood of PRMDs for females when compared to males. Ten studies included a breakdown of PRMD cases by sex. Across these ten studies, the pooled OR for having a PRMD was 1.8802 for females when compared to males, with a 95% confidence interval for the true population OR between 1.285 and 2.7511 (t=3.75,

p=0.0045). Therefore, being female increased the odds of having a PRMD by approximately 88% when compared to males across these studies. The ORs and confidence intervals from these analyses are included in Table 5. The prevalence rates of PRMDs for females relative to males in each of the ten studies are also depicted in the L'Abbe plot in Figure 3, with male prevalence on the x-axis and female prevalence on the y-axis. The overall trend of ORs across studies included in the meta-analysis is also depicted in Figure 3, showing that all studies reported a greater PRMD prevalence for females relative to males, with the exception of Brandfonbrener (1989), which reported approximately equal prevalence rates of females at 29% (95% CI: 27-32%) to males at 28% (95% CI: 23-33%).

Effect of Instrument on Prevalence

Fifteen studies of high methodological quality and rigor reported data regarding instrument-based prevalence of PRMDs. There were six studies that reported prevalence rates for pianists that ranged from 16% to 68%, eight studies that reported prevalence rates for string instrumentalists that ranged from 25% to 87%, four studies that reported prevalence rates for woodwind instrumentalists that ranged from 17% to 44%, and four studies that reported prevalence rates for percussionists that ranged from 50% to 100%. Only one study reported a prevalence rate for brass instrumentalists of 25%.

Interaction Between Sex and Instrument on PRMD Prevalence

Only four studies of good methodological quality and rigor included data on the male and female prevalence of PRMDs within instrument groups. Of the four studies that

reported prevalence estimates for males and females within instrument groups, two studies reported prevalence for pianists (Allsop & Ackland, 2010; Bragge et al., 2008), Vinci et al. (2015) reported on string instrumentalists, and Nemoto & Arino (2007) reported on woodwind instrumentalists. The odds of females having PRMDs relative to males for these four studies are included in Table 6. However, statistical analyses were not included for the interaction between instrument and gender due to the small number of high-quality studies reporting relevant data.

Discussion

Previous literature on PRMDs has provided robust evidence of sex-based disparities in prevalence of PRMDs in musicians. However, very little research has been dedicated to exploring potential mechanisms that account for these sex-based differences. Some research has pointed to physiological mechanisms that may account for sex-based differences in the development of PRMDs (Côté, 2012; Fedorowich et al., 2013; Steinmetz et al., 2010), but no research, to the knowledge of the researchers, has attempted to connect cultural factors such as sex-based instrument choice to higher prevalence of PRMDs in females. Investigating this association is crucial in establishing a more holistic understanding of factors that contribute to this health disparity. In order to address this gap, the present study synthesized previous research on PRMD prevalence in order to test three hypotheses.

The first hypothesis of the present study was that disparities would be observed in the prevalence of PRMDs on the basis of sex, such that biological females would show higher rates of PRMDs than males. Results from the meta-analysis indicate that

female instrumentalists' odds of having a PRMD were 88% higher than that of male instrumentalists. This study contributes quantitative evidence in support of the findings of previous systematic reviews, such as those conducted by Kochem & Silva (2018) and Kok et al. (2016), by conducting a meta-analysis to report a pooled odds ratio for the likelihood of females having PRMDs compared to males.

The second and third hypotheses of this study were that the prevalence of PRMDs would differ between instrument groups and that there would be an interaction between sex and instrument, such that instrument groups found to be associated with greater risk of PRMDs would have a significantly greater proportion of female players to male players. Given the small number of studies of good methodological quality reporting on these factors, we were unable to conduct statistical analyses to test these hypotheses. This lack of high-quality research was a major limitation of the present study, making it impossible to test for interactions between sex and instrument played on prevalence of PRMDs. Future research should include comparisons of the relative risk of male and female instrumentalists and the proportion of female and male players within different instrument groups to investigate the potential interaction of sex and instrument choice in the development of PRMDs.

One consideration relating to the third hypothesis of this study is the possibility of gender-influenced instrument choice serving a protective function for females. For example, Paruup et al. (2011) found that woodwind instrumentalists were at lower risk of PRMDs compared to other instrumentalists. Therefore, the instruments

selected by females, specifically the flute, for which females compose a greater proportion of players than males (Sergeant & Himonides, 2019), may function to reduce females' heightened risk of PRMDs based on sex, particularly when compared to those playing larger instruments. In this context, gender-based instrument choice may be protective against developing PRMDs for females based on an interaction between sex-based physical characteristics and gender-influenced instrument choice. It is important to note, however, that though gender-based instrument choice might be protective in some cases, prevalence of PRMDs appears higher for females across many studies, indicating the need for more research on the contexts in which instrument choice may be protective or a risk factor.

Limitations & Future Directions

Besides the lack of high-quality studies available for testing our second and third hypotheses, there were several additional limitations to the present study. One limitation of this meta-analysis was the lack of distinction between prevalence estimates across different periods of time. For instance, studies reporting point prevalence estimates were included in this analysis alongside studies reporting six-month prevalence estimates. Prevalence estimates have previously been reported to differ based on the length of time across which estimates are made, with larger prevalence reported when the prevalence period was longer (Spronk et al., 2019). However, the quantity of high-quality studies identified by this review did not lend itself to further analyses based on prevalence periods. Future reviews should include separate analyses for all relevant time periods, once additional studies of good methodological quality have been published.

Another limitation of the present study is that previous research reports on differences in PRMD prevalence based on sex. However, instrument choice is often influenced by cultural expectations and norms surrounding gender identity, not necessarily biological sex (Cramer et al., 2002; Hallam et al., 2008; Harrison & O'Neill, 2000). The studies in our sample did not focus on the cultural factors that affect instrument choice, such as perceived associations between instruments and gender identity. Since music is highly reflective of culture, it is possible that it also reflects perceived gender roles within respective societies, which could be an important consideration when analyzing prevalence rates.

Given the finding that female instrumentalists' odds of having a PRMD were 88% higher than that of male instrumentalists, future research should investigate the many different factors that may underlie this critical health disparity. Research investigating potential anthropometric and physiological differences in playing between males and females would expand upon existing findings that suggest biomechanical correlates may in part explain the greater risk of PRMDs for females when compared to males (Steinmetz et al., 2010). However, research investigating cultural factors that may contribute to this disparity, such as gender-based instrument choice, is also needed to further elucidate the variety of mechanisms that underlie female instrumentalists' heightened risk of developing occupational musculoskeletal disorders (Hallam et al., 2008; Harrison & O'Neill, 2000; Wrape et al., 2016). Future research on these biomechanical and cultural mechanisms could inform musical educators on how to

better tailor their teaching to reduce the risk of their students developing PRMDs. For instance, a playing strategy or pedagogical approach that is successful for one student may increase the risk of developing a PRMD for another student with different physical characteristics, such as a smaller hand span. Therefore, expanding understanding of these conditions is imperative to address the critical disparity in health outcomes between male and female instrumentalists through informed musical pedagogy and intervention efforts.

Conclusion

Despite these limitations, this systematic review and meta-analysis provides important contributions to the study of occupational musculoskeletal disorders in musicians. This meta-analysis quantified the odds of having a PRMD for females relative to males based on twenty-three high quality studies. Across these studies, females displayed a greater prevalence of PRMDs than males. This meta-analysis was also the first to attempt to investigate gender-based instrument choice as a potential mechanism underlying the sex-based disparity in PRMD prevalence, although there was not sufficient number of high-quality studies to complete these analyses. Future research should investigate the cultural, anthropometric, and biomechanical factors that explain this sex-based disparity to support musical pedagogy and inform prevention and intervention efforts.

Chapter 3: Equity Impact Statement

Music is a universally practiced art across cultures and societies globally. As such, music acts as a lens through which to observe equity-based disparities that exist within cultures. Previous research supports a sex-based disparity in the prevalence of playing-related musculoskeletal disorders (PRMDs) among musicians, such that females report disproportionately high rates of PRMDs when compared to males (Kochem & Silva, 2018; Kok et al., 2016). Previous research on occupational health disorders, including PRMDs, often explored physiological or musculoskeletal mechanisms to explain these differences (Côté, 2012; Fedorowich, 2013; Steinmetz et al., 2010). However, as music is highly reflective of culture, it is important to also consider experiences of sex and gender when investigating these disparities. Inequities and gender stereotypes are reflected by the ways we perceive different types of instruments as masculine or feminine, and therefore have been shown to have a gender-based influence on the instrument selections young musicians make (Doubleday, 2008; Hallam et al., 2008; Harrison & O'Neill, 2000; Wrape et al., 2016). However, the role of gender-based instrument choice as a potential mechanism through which we can explain the sex-based disparity in PRMD prevalence has not been investigated by previous researchers. An understanding of the cultural, in addition to physical, factors underlying this disparity could be crucial in enhancing our understanding of the issue and the development of prevention and intervention efforts to address PRMDs.

By investigating the relations between instrument played and sex-based prevalence of PRMDs, we aimed to address a significant gap in the existing literature on these potentially debilitating occupational health disorders. We discovered that across 23 high quality studies, being female substantially increased one's odds of having a PRMD by 88% when compared to males. Despite this sex-based disparity in PRMD prevalence, only four studies of good methodological quality reported male and female prevalence within instrument groups. The lack of high-quality research investigating this sex-based disparity prevented us from completing a statistical analysis of the interaction between sex and instrument on PRMD prevalence. Future research must address this critical health disparity to enhance our understanding of PRMDs and promote equitable health outcomes through targeted prevention and intervention programs.

Appendix A: Search Criteria

(“music*” OR “musical N5 instrument*” OR “instrumentalist*”) AND

(“musculoskeletal” OR “PRMD*” OR “performance related disorder*” OR “overuse” OR “overuse N5 syndrome” OR “repetitive motion injur*” OR “cumulative trauma disorder*” OR “occupational disease*” OR “occupational disorder*” OR “occupational health disorder*” OR “WRMD*”)

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