



Canadian Chiropractic Association

Chair	Dr. David Peeace, BSc, DC
JCCA STAFF	
Editor	Kent Stuber, DC, MSc Division of Graduate Education & Research Canadian Memorial Chiropractic College, Calgary, Alberta
Editor Emeritus	Allan Gotlib, C.M., DC Toronto, Ontario
Associate Editors	Jeffrey Quon, DC, PhD School of Population & Public Health Faculty of Medicine, University of British Columbia, Vancouver, British Columbia
	André Bussières, DC, FCCS(C), PhD Faculty of Medicine, McGill University, Montréal, Québec Département chiropratique, Université du Québec à Trois-Rivières, Trois-Rivières, Québe
	Dana J. Lawrence, DC, MMedEd, MA Palmer College of Chiropractic, Davenport, Iowa
Assistant Editors	Pierre Côté, DC, PhD University of Ontario Institute of Technology, Oshawa, Ontario
	<b>B.</b> Kim Humphreys, DC, PhD Head of Chiropractic Medicine, Faculty of Medicine, University of Zurich and University Hospital of Balgrist, Zurich, Switzerland
	Gregory N. Kawchuk, DC, PhD University of Alberta, Edmonton, Alberta
	Mohsen Kazemi, RN, DC, MSc, FRCCSS(C), FCCPOR(C) Faculty of Clinical Education, Graduate Studies and Research, Canadian Memorial Chiropractic College, Toronto, Ontario
	Jill Hayden, DC, PhD Department of Community Health & Epidemiology Faculty of Medicine, Dalhousie University, Halifax, Nova Scotia
Production Co-ordinator	Tami Ehrlich
Advertising	Editor, Journal of the Canadian Chiropractic Association 186 Spadina Avenue, Suite 6, Toronto, Ontario M5T 3B2 Tel: 416-585-7902 877-222-9303 Fax: 416-585-2970
	Email: Dr. Kent Stuber <kstuber@jcca.ca> Website: www.jcca-online.org</kstuber@jcca.ca>
TYPESETTING	
	Thistle Printing Limited 35 Mobile Drive, Toronto, Ontario M4A 2P6

## JCCA Journal of the Canadian Chiropractic Association

(Formerly the Canadian Chiropractic Association Journal) Copyright Registered © by the Canadian Chiropractic Association 1961 Copyright: The Canadian Chiropractic Association, 2017

All rights reserved. Without limiting the rights under copyright above, no part of this publication may be reproduced, stored in or introduced into any retrieval system, or transmitted in any form or by any means (electronic, mechanical, photocopying, recording or otherwise), without the prior written permission with the copyright owner and the publisher.

Published by the Canadian Chiropractic Association and issued quarterly

#### EDITORIAL AND EXECUTIVE OFFICES, 186 SPADINA AVENUE, SUITE 6, TORONTO, CANADA M5T 3B2

General Information: The Journal of the Canadian Chiropractic Association is the official quarterly publication by the Canadian Chiropractic Association. The JCCA is published quarterly by the Canadian Chiropractic Association as a medium of communication between the Association and its members and is a forum for fair comment and discussion of all matters of general interest to the chiropractic profession and the Association. Readers are invited to comment and express their opinions on relevant subjects. Views and opinions in editorials and articles are not to be taken as official expression of the Association's policy unless so stated. Publication of contributed articles does not necessarily imply endorsement in any way of the opinions expressed therein and the Journal and its publisher does not accept any responsibility for them. Business correspondence should be addressed to: the Editor of JCCA, 186 Spadina Avenue, Suite 6, Toronto, Canada M5T 3B2.

#### INDEXING SERVICES

JCCA is indexed by PubMed Central, Scopus, CINAHL (Cumulative Index to Nursing and Allied Health Literature), MANTIS (formerly CHIROLARS), AMED, PASCAL, Index to Chiropractic Literature, and selectively by SPORTDiscus.

## Contents

JCCA Vol 61 No 2 ISSN 0008-3194 (Print) and ISSN 1715-6181 (Electronic)

#### **Original** Articles

- 88 Analyzing injuries among university-level athletes: prevalence, patterns and risk factors Jean Lemoyne, PhD Caroline Poulin, DC, MSc Nadia Richer, DC André Bussières, DC, PhD 96 Associations between low back pain and depression and somatization in a Canadian emerging adult population David Robertson, DC, MSc Dinesh Kumbhare, MD, MSc, FRCPC Paul Nolet, DC, MSc, MPH, FRCCSS (C) John Srbely, DC, PhD Genevieve Newton, DC, PhD 106 Systematic review and meta-analyses of the difference between the spinal level of the palpated and imaged iliac crests Robert Cooperstein, MA, DC Felisha Truong 121 Intra-examiner reliability of measurements of ankle range of motion using a modified inclinometer: a pilot study Patricia Tavares, BSc, DC, FCCOS(C) Victoria Landsman, BSc, MA, PhD Leslie Wiltshire, BSc, DC 128 Dorsal scapular nerve neuropathy: a narrative review of the literature Brad Muir, BSc.(Hons), DC, FRCCSS(C) 145 Perianal abscess mimicking levator ani syndrome: a case report and approach to the differential diagnosis of anorectal pain Paul Mastragostino, HBSc.Kin, DC, FCCS (C) Alexander D. Lee, BSc, DC, FRCCSS(C) Patrick J. Battaglia, DC, DACBR
- 153 Innovative application of Cox Flexion Distraction Decompression to the knee: a retrospective case series Luigi Albano, BSc, DC

#### Imaging Case Reviews

162 High-grade spondylolytic spondylolisthesis Peter C. Emary, DC, MSc Stefan A. Eberspaecher, DC John A. Taylor, DC, DACBR

## Contents

JCCA Vol 61 No 2 ISSN 0008-3194 (Print) and ISSN 1715-6181 (Electronic)

A rare case of Eagle syndrome and diffuse idiopathic skeletal hyperostosis in the cervical spine
 Peter C. Emary, DC, MSc
 Marshall Dornink, DC
 John A. Taylor, DC, DACBR

#### **Book Review**

171 The Basic Science of Pain. An Illustrated and Clinically Orientated Guide Peter Stilwell, BKin, DC, MSc

### **Editorial Board**

Alan H Adams, DC Texas Chiropractic College Pasadena, Texas

Kelly E. Donkers Ainsworth, DC, MD, FRCPC Staff Pediatric Radiologist McMaster University Hamilton, Ontario

*Carlo Ammendolia*, DC, PhD University of Toronto

Samuel Bederman, MD, PhD, FRCSC Department of Orthopedic Surgery University of California at Irvine Orange, CA

Paul Bruno, DC, PhD Faculty of Kinesiology and Health Studies University of Regina

Brian Budgell, DC, PhD CMCC

Jason Busse, DC, PhD McMaster University

J David Cassidy, DC, MSc, PhD, FCCS(C), Dr Med Sc University of Southern Denmark

Scott Cheatham, PT, DPT, PhD(C), ATC California State University Dominguez Hills Carson, California.

*Raphael K Chow*, MD, FRCP(C) University of Toronto

Colin M Crawford, B App Sc (Chiro), FCCS(C), MSc, Grad Dip Neuro, MB BS Perth, Australia

*Edward Crowther*, DC, EdD International Medical University Kuala Lumpur, Malaysia

*Diana De Carvalho*, DC, PhD Memorial University St. John's, Newfoundland

Martin Descarreaux, DC, PhD Université du Québec à Trois-Rivières

John A. Dufton, DC, MSc, MD, FRCPC Staff Radiologist University Hospital of Northern British Columbia Prince George, British Columbia

*Peter Emary*, BSc, DC, MSc Cambridge, Ontario

Mark Erwin, DC, PhD University of Toronto

*Brian Gleberzon*, DC, MHSc CMCC

*Richard Goldford*, BSc, DC, MBA, FRCCSS(C), FCCPOR(C) Toronto, Ontario

*Bart Green*, DC, MSEd, DACBSP Naval Medical Center, San Diego San Diego, California

*David Gryfe*, BSc, DC, FRCCSS(C) Etobicoke, Ontario

*François Hains*, DC, FCCS(C), MSc Dorval, Québec

*Scott Haldeman*, DC, MD, PhD, FRCP(C) University of California Irvine, California

*Jill Hayden*, DC, PhD Dalhousie University Halifax, NS

Walter Herzog, PhD University of Calgary

*Thomas E Hyde*, BA, DC, DACBSP N Miami Beach, Florida

*Claire Johnson*, DC, MSEd, DACBSP National University of Health Sciences Lombard, Illinois

Mohsen Kazemi, RN, DC, FRCCSS(C), FCCPOR(C), MSc, PhD(c) CMCC

Clark R Konczak, MSc, DC, DABCO, FCCO(C), FRCCSS(C) Victoria, BC

Deborah Kopansky-Giles, DC, FCCS(C), FICC, MSc St. Michael's Hospital Toronto, Ontario

*Doug M Lawson*, BA, DC, MSc D'Youville College

*Cynthia Long*, PhD Palmer Centre for Chiropractic Research Davenport, Iowa

*William C Meeker*, DC, MPH Palmer Chiropractic University System San Jose, CA

*Michelle Mick*, DC, DACBR St. Paul, Minnesota

Silvano Mior, DC,FCCS(C), PhD CMCC

*Robert D Mootz*, DC Associate Medical Director for Chiropractic, State of Washington Department of Labor and Industries Olympia, WA *Bernadette Murphy*, DC, PhD University of Ontario Institute of Technology

Martin Normand, DC, PhD UQTR

Jason Pajaczkowski, BSc, BS, DC, FRCCSS(C), FCCPOR(C) CMCC

*John Papa*, DC, FCCPOR(C) New Hamburg, Ontario

Steven Passmore, DC, PhD Faculty of Medicine University of Manitoba

Stephen Perle, DC, MS University of Bridgeport Bridgeport, CT

*Reed B Phillips*, DC, PhD, DACBR Southern California University of Health Sciences

Mathieu Piché, DC, PhD UQTR

John J Riva, DC, MSc Department of Family Medicine McMaster University Hamilton, Ontario

Sandy Sajko, DC, MSc, RCCSS(C) Oakville, Ontario

John Z Srbely, DC, PhD University of Guelph

*Brynne Stainsby*, BA, DC, FCCS(C) CMCC

*Igor Steiman*, MSc, DC, FCCS(C) CMCC

John S Stites, DC, DACBR Palmer College of Chiropractic Davenport, Iowa

John A M Taylor, DC, DACBR, FCCR(C) D'Youville College Buffalo, NY

Haymo Thiel, DC, MSc (Orth), FCCS(C), Dip Med Ed, PhD Anglo-European College of Chiropractic Bournemouth, England

*Gabrielle M van der Velde*, BSc, DC, FCCS(C), PhD Toronto Health Economics and Technology Assessment Collaborative University of Toronto

Marja J Verhoef, PhD University of Calgary

## Analyzing injuries among university-level athletes: prevalence, patterns and risk factors

Jean Lemoyne, PhD<sup>1</sup> Caroline Poulin, DC, MSc<sup>2</sup> Nadia Richer, DC<sup>2</sup> André Bussières, DC, PhD<sup>2,3</sup>

Background: Scientific evidence suggests many health benefits are associated with sport participation. However, high intensity participation may be related to an increased risk of musculoskeletal injuries.

Objectives: This study aims to: 1) describe the prevalence and patterns of sports injuries, and 2) identify its associated risk factors.

Methods: A cross-sectional design was used. University level athletes, involved in 7 sport disciplines reported musculoskeletal injuries sustained in the past year, as well as potential risk factors: training volume and antecedent sport participation. Group comparisons were conducted.

Results: 82 athletes participated in the study. Respondents sustained over two injuries per year. Significant differences were found for sport category and type of injury. No differences were observed regarding antecedent sport participation.

Discussion: High prevalence and sport-specific

Contexte : Les observations scientifiques laissent entendre que de nombreux avantages pour la santé sont associés à la participation sportive. Cependant, la participation à haute intensité pourrait être associée à un risque accru de blessures musculo-squelettiques.

Objectifs : Cette étude vise : 1) à décrire la prévalence et les tendances en matière de blessures sportives et 2) à en déterminer les facteurs de risque connexes.

Méthodologie : On a utilisé une analyse transversale. Des athlètes universitaires qui participaient à sept disciplines sportives ont déclaré des blessures musculosquelettiques au cours de la dernière année, ainsi que des facteurs de risque potentiels : le volume de l'entraînement et l'historique de participation sportive. On a réalisé des comparaisons de groupes.

Résultats : En tout, 82 athlètes ont participé à l'étude. Les répondants ont subi plus de deux blessures par année. On a constaté d'importantes différences sur le plan de la catégorie sportive et du type de blessure. On

<sup>1</sup> Université du Québec à Trois-Rivières, Department of Human Kinetics

<sup>2</sup> Université du Québec à Trois-Rivières, Department of Chiropractic

<sup>3</sup> McGill University, School of Physical and Occupational Therapy, Faculty of Medicine

The authors declare no conflict of interest related with this project. Authors also confirmed that no funding was obtained to conduct this research.

Corresponding author: Jean Lemoyne Université du Québec à Trois-Rivières, Department of Human Kinetics, 3351, Boulevard des Forges, CP.500. Trois-Rivières (Québec) G9A 5H7 E-mail: jean.lemoyne@uqtr.ca Tel: 819-376-5011 ext. 3794 Fax: 819-376-5092

© JCCA 2017

injuries observed in university sport should be of concern to athletes, therapists, coaches and sport organizers.

Conclusion: This study contributed to a better knowledge of injury patterns among university athletes, and suggests further practical and research implications. n'a pas observé de différence concernant l'historique de participation sportive.

Discussion : La prévalence élevée et les blessures propres au sport observées dans le sport universitaire devraient préoccuper les athlètes, thérapeutes, entraîneurs et organisateurs d'événements sportifs.

Conclusion : Cette étude a contribué à une meilleure connaissance des tendances en matière de blessures chez les athlètes universitaires et laisse entendre d'autres répercussions sur le plan concret et de la recherche.

(JCCA. 2017;61(2):84-95)

(JCCA. 2017;61(2):84-95)

KEY WORDS: musculoskeletal injuries, college sport, prevalence, risk factors, injury prevention, chiropractic

#### Introduction

A large body of evidence supports the health benefits of participation in sport.<sup>1</sup> However, research also reveals that the excessive practice of sport, especially in the competitive context, is associated with an increased risk of musculoskeletal injuries.<sup>2,3</sup> Sport injuries are associated with high direct and indirect costs, and can lead to early sport retirement for up to 24% of athletes.<sup>4</sup> Sport injury may also lead to decreased sport participation and associated all-cause morbidity, overweight/obesity and post-traumatic osteoarthritis.<sup>5</sup> A cross-sectional study of 236 young elite athletes suggested an injury prevalence rate three to five times higher than the general population.<sup>6</sup> Multiple mechanisms contribute to explaining the high prevalence of musculoskeletal injuries among competitive level athletes.7 According to Almeida8, training volume is an important risk factor that was associated with sport injuries. Specifically, the number of hours of vigorous training was significantly correlated with the presence of musculoskeletal overuse injuries. Furthermore, high training regimes also appear to increase the risk of sustaining acute injuries in high contact team sports such as rugby.9

Not surprisingly perhaps, antecedent competitive level sport participation may be associated with an increased prevalence of musculoskeletal injuries. According to Malina<sup>10</sup>, antecedent sport participation is defined as being involved in systematic training in a single sport at a relaMOTS CLÉS : blessures musculo-squelettiques, sport collégial, prévalence, facteurs de risque, prévention des blessures, chiropratique

tively young age. Moderate to high levels of sport participation usually occurs at puberty, in particular within North American sport systems. Caine et al.<sup>11</sup> demonstrated that during adolescence, taking part in elite sports and specializing in a single sport were both associated with a higher prevalence of injuries during those years. For instance, Hall and colleagues showed that the risk of developing anterior knee pain was four times higher among "early specialized" female basketball, soccer and volleyball players.<sup>12</sup> However, despite the Hall study, little is known about the long-term effects of antecedent sport participation from an epidemiological perspective. In this regard, prior research suggests there is a need to improve our understanding of acute and overuse injury patterns among high performing athletes. Investigating the relationship between sport injuries and its potential risk factors is relevant from a health promotion perspective.<sup>5</sup> Collecting data about injury patterns and related risk factors can inform the design of novel strategies to prevent injuries among athletes.

The purpose of the current study was to analyze the injury profile of university-level athletes involved in multiple sport disciplines. This study aimed to 1) describe and compare the injury profiles (e.g. prevalence and distribution) of seven sport disciplines, and 2) determine if reported injuries (acute or overuse) were associated with two risk factors: training volume and early sport specialization (i.e., antecedent sport participation).

#### Methods

A cross-sectional study was administered between November 2014 and January 2015 among university level athletes (50% males (M), 50% females (F), aged between 21 and 29 years old). All athletes (N=120) involved in the university's sport programs were invited to participate in the study. Participants came from seven university-level sports: cross-country running (M and F), cheerleading, golf (M), ice hockey (M), soccer (M and F), swimming (M and F), and volleyball (F). We decided to regroup athletes who were involved in the same sports, resulting in six groups. The institution's research ethical board (CER-14-204-07-25) approved this project. The athletes who agreed to take part in the data collection received an information letter and completed a consent form.

Participants completed an online questionnaire (appendix 1). Questions were developed based on prior studies and antecedent recommendations.<sup>13,14</sup> Three experts assessed the survey questionnaire for face and content validity and a pilot version was tested on five users. The first part of the questionnaire consisted of three questions covering demographic information (age, gender, hand dominance). Secondly, participants were asked to self-report any prior injuries (over lifetime) at the onset of their competition season. Injury was defined in the questionnaire as "a physical complaint or observable damage to body tissue produced by the transfer of energy experienced or sustained by an athlete during participation on athletic training or competition regardless of whether it received medical attention or its consequence with respect to impairments in connection with competition or training".13 For each reported injury; participants were asked to specify the date, specific site, tissue involved, type of injury, possible cause, associated factors, type and duration of invalidity and time to recovery. A maximum of ten "past injuries" could be reported. Preliminary data analyses revealed that none of the participants reported over six "lifetime" injuries. Data collection was facilitated by the presence of a chiropractic intern paired with each team. The last section of the questionnaire measured potential risk factors including training volume and antecedent sport participation.

Training volume was measured using two indicators:

duration and frequency of training sessions. For duration of training, participants had to report the total number of hours per week they invested in training (sport-specific, physical preparation, competition) since the beginning of the season. For the frequency of training, participants had to report the number of weekly sessions they were involved in their sport (training, competition, etc.).

Antecedent sport participation was measured using two items. Participants were asked, "Between age 13 and 18, did you take part in other elite-competitive sports other than the one you are involved in your university team?" We also asked the age at which they started their university team sport.

For descriptive purposes, we calculated the frequency and proportional distribution of injuries by participant characteristics and training volume. Training volume was subdivided into the number of weekly training sessions ( $\leq$ 3, between 4 and 7, and more than 7 sessions per week) and the number of hours of training per week (between 5-10, between 10-15, and > 15 hours). Antecedent sport participation was sub-divided into two categories: participants who took part in sports other than their current discipline between the age of 13 and 18 versus those who did not, meaning no antecedent competitive sports.

First, we verified for distribution normality concerning training volume (duration and frequency), and self-reported injuries. Normal probability plots and verification for excessive skewness / kurtosis were conducted to verify for normality assumptions. In the present case, no excessive values were observed (skewness values between -0.07-0.93, and kurtosis values between -0.11-0.91), but significant values for Wilk-Shapiro normality test, suggest a slight violation of normality assumptions. We conducted ANOVAs with Monte Carlo randomization<sup>15</sup> to verify if training volume and number of injuries differ regarding each team. Then, we compared the injury profile by verifying if the injury profile differed for three risk factors: weekly training sessions, hours of training per week (or training volume) and antecedent sport participation. In cases with a significant F value, we conducted post hoc tests to identify specific group level differences, by using Tukey's HSD test. The mean number of injuries per athlete and prevalence rate (with 95% confidence intervals) was calculated for each sport discipline. We calculated the lifetime injury incidence rate (injury rate per athlete-lifetime). The injury incidence rate was calculated with 95%

Teams	Total (N=81)	Cross country (n=15)	Cheerleading (n= 11)	Ice Hockey (n=22)	Soccer (n=20)	Swimming (n=5)	Volleyball (n=8)	Test statistic (df)	P value
Gender, males (%)	40 (49)	5 (33)	2 (18)	22 (100)	9 (45)	2 (40)	0 (0)	Descriptiv	e only
Age (M±SD)	24.0 ± 1.9	$24.6\pm2.4$	$23.7\pm2.3$	$24.1\pm0.8$	23.1 ± 1.4	$25.2 \pm 2.4$	$24.9\pm2.6$	$F(_{5,75}) = 2.0$	0.08
Duration (Hours / week)	12.3 ± 5.4*	11.5 ± 4.1	9.1 ± 3.1	$16.9 \pm 5.4$	10.5 ± 2.8	15.8 ± 8.7	7.7.±2.2	$F_{(5,73)} = 9.1$	< .001
Frequency (Sessions / week)	4.9 ± 2.9**	7.1 ± 2.9	$2.9 \pm 1.4$	$6.7 \pm 2.8$	$3.0 \pm 0.9$	$6.8 \pm 3.7$	$2.7 \pm 1.2$	$F_{(5,73)} = 13.1$	< .001
Sessions (Hours / session)	3.2 ± 1.9	1.9 ± 1.4	4.5 ± 3.3	$2.9 \pm 1.4$	3.7 ± 1.0	$2.2 \pm 0.8$	$3.5 \pm 2.7$	$F_{(5,73)} = 5.9$	.002
Injuries (% of total)	185 (100) §	21 (11)	28 (16)	47 (25)	58 (31)	12 (6)	19 (11)	$\chi^2_{(5)} = 53.4$	< .001
Lifetime injury rate per athlete (95% CI)	2.26 (1.9-2.6)	1.47 (0.9-2.2)	2.55 (1.7-3.7)	2.01 (1.5-2.7)	2.9 (1.5-3.8)	2.20 (1.2-3.9)	2.38 (1.5-3.7)	$F_{(5,73)} = 1.2$	0.27
Lifetime prevalence (%)	55 (67.1)	6 (22.2)	8 (29.6)	15 (55.5)	17 (63.2)	4 (14.8)	4 (15.4)	$\chi^2_{(5)} = 17.8$	0.01

Table 1.Participants' training volume and injury profile(total injuries, mean, lifetime prevalence and lifetime injuries per athlete)

<sup>§</sup> 2 injuries were reported by the golf player (not included for the analyses).

confidence intervals for Poisson rates, as the number of injuries is typically not normally distributed. The lifetime prevalence (overall prevalence) was defined as the total number of athletes injured divided by the total size of each subgroup. Secondly, we compared injury prevalence, to see if acute injuries were more prevalent than overuse injuries. Self-reported injuries were categorized in two subgroups: acute and chronic (or overuse) based on Fuller *et al.*<sup>16</sup> who define acute injuries as traumas resulting from a single, identifiable event, and chronic-overuse injuries, which correspond to gradual-onset injuries caused by repeated trauma. We then calculated chi-square ( $\chi^2$ ) statistic on a 2x2 crosstab: injury type x sport type.

Finally, we computed the mean injuries (with 95% confidence intervals) per athlete for four risk factors: 1) weekly hours (treated as continuous variable), 2) weekly sessions (treated as continuous variable), and 3) antecedent sport participation (2 levels). Due to the distribution of self-reported injuries, a Poisson regression model analysis was performed to identify significant predictors and to estimate their associated incidence risk ratio. Training volume in terms of hours and weekly sessions were included as predictors for the incidence rate ratios (IRRs) which were obtained by exponentiation of the regression coefficients. Due to the potential biased estimates which can be obtained due to unequal sample sizes on sport type

 $(n_{individual} = 21, n_{team} = 61)$ , we accounted for the sport type effect by adding this variable as a clustering variable.

#### Results

Table 1 presents the characteristics of the sample population and training volume for each sport discipline. Eightytwo university-level athletes, (50% male (n=41)) returned a completed survey questionnaire, corresponding to a response rate of 68%. Non-response (N= 38, 32% of total population) may be explained by multiple factors, such as refusal to take part to the study, or simply athletes not opening the invitation message sent by the research staff. Respondents had a mean age of  $24.0 \pm 1.9$  (range: 21-29 years) and were involved in seven sport disciplines: cross-country running (n=15), cheerleading (n=11), golf (n=1), ice hockey (n=22), soccer (n=20), swimming (n=5), and volleyball (n=8). Due to a low response rate (n=1) from the men's golf team, this sport category was excluded from the analyses. Moreover, 41% of participants reported not having taking part in other activities than their specific sport disciplines between the age of 13 and 18.

On average, across the six sport disciplines, athletes trained 12.3 ( $\pm$  5.4) hours per week, and performed an average of 4.95 ( $\pm$  2.9) sessions per week. Sessions lasted on average 3.2 ( $\pm$  1.9) hours. In summary, post-hoc an-

	Total (%)	Individual sports (% of total)	Team sports (% of total)
Concussion	24 (12)	4 (2)	20 (11)
Sprain	67 (39)	10 ( 6)	57 (33)
Muscle tear	30 (17)	13 (7)	17 (10)
Fracture	27 (16)	9 ( 6)	18 (10)
Dislocation / luxation	5 (3)	1 ( 1)	4 (2)
Acute injuries (total)	153 (87)	37 (76)	116 (92) §§
Overuse injuries (total) (Tendonitis, Bursitis, Shinsplints)	22 (13)	12 (24) §	10 ( 8)

Table 2.Types of injuries by sport category<br/>(for descriptive purposes)

\*\*p < 0.01

2 injuries were reported by the golf player (not included for the analyses). A total of 175 injuries (out of 185) are reported (10 missing data for injury type).

alyses revealed that volleyball players and cheerleading participants were involved in fewer training hours than the other groups ( $F_{(5,73)} = 9.12$ , p < .001). Group comparisons also revealed significant group differences regarding weekly sessions ( $F_{(5,73)} = 8.01$ , p < .001), in favor of ice hockey, swimming and cross-country participants. Number of hours per session were significantly lower among cheerleading participants ( $F_{(5,73)} = 5.92$ , p =.002). Despite a significantly higher number of injuries reported by soc-

cer and hockey players ( $\chi^2 = 53.4$ , p < .001) and on lifetime prevalence ( $\chi^2 = 17.8$ , p = .01), no significant differences were observed regarding the average number of lifetime injury rate per athlete ( $F_{(5,73)} = 1.23$ , p = .27). In summary, participants reported an average of 2.28 injuries per athlete; with a prevalence of 91% (only 7 athletes reported having no injuries). The overall "lifetime" prevalence of injuries (one injury or more) was 67.1%.

As shown on Table 2, 185 musculoskeletal injuries were reported, suggesting an average of more than two injuries per athlete (mean= 2.3; 95% CI 2.14-2.36). The distribution of injuries was as follows: ankle (20%), knee (15%), shoulder (13%), head (11%) and back (7%). Specific patterns of injury type and sport disciplines were also observed. Acute injuries were predominant, representing 87% of all reported injuries. Sixty-six percent of acute or traumatic injuries (sprain, facture, etc.) occurred during team sports, whereas overuse injuries (tendonitis, etc.) were slightly more common in individual sports (54% versus 46%). Participants involved in team sports were more affected by acute injuries (92% versus 76%). Conversely, those in individual sports reported a significantly higher level of overuse injuries (24% versus 8%). These results will be addressed in more detail in the discussion section.

For the second objective of the study, results from the

Table 3.	
Multivariate estimates of IRR and 95% CI a	adjusted for injuries per athlete.

Risk factors	Reported injuries	Mean (95% CI) injuries per athlete	Pseudo R <sup>2</sup>	IRR <sup>a</sup> (95% CI) injuries	Wald $\chi^2_{(df)}$	р
<sup>b</sup> Training sessions (n) ≤ 3 (n=36) 4 - 7 (n=26) ≥ 8 (n==18)	89 60 36	2.44 (1.97 - 2.92) 2.35 (1.79 - 2.90) 2.00 (1.33 - 2.67)		0.96 (0.92 - 1.02)	13.33 (10)	0.21
<b><sup>b</sup>Training (hours)</b> ≤ 10 (n= 36) 10-15 (n= 37) > 15 (n= 7)	79 95 16	2.19 (1.69 - 2.64) 2.51 (2.05 - 2.98) 2.00 (0.93 - 3.07)		1.00 (0.96 - 1.05)	22.72 (19)	0.25
Antecedent sport participation Yes (n = 33) No (n = 49)	92 95	2.36 (1.75 - 2.74) 2.24 (1.95 - 2.78)		1.06 (0.92 - 1.13)	3.57 (1)	0.06
<b>Sport type</b> Team (n=61) Individual (n=20)	133 52	2.46 (2.07 - 2.84) 1.67 (1.16 - 2.17)				

CI = Confidence interval; IRR = Incidence rate ratio; df = degrees of freedom

<sup>a</sup> Coefficients are on an exponential scale (exp[ $\beta$ ] = IRR); 95% Bootstrap Confidence Interval.

<sup>b</sup> Training volume (sessions and hours) was treated as a continuous variable

Poisson regression analysis suggested that when sport type was controlled as a clustering effect, the occurrence of injury was not associated with either the number of weekly training sessions (Wald  $\chi^2$  (df) = 13.33 (1), p = .21), training volume in terms of weekly hours (Wald  $\chi^2$ (df) = 22.72 (19), p = .25), and antecedent sport participation (Wald  $\chi^2$  (df) = 3.58 (1), p = .06). Table 3 presents the incidence rate ratios (IRRs) and associated 95% confidence intervals (CIs) for each variable.

#### Discussion

The objective of this study was to determine the injury profile in a population of university-level competitive athletes. Awareness of the injury profile and of risk factors among university level athletes may help develop strategies to reduce the risk of injuries. Our results indicated that injuries appear to be common among university-level athletes, with an average of over two injuries per athlete per year. Such high prevalence should be of concern to athletes, coaches and sport organizers. Ankle, knee and shoulder injuries were the most frequently affected areas, similar to what has been reported elsewhere.<sup>8</sup>

This study also suggested that the injury profile differs according to type of sport, with acute injuries being more common in team sports. However, the present investigation cannot specifically establish cause and effect relationships to explain the athletes' injuries. Such preoccupation was beyond the scope of this investigation, and large samples, systematic follow-ups, and multiple sport samples would have been necessary to establish causal relationships of injury patterns. Several factors may explain our findings. First, contacts with opponents and/or teammates are common in team sports such as ice hockey and soccer.<sup>17</sup> Collisions with teammates are also frequently reported in volleyball, especially when players fall near the net following an attack.<sup>18</sup> Acute injuries may also be caused by ball contact (e.g. serve and smash reception, blocks). In cheerleading, acute injuries can result from falling on the floor after being thrown by teammates. In this regard, it is plausible to suggest that the nature of sport, by its exposure to contact may be a factor that could explain differences between athletes' injury patterns. For example, athletes involved in individual sports (and low risk of contact) undergo a highly repetitive training regime, which is a factor that could possibly explain the higher level of overuse injury.<sup>19</sup> However, it is important to specify that the small sample size for individual sports and the limited number of individual contact sports (e.g. boxing, taekwondo, etc.) are a limitation of this study. Furthermore, the relatively small sample size prevents us from drawing conclusive interpretations about sport type differences. Future research designs should try to recruit among a larger pool of athletes, who participate in a wider range of Canadian, university-level sport disciplines. In summary, further research should increase sample size and therefore expand the number of sports to draw conclusions that are more generalizable.

Together, these findings suggest stakeholders (therapists, trainers, coaches, etc.) should consider tailoring injury prevention plans. Reviews and meta-analyses of effective interventions to reduce the risk of sports-related injuries suggest prevention plans may include insoles (Odd Ratio [OR]: 0.51, 95% CI 0.32-0.81), external joint supports (OR: 0.40, 95% CI 0.30-0.53), and specific training programs (OR: 0.55, 95% CI 0.46-0.66)<sup>20</sup>. Further, the pooled estimate of randomized controlled trials examining a preventative effect of neuromuscular training in the reduction of lower extremity injuries in youth team sport (soccer, European handball, basketball) demonstrates a significant overall protective effect injury (incidence rate ratio: IRR=0.64 (95% CI 0.49-0.84) or a 36% reduction in lower extremity injury risk.<sup>21</sup> Clearly, much can be done to reduce injury risks in young athletes.

The present study also tried to shed some light on the possible associations between training volume (number of sessions and hours trained per week), antecedent sport participation and the participants' injuries. While our findings failed to reveal significant associations, possibly because of the low sample size, other studies suggest that athletes exposed to a higher training load, including high intensity, and high number of hours of training, are facing a higher risk of injury.<sup>22,23</sup> Little is known about the longterm consequences of antecedent sport participation and the development of overuse injuries.12 Use of more specific measures of early specialization, instead of antecedent sport participation (as a potential risk factor) may contribute to delineating possible risks. For instance, number of antecedent sport disciplines, amount of training per age group, and competitive level and number of years in the specialized and non-specialized sport disciplines may be factors to consider.

This study contributed to enhancing knowledge on multiple facets related to the field of sport traumatology. Our results suggest further investigation about the potential confounding effect of contact, and propose investigating among a larger pool of athletes. From this perspective, it would be relevant in future research designs to identify individual sport disciplines, which could be categorized as contact sports and therefore explain more precisely injury patterns. To our knowledge, this is the first study to provide a detailed portrait of the athletes within their teaching institution. From this perspective, our study suggests the need for coaches to consider recommending regular medical follow-ups of athlete's and injury prevention programs. Further research should focus on a larger scale data collection to have a more detailed picture of university athletes' injury profile.

The data collection procedures used for this investigation contributed to gathering relevant information related to athletes' injuries, and contributed to helping health care team in their interventions (e.g. prevention, follow-ups). The questionnaire was adapted to the needs of our stakeholders (coaches, athletes, medical staff and researchers) wishing to more effectively monitor athlete's progress.

Despite its strengths, this study has several limitations. First, our sample was relatively small. This constraint limits generalization of the results to other athletic populations, and to other universities' sports programs. A larger scale study, involving other provincial varsity sport programs would allow for a more complete description of Quebec's university athletes' profile. Second, self-report measures have the potential of recall bias, which can influence the results. Antecedent research showed that recall was associated with multiple challenges in a 12-month recall period among Australian football players.<sup>24</sup> In study recall bias was very likely, and participants may have reported only what they perceived as their most important injuries, resulting in a biased estimation of their lifetime injuries. To reduce such potential limitations, further studies should consider the feasibility of prospective study designs, an approach that would provide more valid estimates of antecedent injuries. Third, our categorization of sport injuries lacked specificity. More detailed injury records about each athlete would have contributed to estimating their injury patterns more precisely, which could have provided a better explanation of their mechanisms. Moreover, using a standardized injury classification tool (e.g. the Orchard Sport Injury Classification<sup>25</sup>, the Sport Medicine Coding System<sup>26</sup>) would also have provided a more detailed profile for each athlete. Fourth, measuring antecedent sport participation using a single item may not properly capture the essence of the domain. Last, as with any cross-sectional design, the causative aspects remain untested. We cannot rule out the possibility that reported injuries caused risk factors (e.g., reduced number of training sessions or hours per week and antecedent sport participation) or that another unmeasured variable is the potential cause of both risk factors and injuries. Future studies should prospectively explore the incidence rate and injury risk factors among competitive sport athletes across several universities and other competitive sports outside of the collegiate system.

#### Conclusion

Injuries are common among university-level athletes. This study revealed that competitive athletes sustained on average more than two injuries each year, with ankle, knee and shoulder injuries being most frequently reported. Further, the injury profile appears to differ according to type of sport, with acute injuries being more common in team sports and overuse injuries in individual sports. The high prevalence of injuries among university-based athletes should be of concern to athletes, coaches and sport administrators as all have an important role to play in designing tailored injury prevention programs. Nonetheless, further studies should aim to include athletes from multiple varsity sport university programs, and consider prospective research designs to provide a fuller picture of the injury profile among these athletes.

#### Practical Implications

- This study suggests that the high prevalence of injuries is a major issue among university level athletes, especially acute injuries.
- Stakeholders should consider that sport injury mechanisms (and injury profile) differ between sport disciplines. Further analyses are needed among other sport disciplines.
- A greater understanding of sport-specific injury mechanisms may help reduce the risk of injuries among university athletes.

#### Acknowledgements

The authors gratefully acknowledge Ms. Laura Mendoza, for her assistance with recruiting participants, data collection and descriptive analysis and Ms. Heather Owens for her assistance with the manuscript. Authors also wish to acknowledge each UQTR's athletic team medical staff for encouraging athletes to take part to the study.

#### References

- 1. Haskell WL, Lee IM, Pate RR, *et al.* Physical activity and public health: updated recommendation for adults from the American College of Sports Medicine and the American Heart Association. Med Sci Sport Exer. 2007; 39(8):1423-1434.
- Alonsa J, Junge A, Renstrom P, *et al.* Sports injuries surveillance during the 2007 IAAF world athletics championships, Clin J Sport Med. 2009; 19(1): 26-31.
- Hamel D, Tremblay B, Nolin B, et al. (2012). Étude des blessures subies au cours de la pratique d'activités récréatives et sportives au Québec en 2009-2010 [Analysis of Quebec's sport and leisure injuries : 2009-2010]. Retrieved online: http://rseq.ca/media/844914/ etudeblessuresrecreasportqc2009-2010.pdf
- Steffen K, Engebretsen L. More data needed on injury risk among young elite athletes. Br J Sport Med. 2010; 44(7): 485-489.
- Richmond S, Fukuchi R, Ezzat A, *et al*. Are joint injury, sport activity, physical activity, obesity, or occupational activities predictors for osteoarthritis? A systematic review. J Ortho Sport Phys Ther. 2013; 43(8):515–524.
- 6. Van Hilst J, Hilgersom NF, Kuilman MC, *et al.* Low back pain in young elite field hockey players, football players and speed skaters: Prevalence and risk factors. J Back Musculoskelet Rehabil. 2015; 28(1): 67-73.
- 7. Yang J, Tibbetts A, Covassin T, *et al*. Epidemiology of oversuse and acute injuries among competitive collegiate athletes. J Athl Training. 2012; 47(2): 198-204.
- Almeida SA, Williams KM, Shaffe, RA, *et al.* Epidemiological patterns of musculoskeletal injuries and physical training. Med Sci Sport Exer. 1999; 31(8): 1176-1182.
- 9. Brooks JH, Fuller CW, Kemp SP, *et al*. An assessment of training volume in professional rugby union and its impact on the incidence, severity, and nature of match and training injuries. J Sport Sci. 2008; 26(8): 863-873.
- Malina RM. Early sport specialization: roots, effectiveness, risks. Curr Sport Med Rep. 2010; 9(6): 364-371.
- 11. Caine D, Maffulli N, Caine C. Epidemiology of injury in

child and adolescent sports: injury rates, risk factors, and prevention. Clin J Sport Med. 2008; 27(1): 19-50.

- Hall R, Foss KB, Hewett TE, *et al.* Sports specialization is associated with an increased risk of developing anterior knee pain in adolescent female athletes. J Sport Rehabil. 2015; 24(1): 31-35.
- Timpka T, Alonso JM, Jacobsson J, *et al.* Injury and illness definitions and data collection procedures for use in epidemiological studies in Athletics (track and field): Consensus statement. Br J Sport Med. 2014; 48: 484-490.
- Kerr Z, Dompier R, Snook E, *et al.* National collegiate athletic association injury surveillance system: review of methods for 2004-2005 through 2013-2014 data collection. J Athl Training. 2014; 49(3): 552-560.
- 15. Edgington, ES. Randomizatrion tests. 1980; New York : M Dekker.
- Fuller CW, Ekstrand J., Junge A, *et al.* Consensus statement on injury definitions and data collection procedures in studies of football (soccer) injuries. Scand J Med Sci. Sports 2006; 16(2): 83-92.
- 17. Emery CA, Meeuwisse WH, McAllister JR. Survey of sport participation and sport injury in Calgary and area high schools. Clin J Sport Med 2006; 16(1): 20-26.
- Verhagen EALM, Van der Beek AJ, Bouter LM, *et al.* A one season prospective cohort study of volleyball injuries. Br J Sports Med. 2004; 38: 477-481.
- Renström P, Johnson RJ. Oversuse injuries in sports. Sports Med. 1985; 2(5): 316-333.
- 20. Leppänen M, Aaltonen S, Parkkari J, *et al.* Interventions to prevent sports related injuries: a systematic review and meta-analysis of randomised controlled trials. Sports Med. 2014; 44(4):473-486.
- Emery CA, Roy TO, Whittaker JL, *et al.* Neuromuscular training injury prevention strategies in youth sport: a systematic review and meta-analysis. Br J Sport Med. 2015; 49(13):865-870.
- 22. Whittaker JL, Small C, Maffey L, *et al*. Risk factors for groin injury in sport: an updated systematic review. Br J Sport Med. 2015; 49(12):803-809.
- 23. Jacobsson J, Timpka T, Kowalski J, *et al*. Injury patterns in Swedish elite athletics: annual incidence, injury types and risk factors. Br J Sport Med. 2013; 47(15):941-952.
- 24. Gabbe BJ, Finch CF, Bennell KL, Wajswelner H. How valid is a self-reported 12-month sports injury history? Br J Sport Med. 2003; 37(6): 545-547.
- Rae K, Orchard J. The orchard sports injury classification system (OSICS) version 10. Clin J Sport Med. 2007; 17(3): 201-204.
- Meeuwisse WH, Preston Wiley J. The sport medicine diagnostic coding system. Clin J Sport Med. 2007; 17(3): 205-207.

## Associations between low back pain and depression and somatization in a Canadian emerging adult population

David Robertson, DC, MSc<sup>1</sup> Dinesh Kumbhare, MD, MSc, FRCPC<sup>2</sup> Paul Nolet, DC, MSc, MPH, FRCCSS (C)<sup>3</sup> John Srbely, DC, PhD<sup>4</sup> Genevieve Newton, DC, PhD<sup>5</sup>

Introduction: *The association between depression, somatization and low back pain has been minimally investigated in a Canadian emerging adult population.* 

Methods: 1013 first year Canadian university students completed the Modified Zung Depression Index, the Modified Somatic Perception Questionnaire, and a survey about low back pain frequency and intensity. Multinomial logistic regression was used to measure associations between low back pain and depression and somatization, both independently and co-occurring.

Results: Over 50% of subjects reported low back pain across grades, and both depression and somatization were significantly positively associated with low back Introduction : L'association entre la dépression, la conversion et la lombalgie a fait l'objet de très peu d'études au sein d'une population adulte émergente canadienne.

Méthodologie : Au total, 1 013 étudiants universitaires canadiens de première année ont rempli l'indice de dépression de Zung modifié, le questionnaire de perception somatique modifié et un sondage sur la fréquence et l'intensité de la lombalgie. On a utilisé la régression logistique multinominale pour mesurer les associations entre la lombalgie, la dépression et la conversion, tant de façon indépendante que cooccurrente.

Résultats : *Plus de 50 % des sujets ont déclaré de la lombalgie sur plusieurs stades et tant la dépression que la conversion étaient associées de manière positive* 

<sup>1</sup> Royal College of Chiropractic Sports Sciences (C) Resident

Corresponding author: Genevieve Newton Department of Human Health and Nutritional Science, University of Guelph, Guelph, ON E-mail: <u>newton@uoguelph.ca</u> Tel: 519-824-4120 x56822

The study was approved by the Research Ethics Board at the University of Guelph. The authors have no disclaimers to make.

<sup>&</sup>lt;sup>2</sup> Department of Medicine, University of Toronto, Toronto ON

<sup>&</sup>lt;sup>3</sup> Department of Graduate Education and Research, Canadian Memorial Chiropractic College, Toronto ON

<sup>&</sup>lt;sup>4</sup> Department of Human Health and Nutritional Science, University of Guelph, Guelph, ON

<sup>&</sup>lt;sup>5</sup> Department of Human Health and Nutritional Science, University of Guelph, Guelph, ON

pain. Several positive associations between the cooccurrence of somatization and depression with various grades of low back pain were observed.

Discussion: These results suggest that low back pain, depression and somatization are relatively common at the onset of adulthood, and should be considered an important focus of public health.

#### (JCCA. 2017;61(2):96-105)

KEY WORDS: chiropractic, low back pain, depression, association, somatization

et étroite à la lombalgie. On a observé plusieurs associations positives entre la cooccurrence de la conversion et de la dépression et divers stades de lombalgie.

Discussion : Ces résultats laissent entendre que la lombalgie, la dépression et la conversion sont relativement communes au début de l'âge adulte et doivent représenter un facteur important en santé publique.

#### (JCCA. 2017;61(2):96-105)

MOTS CLÉS : chiropratique, lombalgie, dépression, association, conversion

#### Introduction

Low back pain is one of the most common musculoskeletal conditions worldwide, with a lifetime prevalence of 84.1%<sup>1</sup>. LBP is the leading cause of years lived with disability<sup>2</sup> and is associated with worse health-related quality of life<sup>3</sup>. This prevalence has been widely investigated in studies across a variety of populations, including adolescents and adults. However, fewer studies have specifically investigated the prevalence of back pain during "emerging adulthood", particularly in a Canadian population, which is typically defined as ages 18 to 26.4 It is during this time when individuals experience an increased amount of responsibility and independence with respect to their lifestyle choices, and begin to establish long-lasting behaviours that are associated with long-term health risks.5 Importantly, while behaviours during this period may be linked to chronic disease later in life, they may be amenable to change.5 For this reason, evaluation of low back pain and associated risk factors during the emerging adult period is warranted, and may highlight viable targets of behavioural and clinical interventions.

Consideration of psychological factors may be particularly relevant in the emerging adult population, who experience high rates of depression.<sup>6,7</sup> Depression has been shown quite consistently to be associated with back pain in the general adult population<sup>8,9</sup>, although research looking specifically at the association between back pain and depression in emerging adults is limited and equivocal, and to our knowledge, this association has not been investigated in a Canadian context. For example, in a study of 973 male and female college students in the United States by Kennedy *et al.*<sup>10</sup>, psychological factors like feeling sad, exhausted, and overwhelmed were directly associated with the prevalence of low back pain. Similarly, Unalan *et al.*<sup>11</sup> found that scores on the Zung Depression Index were associated with back pain in their study of 250 Turkish vocational students. In contrast, a study of 170 female nursing students in Australia by Mitchell *et al.*<sup>12</sup> found that there were no differences in depression scores between low back pain and control students. While it seems likely that back pain would be similarly associated with depression in the emerging adult population, the definitiveness of this conclusion is limited by the small number of research studies in this area.

Heightened somatic awareness, also known as somatization, is associated with depression<sup>13</sup>, which makes it logical to explore these psychological factors concurrently. Somatization is defined as a "complex array of behaviors characterized by an abundant usage of body expressions and language to convey feelings of personal complaint and social distress, amplifying or distorting sometimes subtle physiological changes"<sup>14</sup>, and somatization can be clinically diagnosed as a somatoform disorder when physical symptoms suggestive of illness or injury are unexplainable by a known medical condition<sup>15</sup>. Clinical co-occurrence of depressive and somatoform disorders is quite common.<sup>16</sup> As with depression, somatization symptoms have been positively associated with low back pain in a general adult population<sup>17</sup>, although a recent study found that patients with chronic low back pain were no more likely to be diagnosed with somatoform disorders than patients without back pain<sup>18</sup>. Both depression and somatization have been found to co-occur in adult patients with non-specific chronic low back pain.<sup>19</sup> Somatoform disorders are known to occur in adolescents and young adults<sup>20,21</sup>, although the association between somatization and low back pain, and the co-occurrence of depressive and somatization symptoms with low back pain, have been minimally investigated in an emerging adult population in general and in Canada specifically.

Given the lack of literature regarding the association between low back pain and depressive and somatization symptoms in an emerging Canadian adult population, the purpose of the present study was to investigate these associations in a sample of first year students at a Canadian university. It was hypothesized that as in the general adult population, depressive and somatization symptoms would be positively associated with low back pain in an emerging adult population. The results of this study will provide novel insight into the potential co-occurrence of these conditions within an emerging adult population and inform future research in the therapeutic management and prevention of low back pain.

#### Methods

#### Participants

Subjects were undergraduate students enrolled in a firstyear biology course at the University of Guelph in Fall 2011, Winter 2012, Fall 2012, and Winter 2013. As part of the course, students completed health questionnaires and surveys that were incorporated in the laboratory portion of the course curriculum. Students received 0.5% for completion of each survey. The questionnaires and surveys were linked to the topics that were covered in the course, including back pain and mental health. The purpose of completing the questionnaires and surveys was to provide students with feedback regarding their personal experiences with these health issues. Students were encouraged to seek medical help for health problems identified through the course of survey and questionnaire completion, and were directed to Student Health Services for assistance. During the final week of the semester, the researchers presented the purpose of the study to the students and requested their informed consent to analyze the data previously collected. A total of 1013 students (781 female and 205 male students) provided consent to have their survey data analyzed, which represents 31% of all students enrolled in the course. This study was approved by the Research Ethics Board at the University of Guelph. All students in each course were invited to participate in the study, and consenting subjects gave written consent to have collected data analyzed. Because students completed the surveys as part of the course, no inclusion or exclusion criteria were applied to aggregate course data collection. The exclusion criteria applied for participation in the research study was that students must have completed the three surveys in their entirety, although personal data regarding subject gender and age could be missing. Due to the nature of data collection in this study, information on confounding variables was not collected.

#### Surveys

The questionnaires completed by students included the Modified Zung Depression Index, the Modified Somatic Perception Questionnaire, and questions about low back pain frequency and intensity.

The Modified Zung Depression Index is a validated, self-administered rating scale that has been used to identify depression.<sup>22</sup> The modified version of Zung's original scale was developed by Main et al.23 as part of the Distress and Risk Assessment Method that is used as a psychological assessment screening tool for those with chronic pain and back pain. The Distress and Risk Assessment Method also assists in the classification of patients into those showing no psychological distress, those at risk and those distressed.<sup>23</sup> It helps guide the clinician to whether a more comprehensive psychological or psycho-physical assessment is warranted. The Modified Zung Depression Index is scored using an adjectival scale with 4 response categories, ranging from "not at all all/little of the time" (score 0) to "most of the time" (score 3). The total score (sum score for all 23 items) ranges from 0 to 69 points. Those scores less than 16 are considered "normal", those between 17 and 33 are considered "at-risk" and those above 34 are considered "depressive". Several studies have investigated the validity and reliability of the Zung index with good outcomes, as reviewed in Mariush  $(2009).^{24}$ 

The Modified Somatic Perception Questionnaire is a

13 item self-report scale for patients with chronic pain or disabilities.<sup>2</sup>. It can help identify somatic complaints that may be associated with psychological responses such as anxiety or depression.<sup>26</sup> The scale was developed by Main as a clinical screening instrument to measure somatic and autonomic perception in patients with low back pain and other chronic pain problems.<sup>25</sup> The questionnaire was designed to identify psychological distress in patients with low back pain and provide information to facilitate the distinction between physical pathology and illness behaviour.<sup>25</sup> The questionnaire includes questions about the occurrence in the last week of various symptoms such as nausea, sweating or feeling faint. The Modified Somatic Perception Questionnaire is scored using an adjectival scale with 4 response categories ranging from "not at all" (0 points) to "could not have been worse" (3 points) for each item. The 13 items that are scored are denoted with an asterisk in Table 3. The total score (sum score of all 13 items) ranges from 0 to 39. Those total scores below 12 are considered "at-risk" and those scores above 12 are considered "distressed". It is important to note that the "at-risk" category includes very low somatization scores, and therefore, for the purposes of the present analysis, this category will be considered as "non-distressed". The Modified Somatic Perception Questionnaire has an internal consistency of alpha = 0.78, and it is significantly correlated with the Zung Depression Index.<sup>26</sup>

Students were also asked to identify the frequency of past or present low back pain, using a four point scale of never, once, intermittent, or chronic. They were also asked to identify the intensity of past or present low back pain, using a four-point scale of none, mild, moderate or severe. Frequency and intensity of low back pain were combined and divided into four graded categories to make a single dependent variable for data analysis. Four levels

Table 1.Grade of low back pain.

Grade	<b>Description</b> (Frequency + Intensity)
0	None or a single episode with mild intensity
1	Intermittent or chronic episodes with mild intensity
2	Single episode of moderate to severe intensity
3	Intermittent or chronic episodes with moderate to severe intensity

of grade of low back pain were determined as illustrated in Table 1. No clinimetric values are available for the low back pain survey.

#### Statistical Analysis

All statistical analyses in this study were conducted in consultation with a statistician at the University of Guelph. All tests were done with SPSS version 20. Using logistic regression, a full model was constructed which included somatization and depression as main effects along with somatization by depression as an interaction term. The interaction term was found to be non-significant (p=0.575) and was not included in the model. For both models, the Likelihood Ratio Chi-Square test and Pseudo-R squared test (p<0.05) indicates the full model statistically predicts the dependent variable better than the intercept-only model alone. For the investigation of low back pain and somatization, the "non-distressed" category was used as the reference, while for low back pain and depression, the "normal" category was the reference group. Using Pearson Chi-Square test, the association between somatization and depression was found to be significant (p<0.05). Therefore, to investigate the specific associations between the co-occurrence of depression and somatization with LBP, the categories of depression and somatization were combined as follows: Non-distressed/At-risk Depression, Non-distressed/Depressed, Distressed/Normal Depression, Distressed/At-Risk Depressed, and Distressed/Depressed. The combination of the Non-Distressed and Normal Depression categories was used as the reference. This statistical approach is consistent with methods used to analyze categorical data<sup>27</sup>, and determines the odds ratio (OR), which represents an index of effect, or effect size. Confidence intervals (CI) were also determined. Significance was accepted at p < p0.05. Descriptive statistics models were also used to determine the prevalence of low back pain within the study.

#### Results

#### Subject Characteristics and Prevalence of Low Back Pain

Subject characteristics including gender, age, prevalence of LBP by grade, and scores on the Zung Depression Index and Modified Somatic Perception Questionnaire are reported in Table 2. The grade of low back pain as a

Table 2.	
Subject Characteristics and Prevalence of	f
Low Back Pain	

Descriptive Statistics	n	%
Sex		
Male	205	20.8
Female	781	79.2
TOTAL	986	
Age (years)		
<17	4	0.4
17	56	5.7
18	653	66.3
19	197	20.0
20	30	3.0
21	15	1.5
22	12	1.2
>22	18	1.8
TOTAL	985	
Modified Zung Depression		
Index		
Normal	434	42.8
At-Risk	484	47.8
Depressive	95	9.4
TOTAL	1,013	
Modified Somatic Perception		
Questionnaire		
At-Risk	899	88.7
Distressed	114	11.3
TOTAL	1,013	
Low Back Pain Grade		
0	496	49.0
1	252	24.9
2	55	5.4
3	210	20.7
TOTAL	1,013	

function of gender is illustrated in Table 3. It should be noted that 27 subjects (2.6%) elected to not disclose their gender and were excluded from the analysis of low back pain and gender. Similarly, 28 subjects (2.7%) elected to not to disclose their age. Notably, a total of 49% of the population were classified as grade 0, 24.9% of the population were classified as Grade 1, 5.4% of the population were classified as Grade 2, and 20.7% of the population were classified as Grade 3. There were more females than males in the subject sample, resulting in more females in each category of low back pain grade. The percentage of each grade within genders was quite comparable,

100

Table 3.Grade of Low Back Pain by Gender

Grade		Male	Female	Total
0	Count	118	368	468
	% within grade	24.3%	75.7%	100%
	% within gender	57.6%	47.1%	49.3%
1	Count	46	200	246
	% within grade	18.7%	81.3%	100%
	% within gender	22.4%	25.6%	24.9%
2	Count	10	41	51
	% within grade	19.6%	80.4%	100%
	% within gender	4.9%	5.2%	5.2%
3	Count	31	172	203
	% within grade	15.3%	84.7%	100%
	% within gender	15.1%	22.0%	20.6%
Total	Count	205	781	986
	% of Total	20.8%	79.2%	100%

Table 4.Low Back Pain and Somatization

Grade	Somatic Category	Sig	Odds Ratio	95% CI
1	Distressed	0.001*	2.594	1.505 to 4.471
2	Distressed	0.305	1.672	0.626 to 4.465
3	Distressed	0.000*	3.219	1.873 to 5.533

Legend: \*p<0.05

although there were 10.5% more males in the grade 0 category, and 6.9% more females in the grade 3 category. Ninety five percent of subjects were between the ages of 17 and 20.

#### Low Back Pain and Somatization

Table 4 presents the unadjusted OR and 95% CI for the association between low back pain and somatization symptoms. The mean  $\pm$  SD score for the Modified Somatic Perception Questionnaire was  $6.52 \pm 4.168$ . The analyses demonstrate that those in the emerging adult population falling within the distressed category of somatic percep-

Depression Category	Sig	Odds Ratio	95% CI
Depressive	0.810	1.077	0.589 to 1.968
Depressive	0.468	1.510	0.496 to 4.598
Depressive	0.116	1.642	0.884 to 3.049
At-Risk	0.132	1.282	0.928 to 1.771
At-Risk	0.026*	2.002	1.088 to 3.684
At-Risk	0.001*	1.874	1.310 to 2.681
	Depression Depressive Depressive Depressive At-Risk At-Risk At-Risk	Depression CategorySigDepressive0.810Depressive0.468Depressive0.116At-Risk0.132At-Risk0.026*At-Risk0.001*	Depression CategorySigOdds RatioDepressive0.8101.077Depressive0.4681.510Depressive0.1161.642At-Risk0.1321.282At-Risk0.026*2.002At-Risk0.001*1.874

Table 5.Low Back Pain and Depression

Legend: \*p<0.05

tion are more likely than people in the non-distressed category to have low back pain. Individuals in the distressed somatic perception category were 2.594 times more likely to report grade 1 low back pain and 3.219 times more likely to report grade 3 low back pain than those in the non-distressed category. A non-significant direct association was seen between grade 2 low back pain and somatization.

#### Low Back Pain and Depression

Table 5 presents the unadjusted OR and 95% CI for the association between low back pain and depressive symptoms. The mean  $\pm$  SD score for the Modified Zung Depression Index was  $19.14 \pm 9.796$ . The analyses demonstrate that people in the at-risk category of depression were more likely than people in the normal depression category to be classified as having low back pain. Significant associations were observed for the at-risk depression category and grades 2 and 3 low back pain, whereby individuals in the at-risk depression category were 2.002 times more likely classified with grade 2 and 1.874 time more likely to be classified with grade 3 low back pain than those in the normal depression category. Non-significant direct associations were seen between the depressive category of depression and grade 2 and 3 low back pain, and the at-risk depression category and grade 1 low back pain.

## Low back pain, Depression and Somatization Co-occurrence

Table 6 presents the unadjusted OR and CI for the association between low back pain and the co-occurrence of depressive and somatization symptoms. The analy-

LBP Grade	Combined Category	Sig	<b>Odds Ratio</b>	95% CI
1	Distressed/Depressed	0.143	1.837	0.815 to 4.141
1	Distressed/At-Risk Depression	0.000*	3.966	1.855 to 8.482
1	Distressed/Normal Depression	0.012*	5.845	1.481 to 23.069
1	Non-distressed/Depressed	0.241	1.525	0.754 to 3.086
1	Non-distressed/At-risk Depression	0.121	1.303	0.933 to 1.819
2	Distressed/Depressed	0.416	1.906	0.402 to 9.026
2	Distressed/At-Risk Depression	0.066	3.574	0.920 to 13.886
2	Distressed/Normal Depression	0.186	4.765	0.470 to 48.289
2	Non-distressed/Depressed	0.348	1.864	0.508 to 6.839
2	Non-distressed/At-risk Depression	0.023*	2.073	1.107 to 3.881
3	Distressed/Depressed	0.001*	3.857	1.790 to 8.309
3	Distressed/At-Risk Depression	0.000*	7.393	3.489 to 15.666
3	Distressed/Normal Depression	0.103	3.857	0.760 to 19.571
3	Non-distressed/Depressed	0.068	2.012	0.950 to 4.264
3	Non-distressed/At-risk Depression	0.002*	1.813	1.252 to 2.624

Table 6.Low Back Pain Grade and Co-Occurrence of Symptoms

Legend: \*p<0.05

ses demonstrate that there was a significant association between the co-occurrence of the distressed category of somatization and the category of at-risk depression with grade 1 and 3 LBP. This association was strongest for subjects in the grade 3 category of low back pain. Subjects in the grade 3 category of low back pain also showed a strong association with the co-occurrence of the distressed category of somatization and the depressed category of depression.

#### Discussion

The objective of this study was to investigate the associations between low back pain, depression and somatization, both independently and co-occurring, in an emerging adult population. Our findings demonstrate that both depression and somatization are positively associated with low back pain, both independently and co-occurring. These results are consistent with the literature regarding low back pain, somatization, and depression in a general adult population and suggest that these conditions are relatively common at the onset of adulthood.

The results of this study support the evidence that low back pain is quite prevalent in an emerging adult population, with over half of the participants reporting current or past low back pain across grades. Subjects in this study most commonly reported experiencing back pain in the lowest grade category, with nearly 25% reporting intermittent or chronic episodes of mild intensity. Another nearly 21% reported intermittent or chronic episodes with moderate to severe intensity while only 5% experienced a single episode of moderate to severe intensity. These results are comparable to those previously reported by Cakmak et al.28 who examined the prevalence and risk factors associated with low back pain in an emerging adult population in Turkey. The authors surveyed 1,552 university-aged students between 17 and 26 years of age and reported a low back pain prevalence of 40.9% within this population and the prevalence increased with age.<sup>28</sup> A similar study by Falavigna et al.29 evaluated the association between physiotherapy and medical students with a mean age of approximately 22 years, and reported an even higher lifetime prevalence of low back pain at nearly 80%. While differences in study outcomes exist within the in the emerging adult population, there are a variety of factors to which these differences may be attributed including questionnaires, subject characteristics, study

design and cultural differences. Despite these differences, existing studies demonstrate that low back pain is a common occurrence during early adulthood years.

One relevant consideration regarding low back pain prevalence in the present study is the younger age of this population (17-22 years, with 95% between ages 17 and 20) which falls on the lower end of the emerging adult range and spans the later years of adolescence. A recent meta-analysis of low back pain in children and adolescents found that the lifetime prevalence of low back pain was approximately 40%, further showing a positive association with age.30 In addition to previously described Cakmak et al.28, an association of age with low back pain was similarly observed in a study by Taspinar et al.<sup>31</sup>, who found that University students under 19 years of age experienced less severe back pain than those over 19 years. Similarly, the prevalence of back pain in a general adult population is higher than in children and/or adolescents, with prevalence rates approaching 80%.<sup>1,32,33</sup> To this extent, the results of the present study are consistent with prior literature demonstrating that low back pain prevalence increases with age, placing the emerging adult population on a continuum including childhood, adolescence, and later adulthood.

Low back pain has also consistently been shown to be associated with depression in adults across observational studies, with research suggesting that people experiencing depression are approximately 60% more likely to develop back pain in their lifetime versus non-depressed people.<sup>8</sup> This has important implications on the potential impact of low back pain to society, since the presence of depression has been shown to have a negative effect on the course of recovery of low back.<sup>34</sup> Persistent low back pain also increases the risk of developing depressive symptoms.<sup>35</sup> In the present study, the odds of participants in the at-risk categories of depression reporting low back pain were increased, with significant associations observed at the higher grades of low back pain. This observation suggests that, in a young emerging adult population, depression is a risk factor for low back pain and is consistent with the findings across later adulthood. These findings align with previous research by Kennedy et al.<sup>10</sup> and Unalan et al.<sup>11</sup>, who reported similar associations between low back pain and depressive symptoms. Only one study did not observe a difference in depression scores between low back pain and control students<sup>12</sup>, however, this study was limited

by small sample size of subjects reporting depression. Despite this, the collective literature in this area strongly supports an association between low back pain and depression in both emerging and general adult populations.

As with depression, somatization has also been associated with low back pain in adults<sup>17</sup>, with strong associations also observed with low back pain severity and disability<sup>19</sup>. In contrast to depression, the association between low back pain and somatization in both the emerging and general adult populations is poorly understood owing to the limited research attention it has received. Only one recent study in adults failed to show any relationship between somatization and low back pain outcomes.<sup>18</sup>

The results of the present study provide evidence for an association between somatization and low back pain in a young emerging adult population. Participants in the distressed category of somatization were more likely to experience low back pain than those in the non-distressed category and this association was particularly robust for the highest grade of low back pain defined as intermittent or chronic episodes with moderate to severe intensity. The similarities in findings between low back pain, somatization and depression are unsurprising given the established relationship between these psychological factors in the literature.<sup>13</sup> Furthermore, the observed relationship between low back pain and the co-occurrence of low back pain with depressive and somatization symptoms are consistent with the literature that somatic perception and depression are risk factors for low back pain in general adulthood<sup>36</sup>, and the co-occurrence of depression and somatization in adults with low back pain<sup>19</sup>.

Consideration of psychological factors in relation to low back pain is highly relevant. It has been suggested that negative psychological attributes such as pain catastrophizing and pain-related fear avoidance behaviour, heightened somatic awareness and depression are all associated with greater perceptions of pain and disability.<sup>37</sup> Gatchel *et al.*<sup>38</sup> proposed that there are clear differences in the role of emotional distress in high-risk acute back pain vs. high-risk chronic back pain, as patients with chronic low back pain have shown to have worsened psychosocial sequelae relative to acute low back pain patients. This suggests that these maladaptive psychosocial symptoms may be related to the development of the chronicity of pain.<sup>38</sup> In a 2002 systematic review of psychological factors associated in the development of back pain chronicity, Pincus et al.39 found evidence for the role of somatization, distress and depression in the transition from acute to chronic low back pain. Although the present study suggests that depression and somatization is a risk factor for low back pain in emerging adults, it does not resolve the mechanism(s) that may be responsible for the association between these variables. Crombie et al.40 have suggested that pain is a manifestation of the interaction of cognitive, emotional, motivational, behavioural and physical components, while Carroll et al.41 suggest that the influence of depression and somatic perception can influence low back pain in a number of ways such as passive coping. Further investigation is needed to elucidate the pathways and chronology between low back pain and somatic perception.

There are several limitations that should be considered in the interpretation of the results of this study, the most significant of which being our inability to control for confounding variables that are known to influence back pain. For example, several factors have been shown to influence back pain in University students, including smoking, class attendance, using a computer and using lumbar support.<sup>31</sup> Other factors such as age, sex, inactivity, obesity and race have also been identified as risk factors for the development of back pain in children, adolescents and adults.<sup>42-44</sup> Although these variables were not collected as part of the present study, the strength of the observed associations and the consistency of our findings with previous literature supports the validity of our observations. As well, this study is limited to University students enrolled in one class at a single Canadian institution, which may not be representative of the diversity of the emerging adult population. It should also be noted that a minority of students enrolled in the classes under investigation consented to participate in the research study, and that not all subjects provided data for all outcomes, such as gender and age. However, by recruiting subjects across four classes, our total sample size was quite large, and the number of subjects without all data points represents only a small fraction of the total subject sample. It should also be acknowledged that the early mean age of our sample does not capture the demographics of the broad emerging adulthood population. In spite of this, low back pain has been shown to be a significant issue throughout the world<sup>45</sup>, and the collective geographically

specific studies has not shown large differences between populations. Low back pain has been studied specifically in Canadian adults<sup>1</sup> and adolescents<sup>46</sup>, and low back pain has been shown to be the strongest predictor of major depression in a Canadian adult population<sup>47</sup>. As well, subjects' assessment of low back pain may also be subject to recall bias and subject bias, and there is a slight risk of bias due to the provision of grades for completion of the health surveys. A final limitation of the present study is that the clinical significance of the odds ratios calculated in this study are unclear, although it should be noted that Deyo *et al.*<sup>26</sup> found that the Modified Somatic Perception Questionnaire was only weakly associated with pain outcomes.

In conclusion, the results of this study demonstrate a high prevalence of low back pain in an emerging adult population and that low back pain is directly associated with depression and somatization. These findings are consistent with previous research in adult, emerging adult and adolescent populations. Future studies should examine whether there is an increasing incidence of low back pain and depression and somatization across the full age range spanning the emerging adult population, and aim to elucidate potential mechanisms underlying the association of these variables. The significant burden of musculoskeletal pain and psychological suffering highlights an urgent need for development of preventive interventions targeting this population. We suggest that interventions could be directly integrated into academic curriculums at Universities and colleges given their broad inclusion of the emerging adult population, and should be considered an important target of public health and prevention initiatives.

#### References

- 1. Cassidy JD, Carroll LJ, Côté P. The Saskatchewan health and back pain survey: the prevalence of low back pain and related disability in Saskatchewan adults. Spine. 1998; 23: 1860-1866.
- Vos T, Flaxman AD, Naghavi M, Lozano R, Michaud C, Ezzati M, Memish ZA. Years lived with disability (YLD's) for 1160 sequelae of 289 diseases and injuries 1990–2010: a systematic analysis for the Global Burden of Disease Study 2010. Lancet. 2012; 380: 2163–2196.
- Nolet PS, Côté P, Kristman VL, Carroll LJ, Cassidy JD. Is prevalent low back pain associated with worse health related quality of life six months later? A population based cohort study. Eur Spine J. 2015; 24: 458-466.
- 4. Arnett JJ. Emerging adulthood: A theory of development

from the late teens through the twenties. Am Psychol. 2000; 55: 469.

- Kwan, MY, Faulkner GE, Arbour-Nicitopoulos KP, Cairney J. Prevalence of health-risk behaviours among Canadian post-secondary students: descriptive results from the National College Health Assessment. BMC Public Health. 2013; 13: 548.
- Ferro MA, Gorter JW, Boyle MH. Trajectories of depressive symptoms in Canadian emerging adults. Am J Public Health. 2015; 105: 2322-2327.
- Lisznyai S, Vida K, Nemeth M, Benczur Z. Risk factors for depression in the emerging adulthood. J Couns Psychol. 2014; 3.
- Pinheiro MB, Ferreira ML, Refshauge K, Maher CG, Ordonana JR, Andrade TB, Ferreira PH. Symptoms of depression as a prognostic factor for low back pain: a systematic review. Spine J. 2016; 16: 105-116.
- 9. Pinheiro MB, Ferreira ML, Refshauge K, Ordonana JR, Machado GC, Prado LR, Maher CG, Ferreira PH. Symptoms of depression and risk of new episodes of low back pain: a systematic review and meta-analysis. Arthritis Care Res. 2015; 67: 1591-1603.
- Kennedy C, Kassab O, Gilkey D, Linnel S, Morris D. Psychosocial factors and low back pain among college students. J Am Coll Health. 2008; 57: 191-196.
- Unalan D, Celikten M, Mazicioglu M, Soyuer F. Depressive symptom profile of Turkish students experiencing back pain. J Soc Behav Pers. 2009; 37: 155-162.
- Mitchell T, O'Sullivan PB, Smith A, Burnett AF, Straker L, Thornton J, Rudd CJ. Biopsychological factors are associated with low back pain in female nursing students: a cross-sectional study. Int J Nurs Stud. 2009; 46: 678-688.
- Lieb R, Meinlschmidt G, Araya R. Epidemiology of the association between somatoform disorders and anxiety and depressive disorders: an update. Psychosom Med. 2007; 69: 860-863.
- 14. Bragazzi NL, Del Puente G, Natta WM. Somatic perception, cultural differences and immigration: results from administration of the Modified Somatic Perception Questionnaire (MSPQ) to a sample of immigrants. Psychol Res Behav Manag. 2014; 7: 161.
- Diagnostic and statistical manual of mental disorders.
  5th. ed. Washington: American Psychiatric Association Publishing. 2013.
- Bener A, Al-Kazaz M, Ftouni D, Al-Harthy M, Dafeeah EE. Diagnostic overlap of depressive, anxiety, stress and somatoform disorders in primary care. Asia Pac Psychiatry. 2013; 5: E29-38.
- 17. Bener A, Verjee M, Dafeeah EE, Falah O, Al-Juhaishi T, Schlogl J, Sedeeq A, Khan S. Psychological factors: anxiety, depression, and somatization symptoms in low back pain patients. J Pain Res. 2013; 6: 95-101.
- 18. Ramon-Roquin A, Pecquenard F, Schers H, Van Weel C,

Oskam S, Van Boven K. Psychological, musculoskeletal and somatoform comorbidity in patients with chronic low back pain: original results from the Dutch Transition Project. Fam Pract. 2015; 32: 297-304.

- 19. Licciardone JC, Gatchel RJ, Kearns CM, Minotti DE. Depression, somatization, and somatic dysfunction in patients with nonspecific chronic low back pain: results from the OSTEOPATHIC trial. J Am Osteopath Assoc. 2012; 112: 783-791.
- 20. Lieb R, Pfister H, Mastaler M, Wittchen HU. Somatoform syndromes and disorders in a representative population sample of adolescents and young adults: prevalence, comorbidity and impairments. Acta Psychiatrica Scandinavica. 2000; 101: 194-209.
- 21. Lieb R, Zimmerman P, Friis RH, Hofler M, Tholen S, Wittchen HU. The natural course of DSM-IV somatoform disorders and syndromes among adolescents and young adults: a prospective-longitudinal community study. Eur Psychiatry. 2002; 17: 321-331.
- 22. Zung, WW. A self-rating depression scale. Arch Gen Psychiatry. 1965; 12: 63-70.
- 23. Main CJ, Wood PL, Hollis S, Spanswick CC, Waddell G. The distress and risk assessment method: a simple patient classification to identify distress and evaluate the risk of poor outcome. Spine. 1992; 17: 42-52.
- 24. Mariush ME. Psychological Testing in the Age of Managed Behavioral Health Care. Lawrence Ehrlbaum Associates Inc, NH. 2001: 132-133.
- 25. Main CJ. The modified somatic perception questionnaire (MSPQ). J Psychosom Res. 1983; 27: 503-14.
- 26. Deyo RA, Walsh NE, Schoenfeld LS, Ramamurthy S. Studies of the Modified Somatic Perceptions Questionnaire (MSPQ) in patients with back pain: psychometric and predictive properties. Spine. 1989; 14: 507-510.
- 27. Agresti A. An Introduction to Categorical Data Analysis. New York: Wiley. 1996.
- 28. Çakmak A, Yücel B, Özyalçn SN, Bayraktar B, Ural HI, Duruöz MT, Genç A. The frequency and associated factors of low back pain among a younger population in Turkey. Spine. 2004; 29: 1567-1572.
- Falavigna A, Teles AR, Mazzocchin T, de Braga GL, Kleber FD, Barreto F, Beckenkamp NL. Increased prevalence of low back pain among physiotherapy students compared to medical students. Eur Spine J. 2011; 20: 500-505.
- Calvo-Munoz I, Gomez-Conesa A, Sanchez-Meca. Prevalence of low back pain in children and adolescents: a meta-analysis. BMC Pediatr. 2013; 13: 14.
- 31. Taspinar F, Taspinar B, Cavlak U, Celik E. Determining the pain-affecting factors of University students with nonspecific low back pain. J Phys Ther Sci. 2013; 25: 1561-1564.
- 32. Schmidt CO, Raspe H, Pfingsten M, Hasenbring M, Basler HD, Eich W, Kohlmann T. Back pain in the

German adult population: prevalence, severity, and sociodemographic correlates in a multiregional survey. Spine (Phila Pa 1976). 2007; 32: 2005-2011.

- Walker BF, Muller R, Grant WD. Low back pain in Australian adults. Prevalence and associated disability. J Manipulative Physiol Ther. 2004; 27: 238-244.
- 34. Melloh M, Elfering A, Käser A, Salathé CR, Barz T, Aghayev E, Theis JC. Depression impacts the course of recovery in patients with acute low-back pain. J Behav Med. 2013; 39.
- 35. Elfering A, Käser A, Melloh M. Relationship between depressive symptoms and acute low back pain at first medical consultation, three and six weeks of primary care. Psychol Health Med. 2014; 19: 235-246.
- Edmond SL, Werneke MW, Hart DL. Association between centralization, depression, somatization, and disability among patients with nonspecific low back pain. J Orthop Sports Phys Ther. 2010; 40: 801-810.
- Meyer K, Tschopp A, Sprott H, Mannion AF. Association between catastrophizing and self-rated pain and disability in patients with chronic low back pain. J Rehabil Med. 2009; 41: 620-625.
- 38. Gatchel RJ, Bernstein D, Stowell AW, Pransky G. Psychosocial differences between high-risk acute vs. chronic low back pain patients. Pain Pract. 2008; 8: 91-97.
- Pincus T, Burton AK, Vogel S, Field AP. A systematic review of psychological factors as predictors of chronicity/ disability in prospective cohorts of low back pain. Spine. 2002; 27: E109-120.
- 40. Crombie IK, Croft PR, Linton SJ, LeResche L, Von Kroff M. Seattle: IAS Press, 1999: 25-42.
- Carroll LJ, Cassidy JD, Côté P. Depression as a risk factor for onset of an episode of troublesome neck and low back pain. Pain. 2014; 107:134-139.
- Balague F, Troussier B, Salminen JJ. Non-specific low back pain in children and adolescents: risk factors. Eur Spine J. 1999; 8: 429-438.
- Manchikanti L, Singh V, Falco FJ, Benyamin RM, Hirsch JA. Epidemiology of low back pain in adults. Neuromodulation. 2014; 17: 3-10.
- 44. Waterman BR, Belmont PJ, Schoenfeld AJ. Low back pain in the United States: incidence and risk factors for presentation in the emergency setting. Spine J. 2012; 12: 63-70.
- 45. Hoy D, Bain C, Williams G, March L, Brooks P, Blyth F, Woolf A, Vos T, Buchbinder R. A systematic review of the global prevalence of low back pain. Arthritis Rheum. 2012; 64: 2028-2037.
- Feldman DE, Shrier I, Rossignol M, Abenhaim L. Risk factors for the development of low back pain in adolescence. Am J Epidemiol. 2000; 154: 30-36.
- Currie SR, Wang J. Chronic back pain and major depression in the general Canadian population. Pain. 2004; 107: 54-60.

# Systematic review and meta-analyses of the difference between the spinal level of the palpated and imaged iliac crests

Robert Cooperstein, MA, DC<sup>1</sup> Felisha Truong<sup>1</sup>

Objective: The purpose of this study was to undertake a systematic review of the literature to determine and compare, for patient sub-groups, the spinal level of the iliac crests as commonly measured through manual palpation and radiographic imaging procedures.

Methods: Relevant citations were retrieved by searching the PubMed, ICL, CINAHL, AMED, Osteopathic Research Web, OstMed, and MANTIS biomedical databases, and included articles were rated for quality. Search terms included Tuffier\*, intercristal line, intercrestal line, Jacoby's line, lumbar spine, lumbar landmark, pelvic landmark, palpation, and TL (Tuffier's Line). Meta-analyses were performed on the full datasets as well as subsets based on various patient demographics.

Results: Original search strategies retrieved 1301 citations; 47 articles were used for qualitative synthesis and 31 for meta-analyses. Across these studies imaged Objectif : Cette étude visait à entreprendre un examen systématique de la littérature dans le but de déterminer et de comparer, pour les sous-groupes de patients, le niveau rachidien des crêtes iliaques comme on le mesure souvent par palpation manuelle et imagerie radiographique.

Méthodologie : On a tiré des citations pertinentes par le biais d'une recherche dans les bases de données médicales PubMed, ICL, CINAHL, AMED, Osteopathic Research Web, OstMed, et MANTIS, et les articles compris étaient cotés aux fins de qualité. Les termes de recherche comprenaient Tuffier\*, ligne intercrête, ligne de Jacoby, colonne lombaire, repère lombaire, repère pelvien, palpation et LT (ligne de Tuffier). On a effectué des méta-analyses des ensembles de données complètes, ainsi que des sous-ensembles, fondées sur les diverses données démographiques sur les patients.

Résultats : Les premières stratégies de recherche ont permis de tirer 1 301 citations; on a utilisé 47 articles aux fins de synthèse qualitative et 31 pour des métaanalyses. À l'échelle de ces études, les crêtes imagées

Corresponding author: Robert Cooperstein Palmer Chiropractic College, San Jose CA 94577 Tel: 510-207-6009 Fax: 408-944-6009 E-mail: Cooperstein\_r@palmer.edu

Disclaimer: Neither of the investigators have any commercial or other conflict of interest associated with this project. © JCCA 2017

<sup>&</sup>lt;sup>1</sup> Palmer College of Chiropractic, San Jose CA

crests were found to be most consistent with and closest to the L4-5 interspace in females and L4 spinous process in males. In comparison, the spinal level for the palpated crests was nearest to the L3-4 interspace in males and females. The palpated crest line was 0.7 levels cephalad to the imaged crest line in males, and 1.0 levels cephalad to the imaged line in females.

Discussion and Conclusions: During manual palpation, the examiner's fingers contact soft tissue overlying the iliac crests, thereby usually identifying the L3-4 spinal level rather than the assumed L4-5 level. Palpating iliac crests to guide anesthetic injections or manual therapy without appreciating these findings can be hazardous or lead to suboptimal patient care.

(JCCA. 2017;61(2):106-120)

KEY WORDS: chiropractic, palpation, ilium, radiography, anatomic landmarks, lumbar vertebrae

#### Introduction

Practitioners in the health care professions use anatomical landmarks to identify spinal levels, both to enhance diagnostic accuracy and specifically target the site of interventions. Anesthetists require precise placement of epidural catheters to optimize postoperative analgesia and minimize adverse effects. Anatomic landmarks have been used to locate acupuncture points<sup>1</sup>, and surgeons may in part base the location to begin incision on the location of anatomical landmarks<sup>2, p.18</sup>. Manual therapists palpate spinal and pelvic structures to determine boney symmetry and movement capacities. These procedures may involve using anatomical landmarks to numerate spinal levels. To identify lumbar levels, clinicians generally use the iliac crests as a landmark, which are generally thought based on imaging studies, to lie at L4 or L4-5. The line drawn across the superior aspect of the crests is usually called Tuffier's line (TL)<sup>3</sup>, but has also been called the intercrestal line<sup>4-6</sup>, intercristal line<sup>7-10</sup> or Jacoby's line<sup>11</sup>. Other lumbar and thoracic levels are generally identified by counting up or down from the putative location of L4

étaient les plus conformes et proches de l'espace intercostal L4-5 des femmes et de l'apophyse épineuse L4 des hommes. Comparativement, le niveau rachidien des crêtes palpées était plus près de l'espace intercostal L3-4 des hommes et des femmes. La ligne de crête palpée était de niveau 0,7 vers la tête par rapport à la ligne de crête imagée des hommes et de niveau 1,0 vers la tête par rapport à la ligne imagée des femmes.

Discussion et conclusions : Lors de la palpation manuelle, les doigts de l'examinateur touchent aux tissus mous qui recouvrent les crêtes iliaques, ce qui, en général, détermine de ce fait le niveau rachidien L3-4 plutôt que le niveau L4-5 présumé. La palpation des crêtes iliaques visant à guider les injections anesthésiques ou la thérapie manuelle sans souscrire à ces constatations peut s'avérer dangereuse ou mener à des soins sous-optimaux aux patients.

(JCCA. 2017;61(2):106-120)

MOTS CLÉS : chiropratique, palpation, ilion, radiographie, repères anatomiques, vertèbre lombaire

or L4-5. Although Render<sup>12</sup> and others convincingly demonstrated the accuracy of this landmark rule based on the imaged TL, the accuracy and reliability of identifying the L4 level by manually palpating the crests remains in question.

For instance, several studies have evaluated the accuracy of static palpation, which generally uses the palpated iliac crests to identify the L4-5 interspace; these studies have consistently reported errors such that the identified spinal level is almost always cephalad to the intended spinal level<sup>7, 13-18</sup>. In such studies a radio-opaque marker is applied to the skin at the presumed vertebral level, the location of which is then compared to the actual level as established by an imaging procedure<sup>19</sup>. Chakraverty's<sup>7</sup> research convincingly illustrated the basis for this systematic bias: that is, that the spinal level of the palpated crests is more cephalad than the spinal level of TL as seen on imaging studies. Chakraverty et al.7 found that although imaging associated TL with either the L4 or L4-5 spinal levels in 86.7% (mostly female) patients, the spinal level identified using the palpatory crest method was either the



Figure 1. PRISMA Flow Diagram



Figure 2. White markers indicate the spinal level of the palpated

L3 or L3–4 spinal levels in 77.3% of cases, most commonly in females and in patients with higher body mass indices (BMIs). Findings by Kim<sup>20</sup> also underscore the difference between spinal levels identified through imaging and palpatory methods (Figure 2). Several authors<sup>7,21-25</sup> have pointed out that when an examiner's fingers are placed on the "iliac crests", in reality the palpator is compressing soft tissue (in some cases quite a lot of soft tissue) between the fingertips and the crest; this may help to explain the bias toward cephalad measurement errors.

Since palpation and imaging studies have typically reported their data in terms of the frequencies with which the iliac crests intersect the L4-5 or other spinal level, rather than in terms of the mean spinal levels and their 95% confidence intervals, it has been difficult to ascertain the precise association and variability of spinal levels as measured with palpatory or imaged methods. The purpose of this study was to undertake a systematic review of the literature and perform meta-analyses on data for patient sub-groups for the spinal level associated with TL as established by both imaging and palpatory methods, and to report the data in a clinically meaningful way as mean locations and 95% confidence intervals.

Ultimately, this study intended to determine if a common strategy could be suggested for clinicians, especially manual therapists and anesthetists, to improve their ability to accurately locate lumbar and perhaps thoracic vertebral levels, by using TL or possibly an alternative anatomical structure as a landmark.

#### Methods

The primary inclusion criterion for an article to be included in this review was that it concerned the location of the spinal level of TL as established through either comparison with an imaging reference standard or through manual palpation. An included article, either an imaging or palpation study, did not have to explicitly aim at establishing the spinal location of Tuffier's Line, so long as this information could be extracted from the reported results. Exclusion criteria included: reporting results in a manner that precluded calculation of mean location of TL and its standard deviation<sup>26-28</sup>; cadaveric studies<sup>29-31</sup>; studies on the accuracy of identifying L4 or L4-5 but not in relation to TL<sup>14,32</sup>; studies with a very small sample size<sup>18</sup>; studies utilizing a non-imaging reference standard<sup>33</sup>; and review/commentary articles. Databases consulted included PubMed, ICL, CINAHL, AMED, Osteopathic Research Web, OstMed, and MANTIS.

After searching these biomedical databases, the investigators supplemented the search using the global Google search engine. Searches were conducted using the following terms and/or combinations of them: Tuffier\*, intercristal line, intercrestal line, Jacoby's line, lumbar spine, lumbar landmark, pelvic landmark, palpation, and TL. It was not necessary to construct complicated Boolean phrases to limit the number of returned citations, because even very inclusive search terms returned relatively few citations; e.g., "Tuffier's line" (likely to capture many of the relevant citations) returned only 28 citations. The "related citations" function was deployed when articles were retrieved which fit the inclusion criteria, and additional citations were harvested from included articles. Each of the included palpation articles was rated for quality using a modified version of the QUADAS instrument<sup>34</sup>, which usually includes 14 assessment criteria. We excluded one item having to do with the time period between the index and reference standards, which did not appear relevant to studies concerned with a spinal landmark association rather than diagnostic accuracy. The highest attainable score in the modified QUADAS instrument was thus 13. We used a modified version of the Arrivé instrument for the methodological quality of the included imaging studies<sup>35</sup>, which usually has 15 criteria. In the Arrivé instrument four of the 15 assessment criteria involve a reference standard related to the imaging procedure. Since this meta-analysis concerns a spinal landmark association rather than diagnostic accuracy, these four criteria were deemed irrelevant and were dropped. Thus, the lowest attainable score for an article was 11 and the highest was 33, where the lower the score, the higher the quality. The included articles were rated for quality by two reviewers; disagreements between reviewers were resolved by coming to consensus following email exchanges.

For the purpose of meta-analysis (conducted using Open Meta-Analyst, (http://www.cebm.brown.edu/openmeta/), the investigators abstracted the following data from each included article: sample size, mean location of either the iliac crests (palpation articles) or of TL (imaging articles), and standard deviation. When an included article reported data separately for subsets of the data (e.g., males and females, or obese and non-obese patients), these data were regarded as separate studies for the purpose of meta-analysis, provided different study populations were used for each. However, when different imaging procedures were conducted on the same study population, only the study using the most characteristic protocol (e.g., a flexed rather than extended patient positioning procedure) was entered into a meta-analysis, to avoid overweighting that population. On the other hand, the investigators scrutinized these other parameters for the purpose of qualitative analysis. In addition to performing meta-analysis on the full datasets for both the palpation and imaging studies, we conducted other analyses on subsets stratified by patient gender, age, and pregnancy/ post-partum status. There were too few studies available of each type to conduct meta-analysis based on type of imaging (MRI, CT scan, ultrasound, fluoroscopy, x-ray), type of patient positioning (standing, supine, prone, or lateral decubitus), or body mass index (BMI). Since the authors of two palpation studies<sup>17,36</sup> stated that their results might have been biased since the palpators were aware of prior studies that consistently showed the spinal site of the palpated TL cephalad to the imaged TL, we attempted meta-analysis with and without these studies included. However, since the results were very similar, we elected to include these studies in the meta-analyses. Very few of the studies reported the spinal level associated with TL using the spinous process as a reference point; most used the intervertebral interspace, which is more relevant in the practice of anesthesiology. When data were presented as to whether TL intersected the upper, middle, or lower portion of a vertebral body<sup>37,38</sup>, we collapsed the data to make it compatible with the great majority of the studies, which reported the intersection as occurring either within a vertebral body or at an intervertebral interspace.

#### Results

The retrieval process is summarized using the PRISMA flow diagram (Figure 1). The original search retrieved 1301 citations. After excluding 179 duplicates, 1122 unique citations remained. After inspecting their titles, 1067 were excluded from further consideration, leaving 55 abstracts to be read for consideration of possible inclusion. This resulted in the retrieval of 47 full text articles; five additional full text articles (bringing the total to 52) were retrieved either based on a secondary search of the included full text articles, searching with the Google database, or because the first author was personally familiar with them having earlier conducted a literature search on a related subject. Ultimately, 12 palpation articles and 21 imaging articles fit the inclusion criteria and were stratified into various meta-analyses. Six of the included articles7,20,21,25,36,39 contained both radiological and palpation arms. Thus 27 unique articles were included and entered into the study and included in Table 2, which summarizes these articles and includes quality ratings.

Among the imaging studies that included an arm addressing the ability of a palpator to numerate the lumbar spinal level associated with TL, one of them<sup>10</sup> did not provide sufficient detailed information to be included among the palpation articles. Some of the included palpation studies<sup>8,13,15-18,40</sup> reported on the spinal level that was associated with the crests, but did not address the actual spinal level of TL.

Several of the included studies considered the impact of various demographic variables on the location of either the crests or the imaged TL. Although some authors found age insignificant<sup>10,15,41</sup> in adult patient cohorts, and one study found the same in a pediatric cohort<sup>42</sup>, other authors found that the imaged TL was more cephalad in an elderly population<sup>6,23,25,38</sup> and among older pediatric patients<sup>37</sup>. Several studies determined the palpated crests or imaged TL to be more caudal in females <sup>7-9,23,26,27,41,43,44</sup>, although gender was insignificant in a pediatric population<sup>37</sup>. Sagittal plane posture (extension, flexion, or neu-

Spinal level Subjects Lower Upper I^2 % n Row Palpation studies 3.56 1 15 All, exclude pediatric 3.01 3.72 93.5 2 8 Females, all exclude pediatric 3.39 3.16 3.61 92.6 3 3 Males, all, exclude pediatric 3.43 3.36 3.50 5.3 Imaging studies 4 23 All, excluding pediatric, pregnancy 4.35 4.28 4.42 96.1 5 4.44 91.3 6 Females, PP, pregnant, pediatric excluded 4.39 4.51 6 7 Males, pediatric excluded 4.13 4.02 4.25 93.1 7 3 Pediatric studies only 4.86 4.81 4.92 69.1 \*Explanation regarding spinal numeration: "4.5" = L4-5 interspace, so that 4.75 would be more caudal Most representative differences Delta Interpretation Imaged vs. palpatory crest, genders Palp crest almost 1 level cephalad, 8 0.88 combined (mean 5-6 minus mean 2-3) average for both genders Palp crest cephalad 0.7 levels more 9 0.70 Imaged vs palpatory crest, males (3-6) cephalad to crests in males Palp crest almost one level cephalad to 10 Imaged vs. palpatory crest females (2-5) 1.05 crests in females TL nearest L4 in males, nearest 4.5 11 Imaged crest males vs. females (5-6) 0.31 females Male/ female palpatory crests near L3.5, 12 0.04 Palpatory crest males vs. females (2-3) slightly higher in males. Abbreviations: n= number studies included; PP=post-partum; ped=pediatric

Table 1.

Meta-analyses and most representative differences (higher numbers more caudal spinal level\*).

tral) had a modest impact in which TL became more caudal in flexion<sup>4,,22,36</sup>; Lin<sup>25</sup> on the other hand did not find flexion in patient positioning to have this effect. Even though Shiraishi<sup>38</sup> reported that flexion lowered TL compared with extension in males, the findings as reported in percentage terms are at best heuristic; in our secondary analysis, we found the change clinically insignificant, with the mean spinal level of TL in males in flexion dropping only minutely from L4.21 to L4.26. In agreement with our analysis, Snider 9 did not report a flexion effect. Pregnancy shifted TL cephalad<sup>24,45</sup>. Obesity or elevated BMI tended to diminish accuracy in palpation according to Broadbent<sup>13</sup> and raised the spinal level of TL <sup>7,25,37</sup>. Although weight and height had an equivocal impact on the location of the spinal level of the palpated crests, a variety of overlapping factors – increased body mass index<sup>7</sup>, obesity<sup>10,13,36</sup>, and abdominal circumference<sup>25</sup> – resulted in the palpatory TL level being at a more cephalad spinal level.

Table 1 summarizes the results of a number of representative meta-analyses performed on various subsets of the data. The spinal levels corresponding to either the iliac crests in palpation studies or TL in imaging studies are reported as "L" followed by a number with 2 decimal places. For example, "L3.45" indicates that the spinal level was a little cephalad to the L3-4 interspace. Although the status of being pregnant or post-partum impacted imaging results for females, the lack of an effect while identifying the spinal level associated with crest palpation allowed us to include these cohorts in the meta-analytic results for the female palpation articles. Table 1 includes several rows that compare representative means of the imaged and palpatory crests. Figure 6 provides the means and 95% confidence intervals for the imaged vs. palpatory spinal levels corresponding to Tuffier's Line for representative studies.

The most representative of the meta-analyses performed are provided in Figures 3-5, which includes forest plots summarizing the data. Figure 3 includes all non-pediatric

Study	Exam method(s)	Subjects	Imaging/palpatory technique	Findings and comments	Methodological quality*
Amin (2014)	Р	N=100, mean 39yrs, 72.6kg, M 94%, 6 surg.	FL, X	Radiological and US assessments agreed; palpatory TL about ½ level cephalad. Apparent mislabeling of data columns.	12 (Q)
	FL	N=100, mean 39yrs, 72.6kg, M 94%, 6 surg.	Patient supine		16 (A)
	X 1	N=100, mean 39yrs, 72.6kg, M 94%, 6 surg.	?		
	US	N=100, mean 37yrs, 71.2Kg, M 100%	Patient seated		
	X 2	N=100, mean 37yrs, 71.2Kg, M 100%	?		
Baxter (2016)	Р	N=30, neonates, gender	US, Left lateral decubitus	Palpatory crest cephalad to radiological prior reports.	11 (Q)
Broadbent (2000)	Р	N=100, excluded boney tenderness, spinal deformity; in which 7 cases were palpated for the spinal level of TL; age <16 yrs, mean BMI 6, mean 73kg, gender not reported.	MRI, patients received seated and flexed palpatory assessment	Accuracy unaffected by patient position (sitting or lateral), Markers placed far from crest more likely to be misidentified. Obesity impaired accuracy.	11 (Q)
Chakraverty (2007)	Р	N=49 females	Prone	Mean 45 years, mean 73.3kg, Palpated crest cephalad to TL, and more cephalad spinal levels likely identified with increasing patient BMI. The PSIS line identified the S2 spinous process in 51% of cases.	13 (Q)
	Р	N=26 male	Prone		
	F	N=49 females	Prone		20 (A)
	F	N=26 male	Prone		
Chin (2005)	Х	N=48 surgical patients, 62% female, mena 58yrs	Standing lateral, higher crest used as landmark	Slight trend for the L4/L4–L5 level to shift cephalad relative to the iliac crest in flexed/prone position.	19 (A)
Farshad (2015)	MRI	N=62, 55% female, 5 with lumbosacral transitional segments	Coronal evaluation	Crest more implementable and accurate than other anatomical landmarks for correct lumbar numbering with lumbosacral transitional anomalies.	16 (A)
Furness (2002	Р	N=49, 62% female, mean yrs 45, mean BMI 26	Lateral decubitus, knees flexed	Anaesthetists were aware of prior findings and may have been biased; higher rate of caudal errors than most other studies. US also compared with x-ray, data not herein provided. One x-ray could not be marked.	12 (Q)
Horsanali (2015)	Х	N=317 females, 20-99 yrs, 8 brackets similar numbers per bracket	AP radiographs, supine positioning	Age did not impact imaged crest, unlike Rahmani 2011 and Kim 2007.	19 (A)
	Х	N=273 males, 20-99 yrs, 8 brackets similar numbers per bracket	AP radiographs, supine positioning		
Jung (2004) Age unrelated to spinal level of imaged TL.	Х	N=100 volunteers, 50% female	Supine, lateral decubitus	With lumbar flexion, the imaged crest moved caudally.	22 (A)
	Х	Same	Lateral fully flexed x-rays		
Kim (2003)	MRI	N=690, 49.7% female, age>20, mean 63kg	MRI. Patients supine, legs elevated.	The positions of the conus medullaris and Tuffier's line were lower in women than in men and higher with sacralization. Although transitional vertebra effect position of the conus medullaris and Tuffier's line, the safety margin is unchanged. With increased age the conus was lower and TF line was higher.	21 (A)
Kim (2007)	Р	N=72, 73.6% females, males mean yrs 25.4 and mean BMI 21.9, female mean yrs 36.2 and BMI 20.9	Prone	Interexaminer reliability of palpation was significantly greater for PSIS level than for the iliac crest.	11 (Q)
	Х	Same as P group.	PA x-rays		20 (A)
Kim (2014)	US	N=40 females, pregnant	Lateral decubitus	Compared palpated crest with vertebral levels established by US. Vertebral levels were more cephalad in the pregnant women compared to the non-parturient women.	20 (A)
	US	N=40 females, non-pregnant	Lateral decubitus		
Kuhns (1978)	Х	N=50, patients undergoing excretory urography. Mean BMI 25.8, mean weight 68.2kg	Supine, knees flexed	Data reported for 52 patients, although method states $N=50$	26 (A)

## Table 2.Data summary

Study	Exam method(s)	Subjects	Imaging/palpatory technique	Findings and comments	Methodological quality*				
Lee (2011)	Р	N=51 pregnant patients	Seated	BMI did not correlate with a greater disparity between the clinical estimates and ultrasound- determined levels. US more accurate than palpation to identifying the lumbar interspaces.					
	US	Same as P group.	Seated						
Lin (2015)	Р	N=52 volunteers, 67.3% females. Males mean yrs 46.9, BMI 23.4; females mean yrs 48, BMI 23.4	Left lateral decubitus, flexed.	Patients with smaller abdominal circumference, lower BMI, and younger patients had actual intervertebral levels lower than the palpated level. Radiographic TL in relation to spine did not change from lateral to hyperflexion positions.	16 (A)				
	Х	Same as P group	Left lateral decubitus, flexed						
	Х	Same as P group.	AP, patient supine						
McGaugh (2007)	СТ	N=50 females, mean 47.2 yrs	CT, supine, no flexion	Age unrelated to spinal level of imaged TL. PSIS more consistent spinal reference landmark for iliac crest.	19 (A)				
	СТ	N=50 males, mean 47.5 yrs	CT, no flexion						
Pysyk (2010)	Р	N=64 females. Mean 45.8 yrs, BMI 25.8	US, seated, flexed	Compared palpated crest with vertebral levels established by US. Greater proportion males-to- females with a more cephalad palpated compared to imaged TL. Males and taller individuals had palpated TL as high as ZL2-3, similar to reports of Kim, 2003 and Snider 2008.	11 (Q)				
	Р	N=50 males. Mean 45.3 yrs, BMI 27.0	US, seated, flexed						
Render (1996)	Х	N=163	PA supine, hips flexed	TL was <l4 17.8%="" 3.7%="" and="" at="" in="" l3-4="" of="" patients.<="" td=""><td>27 (A)</td></l4>	27 (A)				
Sahin (2014)	Р	N=50 lean pregnant. Mean yrs 31, BMI 26.4	US	Compared palpated crest with vertebral levels established by US. The palpatory crest level of the spine was more cephalad in larger BMI patients.	13 (Q)				
	Р	N=50 obese pregnant. Mean yrs 32.4, BMI 34.1	US						
Sargin (2015)	Х	N=204 female children <16 yrs	Standing AP X-rays	TL was related to age, more caudal in younger children. No differences based on gender. Unlike Tame, 2003, reported spinal level of TL in range L4 to L5-S1.	19 (A)				
	Х	N=315 male children <16 yrs	Standing AP X-rays						
	Х	N=48 females	Standing, neutral	Mean age both genders combined 48.3 yrs. Could not determine if AP, PA, or lateral radiography. No difference between males and females between extension or flexion, whereas in flexion the spinal level of TL became more caudal in males. Spinal level of TL more cephalad in older patient due to spinal degeneration.	20 (A)				
	Х	N=48 females	Standing, flexed						
Shiraishi (2006)	Х	N=48 females	Standing, extended						
Shiraishi (2000)	Х	N=52 males	Standing, neutral						
	Х	N=52 males	Standing, flexed						
	Х	N=52 males	Standing, extended						
Shlotterbeck (2008)	Р	N=99 pregnant females, in which 20 cases were palpated for the spinal level of TL at L4-5	US assessment performed in same position as one in which block performed.	US used to determine spinal level of needle insertion during neuraxial anesthesia. Six punctures were actually carried out at the L1/L2 intervertebral space.	11 (Q)				
	Х	N=33 female	PA prone	Age not recorded, but range 2-45 yrs. No clinical data available, for x-ray arm. Mean BMI 25.9 palpatory arm. BMI did not impact spinal location of TL. No difference in standing and prone results.	19 (A)				
0.1.(2000)	Х	N=27 male	PA prone						
Snider (2008)	Х	N=100 females	PA standing						
	Х	N=100 males	PA standing						
Tame (2003)	MRI	N=49 children, both genders, <10 yrs	Supine	No clinical information provided. No age effect.	21 (A)				
Tanaka (2013)	Р	N=967 C-section cases, in which 20 cases were palpated for the spinal level of TL	X-ray, right lateral decubitus	Abdominal x-ray confirmed level of insertion of epidural catheters. Palpation not reliable for spinal level of TL.	13 (Q)				
Walsh (2011)	Х	N=450, 20-90 yrs,	AP and lateral x-ray	Spinal level of TL increased with age.	19 (A)				
Wattanaruangkowit (2010)	Х	N=270, 6 age brackets 2-80 yrs.	AP and lateral x-ray	No clinical information provided other than age. Spinal level of TL increased with age.	22 (A)				
Whittey (2008)	Р	N=121 post-partum, in which 22 cases were palpated for the spinal level of TL	US, seated	The backs of subjects were examined to identify the puncture site of the spinal or epidural needle. No other clinical information provided.	13 (Q)				
(A) = Arrivé for imaging studies, n/33, lower scores, higher quality. QUADAS for palpation studies, n/13, higher numbers, higher quality									
Abbreviations: P=palpation study; X=x-ray, MRI=magnetic resonance image; CT= computed tomography; F=fluoroscopy; US=ultrasound									



Figure 3. Palpatory crests (pediatric excluded)



Figure 4. Imaged crest, females (excluding pediatric, pregnant/post-partum)



Figure 5. Imaged crest (males, excluding pediatric)

palpation articles. Figure 4 includes imaged crest studies for females, exclusive of pediatric and pregnancy/postpartum patient cohorts. Figure 5 includes the imaged crest studies for males, exclusive of pediatric studies.

The forest plots in Figures 3-5 include a vertical line that represents the central tendency of the combined data, the Grand Mean. The horizontal lines then illustrate the individual study means for the spinal level of the imaged or palpated iliac crests, and their 95% confidence intervals. The diamond represents the point estimate and confidence interval for the combined studies.

#### Discussion

Meta-analytic methods usually calculate the odds ratios for alternative treatments, assigned to experimental and control groups. In that scenario, the forest plot includes a vertical line intersecting the X axis at an odds ratio of one. The individual studies are then mapped to show their mean effects and confidence intervals, which makes it easy to see whether their odds ratio is above or below 1.0, and thus whether the control or experimental treatment is favored. Since we were not looking at alternative treatments, but rather mean spinal levels and 95% CIs, the forest plots in our study include a vertical line that represents the central tendency of the combined data, the Grand Mean. The horizontal lines then illustrate the individual study means and their CIs. Most authors would suggest that overlapping lines suggest studies that show statistical agreement. We pooled data from several smaller studies to establish the relationship of the spinal level identified by iliac crest palpation and that of the imaged TL, including sub-analyses based on demographic and other characteristics. To select the most representative population for determining the difference between the spinal level of the imaged and palpated iliac crests (row 8, Table 1), we excluded studies featuring post-partum and pregnant females; as well as pediatric patients. In addition to reporting the data by gender, we also reported means for combined male and female cohorts. The imaged TL was closest to the L4-5 interspace while the palpated TL was closest to the L3-4 interspace, corresponding to about a one-level difference. The female imaged crest line is at L4.44 (row 5) whereas the imaged male crest line is slightly higher at L4.13 (row 11). The female and male palpatory crests are both very similar, very close to the L3-4 interspace (rows 2 and 3, respectively).



Figure 6. Imaged vs. palpatory spinal levels corresponding to Tuffier's Line.



Broadbent accuracy data, reinterpreted.

Even though the imaged TL is 0.31 levels more caudal in females than in males (4.44 females, 4.13 males), as seen in row 11, the palpated crest line is virtually the same at the L3-4 interspace for both genders (3.39 females, 3.43 males), as seen in row 12. Chakraverty<sup>7</sup> explains why the discrepancy between the imaged TL and palpated TL is greater in women: "Adult females who have more percentage body fat for equivalent BMIs, different fat distribution and who develop progressively greater waist to hip ratios with age than adult males were found to have proportionately more higher levels identified on palpation." The palpator's fingers overlay soft tissue situated above the iliac crests, especially in obese patients or those with relatively abundant subcutaneous fat tissue relative to their body mass index.<sup>7,21-25</sup>

Gallagher *et al.* <sup>46</sup> found that for an equivalent body mass index, women have significantly greater amounts of total body fat than men. The palpated crests in females at L3.39 is slightly more than 1 level higher than the imaged

TL at 4.35 (row 10); while the palpated TL in males at L3.43 is 0.73 levels higher than the imaged TL at L4.13. To get a sense of the relationship between the imaged and palpated TLs for a mixed population of males and females, row 8 compares the means of males and females combined. Irrespective of gender, the spinal level of the palpated crests is almost 1 level more cephalad than that of the imaged TL.

These results suggest that using the palpated crests to determine the site for anesthetic injections can be a very hazardous practice. The safety and success of epidural blocks and cerebrospinal fluid taps depends on accurate palpation of spinal levels<sup>23,26,42</sup>. The greatest risk lies in puncturing the conus medullaris<sup>47</sup>, which on average extends to the lower portion of L1 but may reach the upper portion of L3 <sup>48</sup>. This is very close to the level of the palpated crests in our secondary analyses. Although this is not of pressing concern for the manual therapist, the success of diagnostic and therapeutic interventions in manu-

al therapy may depend to some extent on accurate spinal palpation. The process of identifying spinal targets of intervention in the upright position, and then attempting to locate those same spinal levels in the prone position, can be quite challenging<sup>49</sup>.

Previous studies have shown low accuracy for palpators in numerating lumbar levels and the mistakes tended to be cephalad<sup>7,13-17</sup>. This is clearly due to confounding the imaged and palpatory crest levels in relation to the lumbar spine. It would be instructive to reinterpret the accuracy achieved in these studies after having adjusted for the erroneous landmark rule wherein the palpated crest was assumed equivalent to the imaged TL; the accuracy would then be seen to have been higher than had been previously reported. As an example, Broadbent et al.13 reported that among 200 attempts by anesthetists' to accurately identify a lumbar spinal level, only 29% were accurate. Although this may appear to indict the anesthetists' palpatory skill per se, their error was in part explained by the erroneous landmark rule. Had they understood that the palpatory crest line was closer to the L3-4 level, their accuracy rate would have been reported to be 50.5%. Figure 7 shows the actual error rate as reported, as compared with what would have been reported based on better understanding of the spinal location of the palpatory crests.

A similar confusion has confounded the literature on the accuracy of thoracic spine palpation. Cooperstein has demonstrated that using the scapula to localize thoracic landmarks is very error prone, especially given the inadequacy of spinal landmark rules that are based on conventional wisdom rather than clinical research<sup>50-53</sup>. For instance, the inferior scapular tip lies closer to the T8 rather than T7 spinous process, as is commonly thought.

It is very likely that health professionals, both manual therapists and others, have been diagnosing and treating patients in part by associating spinal levels with scapular and iliac crest landmark rules that are now known to be inaccurate. In manual therapy, an intended site of spine care is often principally determined by history and physical examination, including assessment of positional asymmetry, pain/tenderness, and joint movement capacity. In such cases, it may not be crucial for an individual provider to know the exact level being treated. However, in a clinical setting with multiple providers, charting errors in numerating diagnosed and treated spinal levels could lead to improper care due to addressing non-intended levels. In addition, when physical examinations are obtained in the seated or standing position, or are in part based on imaging or specific neurological findings, it may be challenging to locate the intended spinal level in the prone position.<sup>49</sup> These errors could lead to sub-optimal clinical outcomes, depending on the degree to which specificity in identifying sites of spine and sacroiliac care is clinically important. While correct utilization of landmark associations for the determination of a particular spinal level is plausibly important to the manual therapist, they are crucial to the anesthetist trying to identify safe and accurate locations for epidurals and other injection procedures.

This present study had several limitations. Some of the I^2 % values indicated a high degree of heterogeneity among the included studies; no doubt due to differences in the imaging technology used, the patient positioning in the imaging and palpation studies, the way the palpatory procedures were performed, and differences in the selected patient populations. The need to convert data reported for segments of a vertebra (upper, middle, lower) in some studies, rather than for discrete vertebral or intervertebral levels, may have also increased heterogeneity. In addition to explaining the variability in the data, more importantly this could compromise accuracy in clinical practice, which is something anesthesiologists and manual therapists should take into consideration. Since the QUADAS quality scores for the palpation articles were uniformly high (mean=11.8/13, s=0.94), it would have been fruitless to attempt interpreting results based on study quality. Likewise, the Arrivé scores were quite uniform (mean=20.4/33, s=2.84). Since we used the included articles only to determine spinal levels corresponding to the imaged or palpated iliac crests, and were not concerned with diagnostic accuracy, various criteria in the QUADAS and Arrivé instruments were irrelevant in our study and thus excluded. As a result, the quality scores in our study cannot be compared to possible ratings by other reviewers, who may have used all the assessment criteria. The Render study<sup>12</sup> stood out as of lower quality than the others, with a quality score of 27/30. That stated, the reported value of L4.34 for TL was in line with the estimate of L4.45 reported by other imaging studies including both genders. As is often the case in meta-analysis, some of the articles, for both the imaging and palpation studies, showed statistically different study outcomes. That did not obscure our central finding, which is the tendency in

most patient populations for the spinal level of the palpated TL to lie cephalad to the spinal level of the imaged TL. In no subject population does the 95% confidence interval for the spinal level associated with the iliac crests overlap that of the imaged TL (Figure 6)., confirming statistically different mean spinal levels.

One way of increasing accuracy in numerating lumbar vertebral levels would be for practitioners to deploy a revised landmark association whereby the spinal level, as identified using the palpatory iliac crest method, will usually be the L3 spinous process or the L3-4 interspace. However, the variability noted in these meta-analyses suggests that there may be an even better strategy: since the 2nd sacral tubercle is very dependably situated between the posterior superior iliac spines7,20,44, numerating lumbar levels based on this landmark association might be most accurate. Finding S2 in this manner, a palpator would then identify the most immediately cephalad sacral tubercle as S1, and then an equal distance cephalad to this would be the soft depression corresponding to the hypoplastic L5 spinous process, followed by the L4 spinous process (which will likely be caudal to the palpated iliac crests), followed by the L3 spinous process, and so on.

#### Conclusion

This study confirms reports by previous authors that the spinal level of the palpated crests is cephalad to the imaged TL, and this difference was quantified as being just about 1 spinal level. The difference is greater in females than males. Past studies on the accuracy of lumbar static palpation should be re-interpreted considering these findings, since reported accuracy rates in those studies assumed that TL corresponded to the L4 spinous process or the L4-5 interspace. A more accurate method is recommended to locate the TL spinal level, especially for higher risk procedures including the epidural blocks and cerebrospinal fluid punctures practiced by anesthesiologists. Clinicians would be well-served practicing according to an updated landmark association wherein the palpated crests will usually correspond to the L3-4 interspace; or clinicians may attempt to locate L4 or L4-5 by counting up from the 2<sup>nd</sup> sacral tubercle <sup>7,20,44</sup>; or by using the 10<sup>th</sup> rib<sup>22</sup> as a landmark. Using a combination of those methods would most likely be good clinical practice compared with abject reliance on any one method.

#### References

- 1. Omi S, editor. WHO standard acupuncture point locations in the Western Pacific Region. Manila, Phillippines: World Health Organization, 2008; 2008.
- Little GL, Merrill WH, editors. Complications in Cardiothoracic Surgery: Avoidance and Treatment. 2 ed. Hoboken: Wiley-Blackwell; 2011.
- 3. Tuffier T. The treatment of chronic empyema. Ann Surg. 1920;72(3):266-287.
- Kim JT, Jung CW, Lee JR, Min SW, Bahk JH. Influence of lumbar flexion on the position of the intercrestal line. Reg Anesth Pain Med. 2003;28(6):509-511.
- 5. Mullin L. Radiographic variability of the intercrestal line. J Chiropr Educ. 2006;20(1):37.
- 6. Wattanaruangkowit P, Lakchayapakorn K. The position of the lumbar vertebrae in relation to the intercrestal line. J Med Assoc Thai. 2010;93(11):1294-1300.
- Chakraverty R, Pynsent P, Isaacs K. Which spinal levels are identified by palpation of the iliac crests and the posterior superior iliac spines? J Anat. 2007;210(2):232-236.
- 8. Pysyk CL, Persaud D, Bryson GL, Lui A. Ultrasound assessment of the vertebral level of the palpated intercristal (Tuffier's) line. Can J Anaesth. 2010;57(1):46-49.
- 9. Snider KT, Kribs JW, Snider EJ, Degenhardt BF, Bukowski A, Johnson JC. Reliability of Tuffier's line as an anatomic landmark. Spine. 2008;33(6):E161-165.
- 10. Sahin T, Balaban O, Sahin L, Solak M, Toker K. A randomized controlled trial of preinsertion ultrasound guidance for spinal anaesthesia in pregnancy: outcomes among obese and lean parturients: ultrasound for spinal anesthesia in pregnancy. J Anesth. 2014;28(3):413-419.
- Shiraishi N, Matsumura G. What is the true location of Jacoby's line? Okajimas Folia Anat Jpn. 2006;82(4):111-115.
- 12. Render CA. The reproducibility of the iliac crest as a marker of lumbar spine level. Anaesthesia. 1996;51(11):1070-1071.
- 13. Broadbent CR, Maxwell WB, Ferrie R, Wilson DJ, Gawne-Cain M, Russell R. Ability of anaesthetists to identify a marked lumbar interspace. Anaesthesia. 2000;55(11):1122-1126.
- Snider KT, Snider EJ, Degenhardt BF, Johnson JC, Kribs JW. Palpatory accuracy of lumbar spinous processes using multiple bony landmarks. J Manipulative Physiol Ther. 2011;34(5):306-313.
- Tanaka K, Irikoma S, Kokubo S. Identification of the lumbar interspinous spaces by palpation and verified by X-rays. Revista brasileira de anestesiologia. 2013;63(3):245-248.
- Whitty R, Moore M, Macarthur A. Identification of the lumbar interspinous spaces: palpation versus ultrasound. Anesth Analg. 2008;106(2):538-540.
- 17. Schlotterbeck H, Schaeffer R, Dow WA, Touret Y,
Bailey S, Diemunsch P. Ultrasonographic control of the puncture level for lumbar neuraxial block in obstetric anaesthesia. Br J Anaesth. 2008;100(2):230-234.

- Parate LH, Manjunath B, Tejesh CA, Pujari V. Inaccurate level of intervertebral space estimated by palpation: The ultrasonic revelation. Saudi J Anaesth. 2016;10(3):270-275.
- Cooperstein R, Young M, Haneline M. Criterion validity of static palpation compared with a reference standard. J Chiro Educ. 2015;29(1):74.
- 20. Kim HW, Ko YJ, Rhee WI, Lee JS, Lim JE, Lee SJ, et al. Interexaminer reliability and accuracy of posterior superior iliac spine and iliac crest palpation for spinal level estimations. J Manipulative Physiol Ther. 2007;30(5):386-389.
- 21. Amin WA, Abou Seada MO, Bedair E, Elkersh MM, Karunakaran E. Comparative study between ultrasound determination and clinical assessment of the lumbar interspinous level for spinal anesthesia. Middle East J Anaesthesiol. 2014;22(4):407-412.
- 22. Jung CW, Bahk JH, Lee JH, Lim YJ. The tenth rib line as a new landmark of the lumbar vertebral level during spinal block. Anaesthesia. 2004;59(4):359-363.
- 23. Kim JT, Bahk JH, Sung J. Influence of age and sex on the position of the conus medullaris and Tuffier's line in adults. Anesthesiology. 2003;99(6):1359-1363.
- 24. Lee AJ, Ranasinghe JS, Chehade JM, Arheart K, Saltzman BS, Penning DH, *et al.* Ultrasound assessment of the vertebral level of the intercristal line in pregnancy. Anesth Analg. 2011;113(3):559-564.
- 25. Lin N, Li Y, Bebawy JF, Dong J, Hua L. Abdominal circumference but not the degree of lumbar flexion affects the accuracy of lumbar interspace identification by Tuffier's line palpation method: an observational study. BMC Anesthesiol. 2015;15:9.
- 26. Rahmani M, Vaziri Bozorg SM, Ghasemi Esfe AR, Morteza A, Khalilzadeh O, Pedarzadeh E, *et al*. Evaluating the reliability of anatomic landmarks in safe lumbar puncture using magnetic resonance imaging: does sex matter? Int J Biomed Imaging. 2011;2011:868632.
- Margarido CB, Arzola C, Carvalho JCA. The intercristal line determined by palpation is not a reliable anatomical landmark for neuraxial anesthesia. Can J Anaesth. 2011;58(3):262-266.
- Borghi B, Tognu A, White PF, Paolini S, Van Oven H, Aurini L, *et al*. Soft tissue depression at the iliac crest prominence: a new landmark for identifying the L4-L5 interspace. Minerva Anestesiol. 2012;78(12):1348-1356.
- 29. Lakchayapakorn K, Suwanlikhid N. The interlaminar and the narrowest distances at the L3/4 and L4/5 interspinous spaces and location of the intercrestal line in Thai cadavers. J Med Assoc Thai. 2014;97 Suppl 8:S1-6.
- Windisch G, Ulz H, Feigl G. Reliability of Tuffier's line evaluated on cadaver specimens. Surg Radiol Anat. 2009;31(8):627-630.

- 31. Ievins FA. Accuracy of placement of extradural needles in the L3-4 interspace: comparison of two methods of identifying L4. Br J Anaesth. 1991;66(3):381-382.
- 32. Cooper K, Alexander L, Hancock E, Smith FW. The use of pMRI to validate the identification of palpated bony landmarks. Man Ther. 2013;18(4):289-293.
- 33. Kettani A, Tachinante R, Tazi A. Evaluation of the iliac crest as anatomic landmark for spinal anaesthesia in pregnant women. Ann Fr Anesth Reanim. 2006;25(5):501-504.
- 34. Whiting P, Rutjes AW, Reitsma JB, Bossuyt PM, Kleijnen J. The development of QUADAS: a tool for the quality assessment of studies of diagnostic accuracy included in systematic reviews. BMC medical research methodology. 2003;3:25.
- 35. Arrivé L, Renard R, Carrat F, Belkacem A, Dahan H, Le Hir P, *et al*. A scale of methodological quality for clinical studies of radiologic examinations. Radiology. 2000;217(1):69-74.
- Furness G, Reilly MP, Kuchi S. An evaluation of ultrasound imaging for identification of lumbar intervertebral level. Anaesthesia. 2002;57(3):277-280.
- 37. Sargin M. Radiological evaluation of the Tuffier's Line in pediatric patients. J Clin Analyt Med. 2015:1-4.
- Shiraishi N, Matsumura G. Establishing intercrestal line by posture:--a radiographic evaluation. Okajimas Folia Anat Jpn. 2006;82(4):139-416.
- Hayes J, Borges B, Armstrong D, Srinivasan I. Accuracy of manual palpation vs ultrasound for identifying the L3-L4 intervertebral space level in children. Paediatr Anaesth. 2014;24(5):510-515.
- 40. Baxter B, Evans J, Morris R, Ghafoor U, Nana M, Weldon T, *et al*. Neonatal lumbar puncture: are clinical landmarks accurate? Arch Dis Child Fetal Neonatal Ed. 2016;101(5):F448-450.
- 41. Horsanalı BÖ, Tekgül ZT, Özkalkanlı MY, Adıbelli ZH, Esen ÖE, Durna FY. Radiological evaluation of the line between the crista iliaca (Tuffier's line) in elderly patients. 2015;43:149-153.
- 42. Tame SJ, Burstal R. Investigation of the radiological relationship between iliac crests, conus medullaris and vertebral level in children. Paediatr Anaesth. 2003;13(8):676-680.
- Hubley-Kozey C, Wall J, Hogan D. Effects of a general exercise program on plantarflexor and dorsiflexor strength and power of older women. Top Geriatr Rehabil. 1995;10(3):45-60.
- 44. McGaugh JM, Brismee JM, Dedrick GS, Jones EA, Sizer PS. Comparing the anatomical consistency of the posterior superior iliac spine to the iliac crest as reference landmarks for the lumbopelvic spine: a retrospective radiological study. Clin Anat. 2007;20(7):819-825.
- 45. Kim SH, Kim DY, Han JI, Baik HJ, Park HS, Lee GY, *et al*. Vertebral level of Tuffier's line measured by

Systematic review and meta-analyses of the difference between the spinal level of the palpated and imaged iliac crests

ultrasonography in parturients in the lateral decubitus position. Korean J Anesthesiol. 2014;67(3):181-185.

- 46. Gallagher D, Visser M, Sepulveda D, Pierson RN, Harris T, Heymsfield SB. How useful is body mass index for comparison of body fatness across age, sex, and ethnic groups? Am J Epidemiol. 1996;143(3):228-239.
- 47. Ohtsuka K, Kiyono S, Takimoto M. A potential hazard of spinal cord injury by spinal and epidural anesthesia in a patient with low-placed conus medullaris (author's transl). Masui. 1977;26(9):967-979.
- Saifuddin A, Burnett SJ, White J. The variation of position of the conus medullaris in an adult population. A magnetic resonance imaging study. Spine. 1998;23(13):1452-1456.
- 49. Cooperstein R, Young M. Mapping intended spinal site of care from the upright to prone position: an interexaminer reliability study. Chiropr Man Therap. 2014;22:20.

- 50. Cooperstein R, Haneline M. Spinous process palpation using the scapular tip as a landmark vs a radiographic criterion standard. J Chiropr Med. 2007;6(3):87-93.
- 51. Cooperstein R, Haneline M, Young M. The location of the inferior angle of the scapula in relation to the spine in the upright position: a systematic review of the literature and meta-analysis. Chiropr Man Therap. 2015;23:7.
- 52. Cooperstein R, Haneline MT, Young MD. The location of the inferior angle of the scapula in relation to the spinal level of prone patients. J Can Chiropr Assoc. 2009;53(2):121-128.
- 53. Haneline M, Cooperstein R, Young M, Ross J. Determining spinal level using the inferior angle of the scapula as a reference landmark: a retrospective analysis of 50 radiographs. J Can Chiropr Assoc. 2008;52(1):24-29.

## Intra-examiner reliability of measurements of ankle range of motion using a modified inclinometer: a pilot study

Patricia Tavares, BSc, DC, FCCOS(C)<sup>1</sup> Victoria Landsman, BSc, MA, PhD<sup>2,3</sup> Leslie Wiltshire, BSc, DC<sup>1</sup>

A modified inclinometer was designed for measuring total ankle range of motion (ROM) in the standing position for a large future study. The purpose of this pilot study was to assess the intra-examiner reliability of this new device in order to see if the examiner would be able to produce equally reliable measurements with this instrument as with a routinely used goniometer.

Nineteen young healthy individuals took part in the pilot. The same examiner took the ROM measurements using both devices twice on the same day and one further time 2 or 3 days later. Test-retest reliability was measured using the intraclass correlation coefficient (ICC). The ICC values were 0.86 (95% CI=[0.67; 0.94]) and 0.83 (95% CI=[0.61; 0.93]) for the measurements taken with the goniometer on the same day and for those

On a conçu un inclinomètre modifié pour mesurer l'amplitude du mouvement (ADM) totale de la cheville en position debout pour une grande étude à venir. Cette étude pilote a pour objectif d'évaluer la fiabilité des intra-examinateurs vis-à-vis de ce nouveau dispositif afin de constater si l'examinateur serait en mesure de produire des mesures d'une fiabilité équivalente avec cet instrument par rapport au goniomètre couramment utilisé.

Dix-neuf jeunes personnes en santé ont participé à l'étude pilote. Le même examinateur a pris des mesures de l'ADM avec les deux dispositifs à deux reprises le même jour et une autre fois deux ou trois jours plus tard. On a mesuré la fiabilité de test-retest au moyen du coefficient de corrélation intraclasse (CCI). Les valeurs de CCI étaient de 0,86 (IC à 95 %=[0,67; 0,94]) et de 0,83 (IC de 95 %=[0,61; 0,93]) pour les mesures prises avec le goniomètre le même jour et les mesures prises lors de deux jours différents. Les

<sup>1</sup> Canadian Memorial Chiropractic College

<sup>3</sup> University of Toronto, Dalla Lana School of Public Health, Department of Biostatistics

Corresponding author: Patricia Tavares Canadian Memorial Chiropractic College, 6100 Leslie Street, Toronto, ON M2H 3J1 e-mail: ptavares@cmcc.ca Tel: 416-482-2340

The authors do not have any conflicts of interest to declare with regard to the preparation of this manuscript. Financial support for this study was received from the Canadian Memorial Chiropractic College research department to cover equipment costs. © JCCA 2017

<sup>&</sup>lt;sup>2</sup> Institute for Work and Health

on two different days. The corresponding values for the modified inclinometer were 0.88 (95% CI=[0.72;0.95]) and 0.81 (95% CI=[0.57; 0.92]). Both instruments were found to have very good test-retest reliability.

(JCCA. 2017;61(2):121-127)

KEY WORDS: ankle, range of motion, pilot study, reliability, inclinometer

#### Introduction

Ankle motion is required for humans to walk, run, sit down, climb, etc. A limitation in range therefore may affect quality of life.<sup>1</sup> Reductions in ankle range of motion (ROM) have been associated with co-morbid conditions such as venous ulcers. This association has been established in populations with high socioeconomic status<sup>2,3</sup> where the prevalence of venous ulcers is approximately 1% according to Fowkes FG *et al.*<sup>4</sup>. It is estimated that in Western countries 3 billion dollars is spent annually on the care of venous ulcers.<sup>1</sup> Neither a similar association nor prevalence has been established in a socioeconomically disadvantaged population where the economic burden of venous ulcer care would likely be significant.

We, the authors, plan to assess patients attending mobile chiropractic clinics across the Dominican Republic with the intention of investigating the association between venous disease and venous ulcers and ankle ROM in a socioeconomically disadvantaged population. It is expected that such a study would require a large number of ROM measurements to be taken in a short amount of time, in field conditions with various practical obstacles, in which traditional methods of measuring ankle ROM are not feasible. As a result a sturdy measurement tool would be required.

Goniometers are traditionally used to determine ROM of the ankle joint.<sup>5</sup> Goniometers are considered valid and reliable clinical tools for assessing range of motion of joints of the extremities.<sup>6</sup> A typical goniometric measurement of the ankle is made with the patient's leg supine on the treatment table and the fulcrum at the lateral malleolus whilst maintaining the bottom rod of the goniometer parallel with the tibia and fibula. This procedure requires skill on the part of the examiner and visual estimation

valeurs correspondantes pour l'inclinomètre modifié étaient de 0,88 (IC de 95 %=[0,72;0,95]) et de 0,81 (IC de 95 %=[0,57; 0,92]). On a constaté une très bonne fiabilité de test-retest avec les deux instruments.

(JCCA. 2017;61(2):121-127)

MOTS CLÉS : cheville, amplitude du mouvement, étude pilote, fiabilité, inclinomètre, chiropratique

of the same position of the goniometer arms at the same starting position at each successive measurement.<sup>7</sup> Measuring ROM with a standard plastic goniometer does not control the patient's ability to flex or extend the toes or control for the subtalar range of motion and its influence on ankle ROM. Belczak *et al.*<sup>8</sup> used a goniometer with a plantar support to eliminate the influence on ankle ROM of the other articulations of the foot on ankle ROM. Despite the assumption that the instrument may have been cumbersome to use in the field, the authors were unable to acquire it for testing.

More recently Thornton *et al.*<sup>9</sup> introduced a digital goniometer as an inexpensive, reliable and valid method of measuring functional ROM of the ankle, with the patient's foot on the floor. Although the results are interesting and promising, the proposed measurement methodology would have proved challenging in the conditions that were expected to be seen in the Dominican study. There, clinical conditions were expected to vary from day to day, with uncertain floor surfaces ranging from concrete to dirt. Moreover, the results of this study were not available at the time of the authors' study.

There are a variety of studies that use inclinometers to measure ankle ROM. According to Gerhardt *et al.*<sup>7</sup>, inclinometers "read angle position relative to gravity or to a set neutral -0- position". Inclinometers for measuring ankle ROM, specifically ankle dorsiflexion<sup>10</sup> have shown to be reliable, and functional plantarflexion has been shown to be best quantified using an inclinometer on the dorsum of the foot in modern dancers<sup>11</sup>. To the best of their knowledge, the authors are not aware of another published study that established the reliability and validity of the full ROM of the ankle joint using an inclinometer.

A regular inclinometer does not control for subtalar



Figure 1. Goniometer



Figure 2. Modified inclinometer

joint contribution to ankle ROM even with an extension arm as described by Gerhardt *et al.*<sup>7</sup> The extension arm described by Gerhardt was rather narrow in width, and would not allow a broad contact with the foot, which would reduce the subtalar contribution.<sup>12</sup> For this purpose, the authors designed a device with a wider plantar support to help to control the amount of toe and foot contribution to the ankle ROM. Furthermore the device needed be robust enough for repeated use on many participants. A detailed description of the modified inclinometer is provided in Methods and Materials.

Since the new device is a modified version of an existing inclinometer with an extended arm, we aimed to assess the intra-examiner reliability of the device. Secondarily we wanted to determine if the intra-examiner reliability of this new device was of a similar magnitude to that of a standard goniometer.

#### Methods

Ethical approval for this study was obtained from the Office of Research Administration of the Canadian Memorial Chiropractic College (CMCC). The REB certificate number was 1303X05.

#### Study participants

Participants were fourth year interns from the Canadian Memorial Chiropractic College. Participants were excluded if they had suffered an injury to the low back or lower extremity, including the ankle, within the last week. A recent low back injury may have hindered participants from sitting comfortably and dorsiflexing the ankle in one of the required positions, ie. sitting with a straight leg on the table as part of the goniometer measurements, as will be described.

Nineteen participants, seven males and twelve females, volunteered and were eligible to participate. They represent a homogenuous group of young generally fit individuals, of approximately the same age. Participants were asked to sign consent forms before participating. Participants were asked to maintain their usual level of activity during the entire duration of the study to eliminate bias in consecutive ROM measurements due to injury.

#### Instruments

#### Goniometer

The goniometer used was a usual plastic type 12 inch goniometer by Almedic from Montreal Canada #32-4 (see Figure 1).

#### **Modified inclinometer**

The new device consisted of a Baseline® bubble inclinometer made by Fabrication Enterprises Inc. attached to a long wooden stick designed by authors PT and LW (hereafter, a modified inclinometer). A device used by both Gerhardt<sup>4</sup> and Lea *et al.*<sup>13</sup>. was the original concept for this new apparatus. The apparatus consisted of a straight edge wood base 30 cm long, 3.8 cm wide and 0.95 cm thick. This was cut level, and thick enough not to distort while using the device for multiple measurements. A notch was cut into one end to hold the inclinometer, 8.3 cm long and 0.79 cm wide. The inclinometer was secured to the straight edge, level to the bottom (See Figure 2).

## Participant Position and Procedure Goniometer

The participant was seated with the right leg supine, knee straight, on a chiropractic table, and the other leg off the table. The goniometer's pivot was centered over the ankle (lateral malleolus), and one arm paralleled the fibula and tibia. The other arm followed a line parallel to the 5<sup>th</sup> metatarsal. The patient was asked to actively dorsiflex and plantar flex the ankle from a starting position with the foot relaxed (considered the zero neutral position) with angle measurements taken at each point of dorsiflexion and plantar flexion. The participant was asked to not dorsiflex or plantar flex the toes if possible. The neutral starting positon was returned to after dorsiflexion before commencing plantarflexion. Total ROM was measured as the sum of these two individual measurements. The test was repeated with the knee flexed to 45 degrees with a pillow under the supine knee. The average of the two total ROMs, one with the knee straight and one with the knee bent, was used to calculate the final value for the ROM using the goniometer<sup>5</sup>.

#### **Modified Inclinometer**

The participant was in a standing position with the right knee on a chiropractic table, a modified position from that of Gerhardt's<sup>7</sup> method. The bent leg was positioned such that 50% of the lower part of the leg was on the surface of the table and 50% of the leg was off the table. The participant was asked to weight bear with the standing leg, not with the bent leg. The wooden arm was placed on the foot, with the foot in the most relaxed position possible. The inclinometer was placed at the heel so that the foot and toes made full contact with the wooden base. The contact was maintained in an effort to reduce the subtalar movement that might alter the ankle ROM.<sup>12</sup> The inclinometer was turned to establish this relaxed position as the zero degree position. The examiner held the bar along the base of the foot, and from the established zero degree position the participant moved the foot into maximum dorsiflexion without using the toes as he/she had been instructed to do. The angle of dorsiflexion was measured. The foot was then moved into the relaxed position again and the inclinometer was reset to zero. The participant was instructed to move into plantar flexion in the same way (See Figure 3). Total range of motion was then determined to be the addition of plantar flexion and dorsiflexion from the established zero position.

#### Measurements

In total, three ROM measurements were taken on each participant using the goniometer and the modified inclinometer each in turn following the procedures described above. Two measurements (Measurement 1 and Measurement 2) were taken on the same day, a few hours apart, and the third measurement (Measurement 3) was taken two or three days later, dependent on the interns' clinic schedules, to comply with standard intra-examiner design accepted in ROM measurement studies.<sup>6</sup>

All the ROM measurements were taken by the same examiner (PT), at the same location, in one of the clinic's treatment rooms on the clinic floor of the college with goniometer measurements always preceeding the modified inclinometer measurements by a few minutes. The interns appeared in random order for each of the three measurements. The measurements were recorded by an independent intern to ensure that the examiner (PT) was blinded to the previous measurement. To ensure the stability of the condition of the ankle of the volunteers, those who had suffered injures in the period between the second and the third measurements were excluded. In this study, none of the participants reported any injuries.



Figure 3. Modified inclinometer measuring plantar flexion relative to a neutral relaxed position called zero.

#### Statistical Analysis

Means and standard deviations were calculated for each measurement of the ROM taken with a goniometer and a modified inclinometer.

Intraclass correlation coefficient (ICC) was used to assess the intra-examiner reliabilty. Based on classification defined by Fleiss and Shrout<sup>14</sup>, we used ICC(2;1) as this measure is commonly used in the literature on test-retest reliability<sup>15,16</sup>. ICC(2;1) accounts for variability between the subjects and between the occasions on which the measurements were taken and can be calculated by fitting a two-way ANOVA model with subjects and occasions as factors. Two ICCs were obtained for each instrument to measure the reliability between Measurement 1 and Measurement 2, and between Measurement 1 and Measurement 3. The calculations were performed using the package *irr*<sup>17</sup> for R software<sup>18</sup>.

#### Results

All the participants (n=19) were included in all three measurements. The mean and standard deviations of ankle ROM for each measurement are included in Table 1. ICC values for the goniometer were 0.86 (same day meas-

Table 1.
Descriptive statistics for the ankle ROM using
goniometer and modified inclinometer.

Test	Goniometer (mean [SD])	Modified inclinometer (mean [SD])
Measurement 1 (n=19)	59.08 [14.34]	69.26 [17.65]
Measurement 2 (n=19)	60.66 [16.45]	67.42 [14.95]
Measurement 3 (n=19)	60.26 [14.07]	70.68 [14.33]

urements) and 0.83 (measurements over different days). For the inclinometer these values were 0.88 and 0.81 respectively (Table 2). These values indicate very good reliability.<sup>19</sup>

#### Discussion

Currently the goniometer method used in this study is still the accepted way of measuring true range of motion of the ankle.<sup>10</sup> Goniometers, although commonly used to measure ankle ROM, are difficult to use when a high volume of measurements need to be taken in a short period of time in field conditions. The method using a digital goniometer, most recently proposed by Thornton *et al.*<sup>9</sup>, seems to be a promising improvement to previous goniometry methods. However, its use required that the participants stand barefoot on the ground/floor, which would not have been prudent in a mobile clinic setting in a developing country where the ground surface would be less than ideal, and inconsistent on each day of measurement.

The intra-examiner reliability of the goniometer and modified inclinometer were obtained from the measurements of ROM taken on participants on the same day, as well as the measurements taken on two different days. In both cases, very good (above 0.80) ICCs were obtained, suggesting the two devices have comparably very good reliability.

Control for toe contribution, convenient patient position and ease of use combined with very good intra-examiner reliability make the modified inclinometer the device of choice for a large population study in which the measurements will be performed by the same examiner. However, it should be emphasized that clinimetric values of this new device have not been established yet and hence its use in clinical practice is not possible. Once an inter-rater reliability and validity of the modified inclin-

Table 2.
<i>ICC</i> (2;1) values and 95% confidence intervals (CI) obtained for goniometer and modified inclinometer.

Goniometer (ICC [ 95% CI])		Modified inclinometer (ICC [95% CI])		
Measurement 1Measurement 1- Measurement 2- Measurement 3		Measurement 1 – Measurement 2	Measurement 1 – Measurement 3	
0.856 (0.665; 0.942) 0.828 (0.607; 0.930)		0.883 (0.722; 0.953)	0.811 (0.574; 0.923)	

ometer is established in future studies, the modified inclinometer can potentially become the device of choice in a regular clinical practice when relative improvements in ankle range of motion is the key required measurement.

It is worth mentioning that the ROM measurements obtained using a goniometer were systematically smaller than the ROM measurements obtained using the modified inclinometer by approximately 10 degrees (Table 1). A similar finding was reported in another study which compared goniometer to inclinometer measurements of ankle dorsiflexion<sup>11</sup>.

There are a few possible explanations for this finding. Likely, it is because the goniometer averaged the straight leg and the bent knee, and calf muscles with a straight leg limit ankle dorsiflexion. Baumbach *et al.*<sup>20</sup> found that knee flexion of 20 degrees is enough to eliminate the effect of the gastrocnemius muscle on dorsiflexion of the ankle.

The systematic discrepancy between the two measurements can also be explained, in part, because the inclinometer measurements were always done after the goniometer ones, and it is likely that repeated measurements increased the ROM values. It would be important to randomize the order of the measurements in future studies to eliminate possible bias due to repeated measurements.

Currently the bubble inclinometer used in this study did not have the ability to establish the vertical gravity -0- position<sup>7</sup> in order to determine the absolute values for dorsiflexion and plantar flexion separately. However this inclinometer allowed the measurement of the total ROM (total of dorsiflexion plus plantarflexion from a set neutral -0- position) which was what would be required for the larger study. Further modification of this inclinometer to include the establishment of a vertical gravity -0- position would be important for future validity studies.

#### Conclusion

This study found a very good intra-examiner reliability for the modified inclinometer. This inclinometer is a sturdy device that helps to control for toe contribution to the ankle ROM, and allows taking measurements in the standing rather than in the supine position as is tradionally done when using a goniometer.

These obvious advantages of the new device and very good intra-examiner reliability make it suitable for use in a large population-based study in the Dominican Republic involving approximately a thousand patients attending mobile chiropractic clinics.

#### References

- 1. Dix FP, Brooke R, McCollum CN. Venous disease is associated with an impaired range of ankle movement. Eur J Vasc Endoasc Surg. 2003;25:556-561.
- Belczak C, Cavalheri G, de Godoy J, Caffaro R, Belczak S. Relação entre a mobilidade da articulação talocrural e a úlcera veosa. J Vasc Bras. 2007;6(2):149-155.
- Fowkes FG, Evans CJ, Lee AJ. Prevalence and risk factors of chronic venous insufficiency. Angiology. 2001; 52 Suppl 1:S5-15.
- 4. Eberhardt R, Raffetto J. Chronic venous insufficiency. Circulation. 2005;111:2398-2409.
- Cocchiarella L, Andersson GB. Guides to the evaluation of permanent impairment.5th. ed. United States of America: American Medical Association, 2001: 535.
- 6. Gajdosik R, Bohannon R. Clinical measurement of range of motion: Review of goniometry emphasizing reliability and validity. Phys Ther. 67(12): 1867-1872.
- Gerhardt J, Cocchiarella L, Lea R. The practical guide to range of motion Assessment. United States of America: American Medical Association, 2002:94-95.
- Belczak C, Cavalheri G, de Godoy J, Caffaro R, Belczak S. Relação entre a mobilidade da articulação talocrural e a úlcera veosa. J Vasc Bras. 2007; 6(2): 149-155.
- Thornton J, Sabah S, Segaren N, Cullen N, Goldberg A. Validated method for measuring functional range of motion in patients with ankle arthritis. Foot Ankle Int. 2016:37(8): 868-873.
- Konor M, Morton S, Eckerson JM, Grindstaff TL. Reliability of three measures of ankle dorsiflexion range of motion. Int J Sports Phys Ther. 2012; 7(3): 279-287.
- Dickson D, Hollman-Gage K, Ojofeitimi S and Bronner S. Comparison of functional ankle motion measures in modern dancers. JDMS. 2012; 16(3): 116.
- Weaver K, Price R, Czerniecki J, Sangeorzan B. Design and validation of an instrument package designed to increase the reliability of ankle range of motion measurements. J Rehabil Res Dev. 2001; 38(5): 471-475.
- Lea R, Gerhardt J. Current concepts review. Range-ofmotion measurements. J Bone Joint Surg Am. 1995; 77A(5): 784-798.
- 14. Shrout P, Fleiss J. Intraclass correlations: uses in assessing rater reliability. Psychol Bull. 1979; 86(2): 420-428.
- 15. Hogg-Johnson, S. Differences in reported psychometric properties of the Neck Disability Index: patient population or choice of methods? Spine J. 2009, 9(10): 854-856.
- Stratford, P.W. On "Test-retest reliability and minimal detectable change on balance ..." Steffen T, Seney M. Phys Ther 2008; 88: 733-46. Phys Ther. 2008; 88: 888-90.
- 17. Matthias Gamer, Jim Lemon, Ian Fellows, and Puspendra

Singh (2012). irr: Various Coefficients of Interrater Reliability and Agreement. R package version 0.84. https:// CRAN.R-project.org/package=irr

- R Core Team (2012). R: A language and environment for statistical computing. R Foundation for Statistical Computing, Vienna, Austria. ISBN 3-900051-07-0, URL http://www.R-project.org/
- Fleiss J.L. Reliability of measurement. The design and analysis of clinical experiments. New York: John Wiley & Sons, 1986: 1-32.
- Baumbach S, Brumann M, Binder J, Mutschler W, Regauer M, Polzer H. The influence of knee position on ankle dorsiflexion- a biometric study. BMC Musculoskelet Disord. 2014; 15: 246.

# Dorsal scapular nerve neuropathy: a narrative review of the literature

Brad Muir, BSc.(Hons), DC, FRCCSS(C)<sup>1</sup>

Objective: The purpose of this paper is to elucidate this little known cause of upper back pain through a narrative review of the literature and to discuss the possible role of the dorsal scapular nerve (DSN) in the etiopathology of other similar diagnoses in this area including cervicogenic dorsalgia (CD), notalgia paresthetica (NP), SICK scapula and a posterolateral arm pain pattern.

Background: Dorsal scapular nerve (DSN) neuropathy has been a rarely thought of differential diagnosis for mid scapular, upper to mid back and costovertebral pain. These are common conditions presenting to chiropractic, physiotherapy, massage therapy and medical offices.

Methods: The methods used to gather articles for this paper included: searching electronic databases; and hand searching relevant references from journal articles and textbook chapters.

Results: One hundred-fourteen articles were retrieved. After removing duplicates, there were 57 articles of which 29 were retrieved. There were 26 articles and textbook chapters retrieved by hand searching equaling Objectif : Ce document a pour objectif d'élucider cette cause peu connue de douleur dans le haut du dos par un examen narratif de la littérature, ainsi que de discuter du rôle possible du nerf scapulaire dorsal (NSD) dans l'étiopathologie d'autres diagnostics semblables dans ce domaine, y compris la dorsalgie cervicogénique (DC), la notalgie paresthésique (NP), l'omoplate SICK et un schéma de douleur postérolatérale au bras.

Contexte : La neuropathie du nerf scapulaire dorsal (NSD) constitue un diagnostic différentiel rare pour la douleur mi-scapulaire, costo-vertébrale et au bas/haut du dos. Il s'agit de troubles communs qui surgissent dans les cabinets de chiropratique, de physiothérapie, de massothérapie et de médecin.

Méthodologie : Les méthodes utilisées pour rassembler les articles de ce document comprenaient la recherche dans des bases de données électroniques et la recherche manuelle de références pertinentes dans des articles de journaux et des chapitres de traités.

Résultats : On a extrait 114 articles. Une fois les dédoublements éliminés, il y avait 57 articles, desquels 29 ont été extraits. Il y avait 26 articles et chapitres de traités extraits à la main, ce qui donne 55 articles

Corresponding author: Brad Muir Canadian Memorial Chiropractic College, 6100 Leslie Street, Toronto, ON M2H 3J1 e-mail: bmuir@cmcc.ca Tel: 416-482-2340

The author does not have any conflicts of interest to declare with regard to the writing of this manuscript. @ JCCA 2017

<sup>&</sup>lt;sup>1</sup> Canadian Memorial Chiropractic College

discogenie pani, costo vertec

J Can Chiropr Assoc 2017; 61(2)

55 articles retrieved of which 47 relevant articles were used in this report.

Discussion: The anatomy, pathway and function of the dorsal scapular nerve can be varied and exceptionally rarely may include a sensory component. The signs and symptoms, therefore, may include pain, atrophy, scapular winging, and dysesthesia. The mechanism of injury to the DSN is also quite varied ranging from postural to overuse in overhead work and sport. Other conditions in this area, including CD, NP, SICK scapula and a posterolateral arm pain pattern bear a striking resemblance to DSN neuropathy.

Conclusion: DSN neuropathy should be included in the list of common differential diagnoses of upper and mid-thoracic pain, stiffness, dysesthesia and dysfunction. The study also brings forward interesting connections between DSN neuropathy, CD, NP, SICK scapula and a posterolateral arm pain pattern.

#### (JCCA. 2017;61(2):128-144)

KEY WORDS: dorsal scapular nerve, nervus dorsalis scapulae, mid-thoracic pain, cervicogenic dorsalgia, notalgia paresthetica, SICK scapula, posterolateral arm pain pattern

#### Introduction

Dorsal scapular nerve (DSN) (Latin: nervus dorsalis scapulae) neuropathy has been a rarely thought of differential diagnosis for mid scapular, upper to mid back pain.<sup>1</sup> Upper to mid-thoracic and costovertebral pain and stiffness are a common entity presenting to chiropractic, physiotherapy, massage therapy and medical offices<sup>2-4</sup> especially following motor vehicle accidents<sup>5</sup>. Patients may report pain, stiffness, dysesthesia and dysfunction (scapular, thoracic and costovertebral) of an acute or chronic nature in this area with only temporary relief following normal care by their health professional.<sup>1</sup>

In a study by Sultan *et al.*<sup>1</sup>, 55 patients with unilateral interscapular pain were evaluated. The diagnosis for these patients varied from no diagnosis to thoracic degenerative discogenic pain, costovertebral joint dysfunction, levator

*extraits, desquels 47 articles pertinents ont été utilisés pour ce rapport.* 

Discussion : L'anatomie, la voie et la fonction du nerf scapulaire dorsal peuvent être variées et, exceptionnellement, comprendre un facteur sensoriel. Par conséquent, les signes et symptômes peuvent comprendre la douleur, l'atrophie, le décollement scapulaire et la dysesthésie. Le mécanisme de blessure du NSD est lui aussi très varié, allant de la posture au travail/sport au-dessus de la tête. D'autres troubles dans ce domaine, dont la DC, la NP, l'omoplate SICK et un schéma de douleur postéro-latérale au bras, ressemblent étrangement à la neuropathie du NSD.

Conclusion : Il faut inclure la neuropathie du NSD dans la liste des diagnostics différentiels communs pour la douleur thoracique supérieure et médiane, la raideur, la dysesthésie et le dysfonctionnement. De plus, l'étude met en évidence d'intéressants liens entre la neuropathie du NSD, la DC, la NP, l'omoplate SICK et un schéma de douleur postéro-latérale au bras.

(JCCA. 2017;61(2):128-144)

MOTS CLÉS : chiropratique, nerf scapulaire dorsal, nervus dorsalis scapulae, douleur thoracique médiane, dorsalgie cervicogénique, notalgie paresthésique, omoplate SICK, schéma de douleur postéro-latérale au bras

scapulae syndrome, thoracic facet syndrome, dorsal back strain, myofascial pain of the rhomboids and finally DSN entrapment.<sup>1</sup> These are common diagnoses rendered for pain in this area and the referral for an electrophysiologic study would suggest a lack of progress with normal care. The study found that there was evidence of DSN neuropathy in 29 patients (52.7%) and another five were at the upper cutoff limit which would bring the total to 61.8% if these were counted as having DSN involvement.<sup>1</sup> This would suggest that DSN neuropathy should be included in the differential diagnoses for upper to midback pain, stiffness and dysfunction. Its consideration may in fact add to, or change, the diagnosis and provide an avenue for longer lasting relief with an appropriate treatment strategy.

The signs and symptoms of DSN neuropathy bear a

129

striking resemblance to several other diagnoses or findings in the cervicothoracic, scapular and posterolateral arm areas including cervicogenic dorsalgia (CD), notalgia paresthetica (NP), SICK scapula and a posterolateral arm pain pattern. First described by Maigne<sup>6,7</sup>, cervicogenic dorsalgia (CD) is not well recognized outside of chiropractic literature (a literature search in PubMed revealed no articles). Anatomically, the DSN provides that direct link from the mid to lower cervical spine to the mid-scapular region.<sup>8-10</sup> Notalgia paresthetica (NP) is a condition of the upper to mid-thoracic spine that involves pruritis, numbness and tingling, and pain.<sup>11-15</sup> The etiology of this condition is elusive, and its treatment using conservative therapies has also been met with mixed results.<sup>11,13,16</sup> In the paper by Sultan et al.<sup>1</sup>, two of the 29 patients that tested positive for DSN neuropathy had pruritis in the upper to mid-back area with one of those having a decreased ability to sense a pinprick over the area. This, along with a dissection report by AF Dixon in 1896 that showed cutaneous nerve fibres from the DSN innervating an area of the mid-thoracic spine<sup>17</sup>, suggests a possible connection between NP and DSN neuropathy. "SICK scapula", an acronym for the condition Scapular malposition, Inferior medial border prominence, Coracoid pain and malposition, and dysKinesis of scapular movement<sup>18</sup>, is a condition that has been reported to have rhomboid related scapular winging which suggests DSN involvement. SICK scapula is speculated to be a component of several shoulder related diagnoses including impingement, labral tears and rotator cuff tendinopathies and tears.<sup>18</sup> DSN neuropathy may cause scapular winging due to rhomboid atrophy19-20 and may need to be considered as a component of SICK scapula. Several authors mention that DSN neuropathy may be related to a posterolateral arm and forearm pain pattern<sup>21-23</sup> that may include the neck, axilla and lateral thoracic wall<sup>21</sup>. This paper will explore the possibility that the pain pattern may be due to a peripheral nerve neuropathy that includes the DSN, suprascapular nerve, long thoracic and radial nerve with the C5 nerve root being a common thread. A possible mechanism that may explain the etiopathology behind these nerves being affected is explored. The mechanism behind these nerves combining to create this pain pattern while other C5 nerves including the axillary, median, or musculocutaneous nerve, are rarely affected, is unknown and not in the scope of this paper.

Table 1.Sources used in the search for articlesfor this manuscript.

- PubMed Key Words: dorsal scapular nerve, dorsoscapular nerve, nervus dorsalis scapulae, mid-thoracic pain
- EbscoHost that included: Alt HealthWatch, AMED, CINAHL, eBook Collection, ERIC, Medline, Nursing & Allied Health Collection, Psychology and Behavioral Sciences Collection, Rehabilitation & Sports Medicine Source, SportDiscus, American Doctoral Dissertations Key Words: dorsal scapular nerve, dorsoscapular nerve, nervus dorsalis scapulae, mid-thoracic pain
- Index to Chiropractic Literature Key Words: dorsal scapular nerve, dorsoscapular nerve, nervus dorsalis scapulae, mid-thoracic pain
- Hand searches of references from the retrieved articles and textbooks

The purpose of this paper is to elucidate this little known, but emerging, cause of upper back pain. This paper will include a narrative review of the anatomy and function of the DSN, along with the epidemiology, signs and symptoms and possible mechanisms of injury of DSN neuropathy. It will also explore the possible role of DSN neuropathy in other diagnoses in this area including CD, NP, SICK scapula and a posterolateral arm pain pattern.

#### Methods

The methods used to gather articles for this paper included: searching several electronic databases and hand searching relevant references from journal articles and textbook chapters. Table 1 outlines the electronic databases used in the search. The following terms were used in the searches: dorsal scapular nerve, dorsoscapular nerve, nervus dorsalis scapulae, and mid-thoracic pain. It should be noted that "dorsoscapular nerve" is not recognized as a proper anatomic/medical term for the DSN but was found to be used while procuring articles during the literature search.

Due to a relative lack of articles found in the electronic database search, the bulk of the articles and textbook chapters were found by hand searching the references of each article and attempting to purchase or obtain these articles through available college and university libraries. Google Scholar was used to retrieve hand searched arti-

Table 2.Flowchart outlining the search strategy and article aquisition



cles that were not otherwise retrievable and briefly examined to see if all relevant articles had been identified. Due to the lack of limiters, Google Scholar was not used as a main search engine source.

#### Results

Searching the electronic databases, 114 articles were found. Once duplicates were removed there were 57 articles of which twenty-nine were retrieved. There were 26 articles and textbook chapters retrieved by hand searching the articles that had been retrieved equaling 55 articles of which 47 relevant articles were used in this report.

Of the 57 articles from the database search, only 29 articles were retrieved from this list. The articles not retrieved were eliminated due to language issues, and a lack of relevance determined from the abstract, and due to an inability to procure the article. All of the articles retrieved by hand searching were used in this report. The remaining eight articles that were retrieved and not used was due to a lack of relevance to the subjects covered in the narrative review after reading the full article.

#### Discussion

#### Origin

The DSN is typically reported to arise from the anterior ramus of the C5 nerve root directly or as the first branch of the superior trunk of the brachial plexus.8 In a cadaveric study, Ballesteros and Ramirez<sup>8</sup> found that this usual presentation was only in 17.9% of cadavers. In another 30.4%, it still arose from the usual position but shared a common branching trunk with the long thoracic nerve. The other reported variations included C4 in 28.4% and C4 and C5 in 23.1%. In a cadaveric case report by Shilal et al.9, the DSN was found to have input from the C6 nerve root. Interestingly, the long thoracic nerve on this individual arose from the C6 and C7 nerve roots only with an aberrant communicating branch between the dorsal scapular and the long thoracic nerves.

In another cadaveric study by Tubbs et al.<sup>10</sup>, the DSN originated from C5 in 19 of the 20 sides (10 cadavers) with one originating from the C5 and C6 spinal nerves. In a cadaveric study done by Lee et al.<sup>24</sup>, the DSN originated from the fifth cervical anterior ramus in 75.8% of the cases. In 9.0%, it arose from the superior trunk of



Figure 1. Pathway of the dorsal scapular nerve

the plexus while in 7.6% it was from the fourth and fifth cervical anterior rami and in another 7.6% from the sixth cervical anterior rami. In the anatomic study by Chen et al.<sup>21</sup>, one patient had bilateral input to the DSN from C3-4.

In a cadaveric study by Nguyen et al.25, they reported a C5 origin in 70% (16 of 23 cadavers), 22% from C4 and 8% from C6. No combination of nerve roots was reported in this study. In a study by Malessy et al.26 on four cadavers (one side only, two male and two female), the DSN arose from C4 and C5 in all specimens. It seems that the DSN has been erroneously reported as arising from C4 to T1<sup>10,27,28</sup>, and from C8 only<sup>22</sup>.

#### Pathway

The course of the DSN as described by Chen *et al.*<sup>21</sup>, has it immediately pass obliquely and inferiorly through the middle scalene without innervating it. Classically, the middle scalene is described as acting to flex the cervical



Figure 2. *Dissection showing the dorsal scapular nerve*.

The dorsal scapular nerve is shown innervating the levator scapulae, rhomboid minor and rhomboid major muscles. The rhomboid major and minor are reflected from the midline showing the DSN attached to the anterior aspect of each. Also shown is the C3 and C4 nerves innervating the upper portion of the levator scapulae

spine when contracting bilaterally, ipsilaterally laterally flex and contralaterally rotate the cervical spine when acting unilaterally, and to elevate the first and possibly the second rib with forced inspiration.<sup>29-31</sup> Olinger and Homier<sup>29</sup> and Buford *et al.*<sup>32</sup> suggest that the scalenes have the ability to ipsilaterally rotate the cervical spine and can be stretched with both ipsi and contralateral rotation. The middle scalene is normally innervated by branches from the anterior rami of the third through eighth cervical spinal nerves<sup>33</sup>. The middle scalene is often described as an entrapment site of the DSN<sup>21</sup> and thus its function may play a part in determining the role of the cervical spine in the tensioning of the DSN.

The DSN then runs inferiorly and slightly laterally between the superior fibres of the upper trapezius medially and the levator scapulae laterally. It then passes deep to the upper trapezius fibres as they curve/course laterally toward the acromion, lying anterior to the rhomboid major and minor but posterior to the serratus posterior superior muscle (SPS). The main trunk of the DSN lies medial to the medial border of the scapula as it travels inferiorly to the inferior medial border of the scapula that typically ends at the level of the T7 spinous process<sup>31</sup> (see Figure 1 and 2).

A case report by George and Nayak<sup>34</sup> described a variation of the pathway. They found that the DSN came from the anterior ramus of C5 and made a loop around the deep branch of the cervical artery suggesting a possible mechanism of compression. It then continued along its normal path as described above. In the Tubbs *et al.*<sup>10</sup> study, they reported that in all twenty sides, the DSN was intertwined with the dorsal scapular artery and was found along the anterior border of the rhomboid major and minor muscles. In the study by Lee *et al.*<sup>24</sup>, the DSN pierced both the middle and posterior scalene in 6.4% of the cases. Martin and Fish<sup>35</sup> report the DSN piercing the levator scapulae as it travelled inferiorly in nine of their 35 dissected specimens suggesting a possible area of compression. In the cadaveric study by Nguyen *et al.*<sup>25</sup>, the DSN pierced the middle scalene in 74% of their cases, while passing anterior and posterior to the middle scalene in 26% (13% each).

#### Function

The DSN's main function is the innervation of the rhomboid major and minor which retract, elevate and stabilize the scapula. It also acts to rotate the lateral border of the scapula downward.<sup>35</sup>

The DSN will also innervate the levator scapulae (LS) muscle. In a cadaveric study, eleven of the thirty-five specimens showed DSN innervation of the LS.<sup>36</sup> The other source of innervation to the LS is C3 and C4 via the cervical plexus.<sup>36</sup> The LS mainly acts to elevate the scapula, rotate the glenoid cavity inferiorly by rotating the scapula and will retract the scapula.<sup>31</sup> It also acts as an accessory breathing muscle during dysfunctional breathing.<sup>37</sup>

The DSN was reported to innervate the LS only in 48% of the cases described by Nguyen *et al.*<sup>25</sup> In the other 52% of the cases, the DSN innervated the LS and the rhomboid minor and major muscles combined. This is the first study found that describes the DSN as only innervating the LS and no mention was made of what was providing innervation to the rhomboids in these cases.

In the cadaveric study by Malessy *et al.*<sup>26</sup>, they report that the cranial aspect of LS was innervated by a branch from C3, with the caudal aspect innervated by C4 that combined with a C5 branch to supply the rhomboids in all four specimens.

Kida and Tani<sup>38</sup>, in a cadaveric case report showed the DSN sending a branch to innervate the SPS muscle. Vilensky *et al.*<sup>39</sup> describe the medial origin of the SPS as the spinous process of the sixth cervical to the second thoracic. It traverses obliquely, laterally and inferiorly to insert on the upper border of ribs two to five just lateral to their angle. Its normal innervation is the intercostal nerves ranging from T1 to T4 or T5.<sup>39</sup> According to Vilensky *et al.*<sup>39</sup>, most textbooks describe the SPS as having a respiratory function due to its location on the ribs. They suggest that it may not in fact be active during respiration but may have a proprioceptive function for the thoracic spine possibly during respiration.

Normally, the dorsal scapular nerve contains only motor fibres, but an old dissection case report<sup>17</sup> describes sensory fibres from the nerve that innervated a small area of skin at the level of the fifth and sixth thoracic vertebrae. This finding may suggest a connection from the cervical spine to notalgia parasthetica, an elusive diagnosis of pruritis and/or pain and dysesthesia in an area of the mid scapular region.

#### Epidemiology

Musculoskeletal thoracic pain is common in the general population. The annual prevalence of thoracic spine pain in adults ranges from 15.0% in Swedish adults (aged 35 to 45 years) to 34.8% in Swedish working adults (aged 16 to 65 years).<sup>2-4</sup> The lifetime prevalence for thoracic spine pain in adolescents was 15.6 to 19.5%.<sup>3</sup> In a study by Briggs et al.3, it was reported that factors associated with thoracic spine pain in children and adolescents includes female gender, postural changes associated with backpack use, backpack weight, participation in specific sports, chair height at school, and difficulty with homework. In adults, thoracic spine pain was associated with concurrent musculoskeletal symptoms and difficulty performing activities of daily living. Thoracic pain is also commonly associated (65.5%) with neck pain in individuals injured in motor vehicle accidents.5

Pain and dysfunction arising from compression of the DSN has often been reported as rare<sup>23,40</sup>, or not included as a cause of neurologic related shoulder injuries<sup>41</sup>, or as a differential diagnosis in posterior upper thoracic pain<sup>42</sup>. However, a report by Sultan et al.<sup>1</sup> suggests that this may not be the case. In their study, 55 patients with unilateral interscapular pain were evaluated. The diagnosis for these patients varied from no diagnosis to thoracic degenerative discogenic pain, costovertebral joint dysfunction, levator scapulae syndrome, thoracic facet syndrome, dorsal back strain, myofascial pain of the rhomboids and finally DSN entrapment. They found that there was evidence of DSN neuropathy in 29 patients (52.7%) and another five were at the upper cutoff limit which would bring the total to 61.8% if these were counted as having DSN involvement.1

In the study by Chen *et al.*<sup>21</sup>, they examined and treated 36 patients with dorsal scapular nerve compression. There were 28 female and eight male patients whose ages ranged from 29 to 52 years with an average age of 34 years. Of the 36 patients, symptoms were unilateral in 34, two were bilateral, 20 were on the right side and 16 on the left. Sultan *et al.*<sup>1</sup> included 42 women and 13 men, and an age range of 22 to 52 years (average 40.2). There were 29 patients that had symptoms on the right side and 26 on the left. Only 29 of these patients were considered testing positive for DSN neuropathy on electrophysiologic evaluation but these were not broken down into gender.

#### Signs and Symptoms

DSN neuropathy may present on a spectrum from complete function to complete atrophy of the muscles it innervates. This spectrum would include varying levels of: pain intensity and character along a portion of or its entire pathway; and tightness and weakness in the muscles it innervates.<sup>1</sup> Other symptoms may include dysesthesia and pruritis in the midscapular region<sup>1</sup>, and radiation of the pain along the posterolateral aspect of the shoulder, arm, and forearm<sup>21-23,43</sup>. Chen *et al.*<sup>21</sup> also report DSN involvement in neck, axilla, and lateral thoracic wall pain.

Other findings may include a loss of pinprick sensation medial to the scapular border<sup>1</sup>, and varying levels of loss of range of motion of the cervical spine, typically ipsilateral rotation and contralateral lateral flexion<sup>22,40</sup>. A loss of range of motion of the affected side shoulder has also been outlined although no specific movements were described.<sup>1,40</sup> Cervical flexion, ipsilateral lateral flexion<sup>1</sup>, and extension<sup>19</sup> have also been reported to aggravate the pain along the DSN. Pain on palpation of the thoracic spinous<sup>21</sup>, thoracic facet and costotransverse joints may also be present<sup>1</sup>. Relative hypertrophy and spasm of the neck musculature has also been reported.<sup>40</sup> An elongated C7 transverse process has been reported in association with this condition by a few authors.<sup>1,21,40</sup>

Weakness of the rhomboids may cause varying levels of winging of the scapula. Ravindran<sup>20</sup> describes two cases of suprascapular neuropathy in a brother and sister that played volleyball at a high level. Each had electromyographical confirmation of chronic neurogenic changes in the supraspinati, infraspinati and rhomboid muscles with normal findings in the trapezius, deltoid and serratus anterior. The neurogenic changes mentioned presented as muscle weakness and wasting of the infraspinatus, as well as weakness in the supraspinatus and rhomboids. Both had mild winging of the scapula that didn't change with shoulder ranges. It was postulated that they either had a concurrent DSN neuropathy or an anatomical variation of the suprascapular nerve innervating the rhomboids – which has not been reported in cadaveric studies.<sup>20</sup> Moderate to more severe cases of scapular winging due to DSN injury are reported by several authors.<sup>19,28,43,47</sup>

Benedetti *et al.*<sup>43</sup> describe the case of a 24 year old woman that presented four years after a crash in which she sustained a pelvic fracture as well as abdominal and chest trauma. For the past year, she had been experiencing upper back pain with constant left shoulder pain with radiation and weakness of the left upper limb. An MRI showed no sign of a C5 radiculopathy. She presented with left scapular winging along with severe global loss of range, tingling and numbness in the left upper limb and tremor with the initiation of active movment. There was no mention of range of motion findings of the cervical spine. An EMG study was performed confirming signs of denervation of the left rhomboid suggestive of a DSN lesion.

Plezbert and Nicholson<sup>47</sup> report on a case of a 28 year old with persistent cervical and thoracic pain along the medial border of the scapula and left shoulder weakness. The patient had been originally diagnosed with cervical-thoracic myofascitis but was re-examined due to little progress. The re-exam noted weakness in the rhomboid (3/5) and posterior deltoid (4/5) with mild inferior scapular angle winging. Reflexes and sensory testing was found to be within normal limits. Restrictions on palpation were noted in the cervical, upper thoracic and left upper costovertebral joints. Deep palpation of the anterior and middle scalenes, as well as, left cervical rotation and cervical extension recreated the pain along the medial border of the scapula. The patient's diagnosis was subsequently revised to having a left DSN entrapment neuropathy secondary to scalene myofascitis. Radiographs of this patient showed a flattened thoracic kyphosis and a congenital block vertebrae at C2-3.

#### Etiology

The main etiology described for DSN entrapment is hypertrophy of the middle scalene muscle causing compression of the nerve as it passes through.<sup>21</sup> Mondelli *et al.*<sup>48</sup> describes the case of DSN neuropathy in a bodybuilder whom they felt had tractioned the nerve in a hypertrophied middle scalenus muscle during exercises of neck flexion and heavy shoulder raises and lowers.

There are several other case reports in the literature that outline a variety of different mechanisms. In a case

of repetitive lifting, Argyriou et al.44 describe rhomboid atrophy and scapular winging secondary to DSN neuropathy in a worker required to manually handle and carry bags weighing eight kilograms across his body. Haim and Urban<sup>49</sup> describe the case of a rhomboid dystonia following thoracic disc surgery that required DSN blocks to resolve. Debeer et al.<sup>28</sup> report on a case involving a 15 year old girl. The girl was being treated for three years for idiopathic scoliosis utilizing a corrective brace that the authors felt caused a compressive neuropraxia to the DSN and subsequent, severe shoulder dysfunction secondary to scapular winging. Ravindran<sup>20</sup>, as mentioned, describes siblings with suspected DSN neuropathy due to repetitive overhead activity related to volleyball. Kugler et al.<sup>50</sup> mention an estimate by Dubotzky and Leistner that a highly skilled volleyball attacker practicing 16 to 20 hours per week will spike the ball approximately 40 000 times per year. Kaplan et al.45 describe a case of an 18 year old female with a one year history of right scapular pain and mild scapular winging. Denervation of her right rhomboid major and levator scapulae were found and attributed to 30 months of continuous studying (four to five hours per day) for a weekly three hour exam. This case suggests that continuous stretch of the DSN secondary to poor posture may be enough to cause a chronic neuropathy. Akgun et al.<sup>19</sup> describes a more acute case of DSN injury in a 51 year old man that felt a sharp pain in the right shoulder after lifting a heavy box overhead. In another case report, Jerosch et al.51 describes the case of a 19 year old female that anteriorly dislocated her right shoulder following a fall in judo. Two surgeries to stabilize the shoulder were unsuccessful and upon presentation to a new clinic, winged scapula with atrophy of the rhomboids and serratus anterior were noted. EMG confirmed damage to both the long thoracic and DSN.

In the study by Sultan *et al.*<sup>1</sup>, two of the 29 patients with DSN neuropathy developed the condition acutely after lifting heavy objects overhead in a manner similar to Akgun *et al.*<sup>19</sup>. The remainder described a gradual, insidious onset of the condition. In nine of these patients, they related the onset of their symptoms to frequent, repetitive use of their dominant side in overhead activities related to work, recreation and sport. Of these nine patients, three were teachers, two were painters, two electricians, one was a volleyball player and one a basketball player<sup>1</sup>.

Brower<sup>22</sup> also suggests that a traction injury to the

nerve secondary to a whiplash event is a possible etiology although no specific cases were discussed. Benedetti *et* al.<sup>43</sup>, describe a DSN lesion following a crash although there was no mention of it being a car or other vehicle. They felt her DSN injury was likely due to the chest trauma sustained in the accident. No mechanism of injury was mentioned in the case reported by Plezbert and Nicholson.<sup>47</sup>

In summary, the DSN may be injured due to overhead activity during work and sport/recreation that may be repetitive and in some cases involve heavy loads, chronic postural strain, iatrogenic (either post-surgical or post bracing), or following a crash involving a motor vehicle of some kind.

#### Possible role of DSN neuropathy in other conditions

#### Cervicogenic dorsalgia

Maigne suggests that 70% of common dorsal pain originates from the lower cervical spine.<sup>6,52</sup> This compares to the estimate by Sultan *et al.*<sup>1</sup> who found that 53% to 62% of patients in their study with unilateral interscapular pain had DSN neuropathy. Maigne<sup>6</sup> and Terrett<sup>52</sup> both report that women are more affected than men with Terrett<sup>52</sup> estimating it at a 6:1 ratio. This ratio is slightly higher than the DSN gender ratio of 3.5:1 in Chen *et al.*<sup>21</sup> No mention of side to side differences was mentioned in Maigne<sup>6</sup>, Terrett<sup>52</sup> or Engel and Gatterman<sup>7</sup> or how often the condition may be bilateral.

Engel and Gatterman<sup>7</sup> suggest that the cause of the cervical spine irritation that's creating the thoracic spine pain may lie in a variety of structures including the disc and the facet and the diagnosis should include these as the primary diagnosis with the thoracic pain mentioned as an associated symptom. The difficulty in diagnosing this condition is the patient may consistently deny any associated neck pain especially in the subacute and chronic stages<sup>7,52</sup> which is similar to DSN neuropathy<sup>1</sup>.

Maigne<sup>6</sup> and Terrett<sup>52</sup> describes the interscapular pain of CD as well localized or diffuse, a cramping sensation, the sensation of a weight, a burning or painful tension, a feeling of fatigue and/or a deep-seated intrathoracic pain. They also suggest that cervical ROM may range from normal, to minimally to markedly decreased in certain directions. Again, similar to DSN neuropathy, the interscapular pain in the cases described by Terrett<sup>52</sup> were recreated by ipsilateral cervical spine rotation and further increased by cervical extension from the rotated position. The pain usually decreases with rest, however, it may be aggravated during sleeping especially if the patient's preferred position is prone with rotation to the affected side.<sup>6,7,52</sup> There is no mention of radiation of the pain across the scapula and along the posterolateral arm with CD unlike in DSN neuropathy.<sup>21-23</sup>

Maigne<sup>6</sup> also describes a "cervical point of the back" found during the physical exam for CD. Maigne<sup>6</sup> suggests that this point is a consistent point of pain just lateral to the spinous processes (2 cm) which is found with palpation between the T5 and T6 spinous in a large percentage of patients. This is very similar to the pain at the thoracic spinous described by Chen *et al.*<sup>21</sup> in patients with DSN neuropathy. Maigne<sup>6</sup> also describes a mid-thoracic "cellulalgic band" found by skinrolling. This feature has not been described in the literature pertaining to DSN irritation although it has been mentioned with regard to notal-gia paresthetica.<sup>11</sup>

Radiographic findings of the cervical and thoracic spine are often negative similar to those taken in patients with DSN neuropathy. Similar to a DSN neuropathy, CD is often misdiagnosed as a subluxated rib or a trigger point<sup>7,52</sup>, thoracic posterior facet syndrome, thoracic subluxation, T4 syndrome, discogenic disease, costovertebral lesion, intercostal muscle spasm, interscapular or scapular muscle spasm<sup>1</sup>.

The "anterior doorbell sign" described by Maigne<sup>6</sup> consists of recreating the intrathoracic pain by applying pressure to the "responsible level and at the anterolateral part of the spine". Maigne<sup>2</sup> suggests it is an inconstant finding and is positive in six of ten cases of thoracic pain of cervical origin. He also suggests that to find the "cervical doorbell point" you may have to vary the palpation point by a few centimetres<sup>6</sup>. Engel and Gatterman<sup>7</sup> suggest that the "cervical doorbell point" nerve root and is often mistaken for a "scalene trigger point". The location of the structure described by Maigne<sup>6</sup>, and Engel and Gatterman<sup>7</sup> may be the location of the DSN as it pierces the middle scalene as it branches from the anterior rami<sup>21</sup>.

Treatment of CD is focused on the cervical spine<sup>6,7,52</sup> with the predominant therapeutic modality being manipulation and less often exercise and massage or soft tissue therapy to the thoracic and cervical spine musculature. Manipulation of the thoracic facet and costovertebral joints is often met with temporary relief only and should tip the practitioner to a different primary cause of the intrathoracic pain.<sup>52</sup> Plezbert and Nicholson<sup>47</sup> describe the successful treatment of a case of DSN neuropathy secondary to scalene myofascitis. The patient was treated with trigger point therapy of the scalene musculature, high-volt galvanized current to the rhomboid muscle utilizing muscle strengthening settings, as well as chiropractic manipulation to the cervical, thoracic and upper posterior ribs.

There is no mention of scapular winging in any of the cases or papers by Engel and Gatterman<sup>7</sup>, Maigne<sup>6</sup>, or Terrett<sup>52</sup> and none of them mention the possibility of the thoracic pain being the result of a DSN neuropathy.

Considering the few differences, the similarities in presentation of DSN neuropathy and CD suggest that further investigations should be carried out to see if they are in fact one and the same.

#### Notalgia paresthetica

Notalgia paresthetica (NP) is a condition characterized mainly by unilateral pruritis in an area medial to the scapula and lateral to the thoracic spine.<sup>11-15</sup> It is often accompanied by pain, numbness or tingling, paresthesia, or hyperesthesia and is commonly found to have local hyperpigmentation thought to be the result of chronic scratching.<sup>13</sup> One author suggests that it may be that "unreachable itch" that led not only humans but primates (apes in particular) to develop back scratchers to deal with it.<sup>13</sup> Ellis<sup>13</sup> also suggests that "pruritis is an often unrecognized symptom of nerve damage". This condition was first described by Astwazaturow in 1934 but remains difficult to treat even today.<sup>14</sup>

The condition is thought to be neurogenic in origin due to the relative, short term effectiveness of drugs used to aid in the treatment of nerve pain including topical capsaicin, botox, nerve blocks and gabapentin.<sup>13</sup> Most often NP has been attributed to the entrapment of the T2-T6 posterior rami that supply cutaneous innervation to the area but its true etiology remains elusive.<sup>16</sup> Some authors suggest that the entrapment may be due to spasms of the paraspinal musculature, mainly the multifidus, the scapular stabilizers including the rhomboid and trapezius, or impingement due to degenerative changes in the thoracic spine and/or thoracic herniated discs.<sup>53</sup> Other treatments aimed at the proposed neuromusculoskeletal pathologies have also had some success at treating NP including exercise, acupuncture, and osteopathic treatment including muscle energy, soft tissue, inhibition and fascial release of the area<sup>11</sup> as well as ultrasound and radiation physiotherapy<sup>54</sup>. Another case series of patients with long thoracic nerve injury or of the C5-7 cervical roots found that muscle stimulation (EMS) of the serratus anterior was helpful in the treatment of NP.<sup>12</sup>

The findings of AF Dixon<sup>17</sup> in 1896 that the DSN had cutaneous branches that innervated the skin in the mid-scapular area suggests a possible direct link of NP and DSN neuropathy. Sultan *et al.*<sup>1</sup> in fact describes two patients with DSN entrapment that have a concurrent itching sensation along with pain and one of the two patients had reduced pinprick sensation in the area just medial to the scapular border. Along with the pruritis, pain along the medial scapular border seems to be a feature of NP<sup>13</sup> which has already been discussed previously with respect to DSN neuropathy<sup>1</sup>.

Notalgia paresthetica, like DSN neuropathy, is predominantly a chronic condition found in middle to older aged women at a 2:1 or 3:1 ratio versus men.<sup>54</sup> In a review by Perez-Perez<sup>15</sup>, the author's personal observation suggests NP may be as high as 9:1. In a study by Wallengren<sup>55</sup>, their patient distribution was 4:1 women to men, age range 35 to 70 years with a mean age of 59 years, with a unilateral distribution in 19 of the 20 (left side 13, right side six) with one patient having the condition bilaterally.

The DSN has a known anatomical and functional link to the long thoracic nerve<sup>8,9,21</sup> so EMS of the serratus anterior may be beneficial to DSN neuropathy although this has not previously been shown in the literature<sup>12</sup>. The aforementioned cases of NP did not note any assessment of the cervical spine or any exacerbation of symptoms with neck movement, although neck related injury or surgery to the C5-7 area were described in two of the four cases by Wang et al.<sup>12</sup> Also, an NP case of two years duration was reported by Alai et al.56 to have a decreased range of motion in the neck (specific directions were not reported) associated with noticeable bilateral cervical muscle spasm greater on the left. The patient did have a history of multiple motor vehicle accidents resulting in whiplash associated injuries to the neck and infrascapular back area 15 to 20 years prior. The patient's cervical spine MRI revealed degenerative changes and mild disc protrusions of C4 through C7 which were felt to be contributing factors to the NP.56 Richardson<sup>11</sup> also reports a case of NP with associated neck, upper back and low back pain of approximately two years duration with symptoms that began following a rear-end motor vehicle accident. Their patient also demonstrated decreased cervical range of motion as well as tissue texture changes in vertebrae T2 through T7 with tenderness of the corresponding spinous processes and "an appreciable ropy and fibrotic texture at the left scapula"<sup>11</sup>. The tissue texture changes sound similar to those described by Maigne<sup>6</sup> with regard to changes found in CD. In two cases of exercises being used to treat NP, rhomboid weakness was noted causing protracted scapula.<sup>57</sup> It was postulated that the NP was caused by a constant stretch of the T2 to T6 spinal nerves. This mechanism, however, may also cause a constant tension on the DSN resulting in DSN neuropathy and increasing rhomboid weakness if prolonged.<sup>1</sup>

One area where the comparison between DSN neuropathy and NP requires further study is a small portion of the affected area itself. The T6 dermatome falls below the level of the inferior border of the scapula<sup>58</sup> and thus below the level of the rhomboid attachment on the inferior medial border of the scapula. This would suggest that the DSN neuropathy alone would not affect this area although a possible combination of DSN neuropathy and thoracic cutaneous nerve neuropathy, as outlined later in the discussion, may be the potential cause.

#### SICK scapula

SICK scapula is an acronym that stands for Scapular malposition, Inferior medial border prominence, Coracoid pain and malposition, and dysKinesis of scapular movement.<sup>18</sup> SICK scapula is a complex of scapular issues affecting overhead athletes. Certain components of SICK scapula bear a striking resemblance to the findings of DSN neuropathy. Burkhart, Morgan and Kibler<sup>18</sup> report that patients with an isolated SICK scapula "may complain of anterior shoulder pain, posterosuperior scapular pain, superior shoulder pain, proximal lateral arm pain or any combination of the above. In addition, posterosuperior scapular pain may radiate into the ipsilateral paraspinous cervical region or the patient may complain of radicular/thoracic outlet type symptoms into the affected arm, forearm, and hand ...". In particular, the type II pattern of dynamic scapular dyskinesis has entire medial border winging of the affected scapula at rest that becomes more prominent with the cocking and elevation phase of pitching. They suggest that it is associated with upper and lower trapezius and rhomboid muscle weakness.<sup>18</sup> They also suggest that continued SICK scapula can lead to rotator cuff impingement, SLAP lesions and possible "dead arm".<sup>18</sup>

DSN neuropathy should be considered in these patients especially with scapular winging, interscapular pain, and radiation along the posterolateral arm and forearm.<sup>1</sup> DSN neuropathy has been shown to be associated with overuse injuries sustained in overhead athletes.<sup>1,20</sup>

#### Posterolateral Arm Pain – C5 Peripheral Nerve Neuropathy

Many authors report a posterior shoulder/posterolateral arm pain pattern associated with DSN neuropathy, scapular dysfunction and shoulder injuries.<sup>1,18,20-23,43,59</sup> This may be accompanied by occasional pain to the forearm and posterior hand<sup>22,23</sup> and the axilla and lateral thoracic wall<sup>21</sup>.

There is debate in the literature surrounding the existence of active and latent trigger points and their ability to refer pain versus the pain being of peripheral nerve origin.<sup>60,61</sup> If you briefly examine the pain pattern from both sides, there are some interesting similarities and omissions when it comes to this particular pain pattern. From a peripheral nerve standpoint, the pain pattern outlined above would suggest involvement of the DSN, long thoracic (lateral thoracic wall), suprascapular (scapular) and radial nerves (posterolateral arm and forearm) and possibly the axillary (posterolateral shoulder) and musculocutaneous nerves (posterolateral forearm). The common thread for these nerves would be the C5 nerve root suggesting a C5 peripheral nerve neuropathy. Anecdotally, the DSN, suprascapular, long thoracic and radial nerves are more commonly involved as a group.<sup>59</sup> The axillary and/or musculocutaneous nerves are rarely combined with the other four when this pain pattern is present. The median nerve also shares a C5 nerve root but again, anecdotally, is rarely involved. The reason for the inclusion or exclusion of particular C5 related peripheral nerves is unknown.

A review of the particular trigger point referral patterns outlined in Travell and Simons upper limb trigger point manual<sup>62</sup> revealed the following: the rhomboids (innervated by the DSN) refer locally as well as over the scap-

ula; the levator scapula (innervated by the DSN in 31%-100%<sup>25,36</sup>) refers to the rhomboid, scapula and posterior shoulder area; the serratus anterior (innervated by the long thoracic nerve) refers to the rhomboid area as well as the medial arm, forearm and hand (in the distribution of the ulnar nerve); the scalenes (innervated segmentally), supra and infraspinatus (innervated by the suprascapular nerve) and the tricep brachii (innervated by the radial nerve) refer pain along the upper back, across the scapula, down the posterolateral arm to the hand in the exact pain pattern described. The posterior deltoid (innervated by the axillary nerve) will refer locally and into the posterior upper arm while the coracobrachialis (innervated by the musculocutaneous nerve) will refer in the posterolateral arm and forearm.62 The bicep brachii and brachialis (also innervated by the musculocutaneous nerve) will refer locally and to the cubital fossa.62

Neither the trigger point nor the peripheral nerve theories seem to completely explain the posterolateral pain pattern described in DSN neuropathy and further study to determine the cause is suggested.

#### Etiopathology

Sultan *et al.*<sup>1</sup> suggests three possible mechanisms for the cause of pain in patients with a known DSN neuropathy. The first is entrapment or stretch, whether acute or prolonged, of the nerve can induce neuropathic trunk pain which involves the nervi nervorum. The second mechanism is the presence of myofascial pain syndrome with subsequent DSN entrapment in the taut bands of the rhomboids containing the trigger points. The third mechanism is the pain is caused by the stretching of the cutaneous nerves from the thoracic posterior primary rami to the area due to scapula winging – similar to the mechanism proposed to explain NP – even though the pain may be there in the absence of scapular winging.<sup>1</sup>

The first mechanism outlined by Sultan *et al.*<sup>1</sup> is a plausible initiating factor of the neuropathic trunk pain and subsequent pain pattern. This is due to the activation of the nervi nervorum that are nociceptive<sup>63,64</sup> and irritated with stretching of the nerve they innervate. With nerve compression, Mackinnon<sup>65</sup> describes a process by which there is an interruption in the blood-nerve barrier that allows a leakage of fluid from the microvessels, or vasa nervorum, supplying the nerve. The blood-nerve barrier breakdown allows entry to, and an accumulation

of, inflammation related mediators that causes edema and eventually scar formation.<sup>65</sup> Sultan *et al.*<sup>1</sup> outlines a mechanism proposed by Ellis<sup>66</sup> to explain thoracic outlet syndrome that suggests that this type of trauma to the nervi nervorum creates a cycle of inflammation within the perineurium that results in an individual nerve "internal compartment syndrome". Mackinnon describes a similar process calling it a "mini-compartment" syndrome within the nerve.<sup>65</sup> With repeated or continued trauma, there is a vicious cycle of neural desensitization that causes more inflammation with even less trauma.

Along with the inflammation within the nerve, Ellis<sup>66</sup> suggests that highly innervated and inflammatory fibrous bands and persistent adhesions are formed that adhere the nerve to adjacent structures which further limits the movement of the nerve. This seemingly describes the mechanism behind the double crush syndrome as outlined by Upton and McComas.<sup>67</sup> They originally hypothesized that proximal level nerve compression could cause more distal sites to become compressed<sup>65,67</sup> calling it a "double crush". This was further expanded to suggest that it was more of a multiple crush situation and also that more distal compression sites could cause proximal site compression, a reverse double crush.65 The development of the fibrous bands along the route of the nerve would create individual tension sites adding to the inflammation and suggesting the need to evaluate the nerve along its entire route prior to treatment. This could also help explain the estimated 1% to 25% failure rate for carpal tunnel release surgery.68

The increasing compartment pressure in the nerve causes more compression of the nervi nervorum causing further pain and fibroblast proliferation, and continued fibrosis. The inflammation, if not identified and treated appropriately, can spread both perineurally (within the nerve) and endoneurally causing activation of the CNS.<sup>66</sup> Ellis<sup>66</sup> suggests that this can then cause the subsequent inflammation of other adjacent/connected nerves, and can lead to mirror symptoms in the opposite limb.

Although there is debate in the literature regarding the existence of active and latent trigger points<sup>60,61</sup> as a theory of myofascial pain syndrome, most manual therapists would agree that taut, palpable, painful bands in muscle can be found in all areas of the musculoskeletal system. It is conceivable that these taut bands could be initiated by DSN neuropathy and not the reverse. In 21 of the 29 patients with DSN neuropathy in the paper by Sultan *et al.*<sup>1</sup>, the primary diagnosis was myofascial pain syndrome of the rhomboid major with identified taut bands. Further to this, the taut bands could cause entrapment of the thoracic medial cutaneous nerves from the thoracic posterior primary rami that supply this area as they pass directly through the rhomboid and trapezius on their way to the skin.<sup>12</sup> To continue this argument, if DSN neuropathy causes scapular winging in these individuals, traction of these cutaneous branches could occur causing both compression and traction. This would create continued inflammation and nervi nervorum irritation within these nerves causing both pain and sensory changes in the area. (See Figure 3).

This may help to explain the mechanism behind NP. It is interesting to note that two of the patients with DSN neuropathy in the paper by Sultan *et al.*<sup>1</sup> had both pruritis, (one of the two had diminished pinprick sensation) and scapular winging. This may also be the cause of the pain associated with skin rolling in the area as reported by Maigne<sup>6</sup> with regard to CD, as discussed. Interestingly, the "cervical point of the back" outlined by Maigne<sup>6</sup> could also be due to irritation of the skin at the spinous innervated by the medial branch of the posterior primary rami. It may also help to explain the limited therapeutic value of the use of topical applications in NP<sup>55</sup> in that the cutaneous nerves may only play a partial role. (See Figure 3).

The spread of the inflammation into adjacent nerves as outlined by Ellis<sup>66</sup>, would help to explain the posterolateral arm pain pattern (suggested C5 peripheral nerve neuropathy) as outlined above. As the inflammation spreads to the other nerves, intuitively, they also become susceptible to fibrosis and persistent adhesions - their own multiple crush situation.<sup>65</sup> This becomes very important clinically, as Mackinnon further states, "this concept of double or multiple crush is important clinically in patients who demonstrate multiple levels of nerve compression, as failure to diagnose and treat these multiple levels of injury will result in a failure to relieve the patients' symptoms."65. The effectiveness of treating some or all of the C5 nerves (DSN, suprascapular, long thoracic and radial) and the effect on outcomes should be investigated. (See Figure 3).

DSN neuropathy, like other neuropathies, may exist on a spectrum that includes various levels of pain intensi-



Figure 3.

Proposed mechanism of dorsal scapular nerve neuropathy, notalgia paresthetica, C5 peripheral nerve neuropathy and SICK scapula/scapular winging.

ty, dysesthesia and tingling and atrophy of the supplied musculature. It may also show various levels of spread to other adjacent nerves. Anecdotally, most patients reach a particular level of pain only and do not progress to dysesthesia/numbness/tingling or atrophy or irrecoverable nerve damage. It is assumed that the Etiopathology as proposed (in Figure 3) must reach an "equilibrium" of sorts and the patient's pain will wax and wane within a defined range (ie. a 2 to a 4 on the pain scale). How or why this equilibrium is reached in some patients and not others is not within the scope of this paper but may be due to the level of the initial trauma, the length of time of the chronic compressive/traction cycle and/or the nerve fibre type affected by the inflammatory cycle. The cycle could also be affected by further acute trauma (such as a new or second motor vehicle accident) or a change in the overuse related trauma to the affected area (such as a new (or a change) in job description ie. overhead lifting, or constant sitting with poor posture).

#### Conclusion

This article provides a review of the origin, pathway, and function of the DSN along with the epidemiology, clinical presentation and proposed etiologies of DSN neuropathy.

DSN neuropathy may be associated with or may be the entity known as cervicogenic dorsalgia. It may also play a role in notalgia paresthetica, SICK scapula and a commonly reported pain pattern along the posterolateral arm and forearm associated with neck, upper back and shoulder pain. Further study is recommended to explore the possible role of DSN neuropathy in these conditions and the possible etiopathology theorized.

It is recommended that DSN neuropathy be considered as a potential contributor to upper to mid thoracic pain and may be the sole cause in some cases. It is hoped that practitioners will begin to include it in their list of differentials in this group of patients.

### Acknowledgment

Thank you to Dr. Guy Sovak for the preparation and pictures of the dissection used in this paper.

#### References

- 1. Sultan HE, El-Tantawi GA. Role of dorsal scapular nerve entrapment in unilateral interscapular pain. Arch Phys Med Rehab. 2013;94(6):1118-1125.
- Aronsson G, Gustafsson K, Dallner M. Sick but yet at work. An empirical study of sickness and presenteeism. J Epidemiol Community Health. 2000; 54:502-509.
- Briggs AM, Smith AJ, Straker LM, Bragge P. Thoracic spine pain in the general population: prevalence, incidence and associated factors in children, adolescents and adults. A systematic review. BMC Musculoskel Dis. 2009;10(1):77.
- Linton SJ, Hellsing AL, Hallden K. A population-based study of spinal pain among 35-45-year-old individuals. Prevalence, sick leave, and health care use. Spine. 1998; 23:1457-1463.
- Hincapié CA, Cassidy JD, Côté P, Carroll LJ, Guzmán J. Whiplash injury is more than neck pain: a populationbased study of pain localization after traffic injury. J Occupation Environ Med. 2010;52(4):434-440.
- 6. Maigne R. Orthopedic Medicine: a New Approach to Vertebral Manipulations. Springfield, IL: Charles C. Thomas, 1972: 255-266.
- Engel G, Gatterman M. Cervicogenic Dorsalgia in: ed. Gatterman M. Foundations of Chiropractic: Subluxation. Elsevier Heatlh Sciences, 2005. 448-456.
- Ballesteros LE, Ramirez LM. Variations of the origin of collateral branches emerging from the posterior aspect of the brachial plexus. J Brachial Plex Peripher Nerve Inj. 2007; 2: 14.
- 9. Shilal P, Sarda RK, Chhetri K, Lama P, Tamang BK. Aberrant dual origin of the dorsal scapular nerve and its communication with long thoracic nerve: an unusual variation of the brachial plexus. J Clin Diagnost Res. 2015;9(6):AD01.
- Tubbs RS, Tyler-Kabara EC, Aikens AC, Martin JP, Weed LL, Salter EG, Oakes WJ. Surgical anatomy of the dorsal scapular nerve. J Neurosurg. 2005;102(5):910-911.
- 11. Richardson BS, Way BV, Speece AJ. Osteopathic manipulative treatment in the management of notalgia

paresthetica. J Am Osteopath Assoc. 2009;109(11):605-608.

- Wang CK, Gowda A, Barad M, Mackey SC, Carroll IR. Serratus muscle stimulation effectively treats notalgia paresthetica caused by long thoracic nerve dysfunction: a case series. J Brachial Plex Peripher Nerve Inj. 2009; 4:17.
- 13. Ellis C. Notalgia paresthetica: the unreachable itch. Dermatol Pract Concept. 2013; 3(1):3.
- 14. Shin J, Kim YC. Neuropathic itch of the back: a case of notalgia paresthetica. Ann Dermatol. 2014;26(3):392-394.
- 15. Pérez-Pérez LC. General features and treatment of notalgia paresthetica. Skinmed. 2011;9(6):353-358.
- Tacconi P, Manca D, Tamburini G, Cannas A, Giagheddu M. Notalgia paresthetica following neuralgic amyotrophy: a case report. Neurolog Sci. 2004;25(1):27-29.
- 17. Dixon AF. Abnormal distribution of the nervus dorsalis scapulæ, and of certain of the intercostal nerves. J Anat Physiol. 1896; 30(Pt 2): 209–210.
- Burkhart SS, Morgan CD, Ben Kibler W. The disabled throwing shoulder: spectrum of pathology Part III: The SICK scapula, scapular dyskinesis, the kinetic chain, and rehabilitation. Arthroscopy. 2003;19(6):641-661.
- 19. Akgun K, Aktas I, Terzi Y. Winged scapula caused by a dorsal scapular nerve lesion: a case report. Arch Phys Med Rehabil. 2008;89: 2017-2020.
- 20. Ravindran M. Two cases of suprascapular neuropathy in a family. Br J Sports Med. 2003; 37:539-541.
- Chen D, Gu Y, Lao J, Chen L. Dorsal scapular nerve compression. Atypical thoracic outlet syndrome. Chin Med Journ. 1995;108(8):582-585.
- 22. Brower RS. Differential diagnosis of cervical radiculopathy and myelopathy. In: Clark CR, ed. The Cervical Spine. 4th ed. Philadelphia, Lippincott Williams & Wilkins; 2005: 995-1008.
- Hammer WI, ed. Functional Soft-Tissue Examination and Treatment by Manual Methods Third Edition. Sudbury, MA: Jones and Bartlett Publishers, 2007: 140-141.
- 24. Lee HY, Chung IH, Seok W, Kang HS, Lee HS, Ko JS, Lee MS, Park SS. Variations of the ventral rami of the brachial plexus. J Kor Med Sci. 1992;7(1):19-24.
- 25. Nguyen VH, Liu HH, Rosales A, Reeves R. A cadaveric investigation of the dorsal scapular nerve. Anat Res Intl. 2016 Aug 1.
- 26. Malessy MJ, Thomeer RT, Marani E. The dorsoscapular nerve in traumatic brachial plexus lesions. Clin Neurol Neurosurg. 1993;95:17-23.
- 27. Pecina MM, Krmpotic-Nemanie J, Markiewitz AD. Tunnel syndrome: peripheral nerve compression syndromes. Boca Raton: CRC Press, Inc, 1997: 59-61.
- Debeer P, Van Den Eede E, Moens P. Case report: scapular winging: an unusual complication of bracing in idiopathic scoliosis. Clin Orthoped Rel Res. 2007; 461:258-261.

- 29. Olinger AB, Homier P. Functional anatomy of human scalene musculature: rotation of the cervical spine. J Manip Physiol Ther. 2010;33(8):594-602.
- 30. Dahlstrom KA, Olinger AB. Descriptive anatomy of the interscalene triangle and the costoclavicular space and their relationship to thoracic outlet syndrome: a study of 60 cadavers. J Manip Physiol Ther. 2012; 35(5):396-401.
- Moore, KL. Clinically oriented anatomy. 2<sup>nd</sup> Edition. Baltimore: Williams & Wilkins, 1985: 664, 994.
- Buford JA, Yoder SM, Heiss DG, Chidley JV. Actions of the scalene muscles for rotation of the cervical spine in macaque and human. J Orthop Sports Phys Ther. 2002; 32(10):488-496.
- Rusnak-Smith S, Moffat M, Rosen E. Anatomical variations of the scalene triangle: dissection of 10 cadavers. J Orthop Sports Phys Ther. 2001;31(2):70-80.
- George BM, Nayak SB. Neuromuscular variations in the posterior triangle of the neck–a case report. Neuroanat. 2008; 7:8-9.
- Martin RM, Fish DE. Scapular winging: anatomical review, diagnosis, and treatments. Curr Rev Musculoskelet Med. 2008; 1:1-11.
- Frank DK, Wenk E, Stern JC, Gottlieb RD, Moscatello AL. A cadaveric study of the motor nerves to the levator scapulae muscle. Otolaryngol Head Neck Surg. 1997; 117(6):671-80
- 37. Perri MA, Halford E. Pain and faulty breathing: a pilot study. J Bodywork Move Ther. 2004;8(4):297-306.
- Kida MY, Tani M. The human superior posterior serratus muscle supplied by both the intercostal and dorsal scapular nerves. Kaibogaku zasshi. J Anat. 1993;68(2):162-168.
- 39. Vilensky JA, Baltes M, Weikel L, Fortin JD, Fourie LJ. Serratus posterior muscles: anatomy, clinical relevance, and function. Clin Anat. 2001;14(4):237-241.
- Wood VE, Marchinski L. Congenital anomalies of the shoulder. In: Rockwood CA, Matsen FA. Eds. The Shoulder. Vol 1. Philadelphia: WB Saunders; 1998. 99-163.
- 41. Duralde XA. Neurologic injuries in the athlete's shoulder. J Athl Train. 2000;35(3):316.
- 42. Fruth SJ. Differential diagnosis and treatment in a patient with posterior upper thoracic pain. Phys Ther. 2006;86(2):254-268.
- Benedetti MG, Zati A, Stagni SB, Fusaro I, Monesi R, Rotini R. Winged scapula caused by rhomboid paralysis: a case report. Joints. 2016 Oct;4(4):247.
- 44. Argyriou AA, Karanasios P, Makridou A, Makris N. Dorsal scapular neuropathy causing rhomboids palsy and scapular winging. J Back Musculoskel Rehabil. 2015;28(4):883-885.
- 45. Kaplan Y, Kurt S, Karaer H. Isolated dorsal scapular neuropathy associated with repetitive minor trauma: a case report. Arch Neuropsychiatry. 2008; 45:107-109.
- 46. Lee SG, Kim JH, Lee SY, et al. Winged scapula caused by

rhomboideus and trapezius muscle rupture associated with repetitive minor trauma: a case report. J Korean Med Sci. 2006; 21: 581-584.

- Plezbert JA, Nicholson CV. Dorsal scapular nerve entrapment neuropathy: a unique clinical syndrome. J Neuromusculoskel System. 1994; 2(4): 206 – 211.
- Mondelli M, Cioni R, Federico A. Rare mononeuropathies of the upper limb in bodybuilders. Muscle Nerve. 1998;21(6):809-812.
- Haim K, Urban BJ. Dorsal scapular nerve block: description of technique and report of a case. Anesthesiology 1993;78: 361-363.
- Kugler A, Krüger-Franke M, Reininger S, Trouillier HH, Rosemeyer B. Muscular imbalance and shoulder pain in volleyball attackers. Br J Sports Med. 1996;30(3):256-259.
- 51. Jerosch J, Castro WH, Geske B. Damage of the long thoracic and dorsal scapular nerve after traumatic shoulder dislocation: case report and review of the literature. Acta Orthop Belg. 1990; 56: 625-627.
- 52. Terrett AG, Terrett RG. Referred posterior thoracic pain of cervical posterior rami origin: a Ccause of much misdirected treatment. Chiropr J Austral. 2002;32(2):42-51.
- Savk O, Savk E. Investigation of spinal pathology in notalgia paresthetica. J Amer Acad Dermatol. 2005; 52(6):1085-1087.
- 54. Raison-Peyron N, Meunier L, Acevedo M, Meynadier J. Notalgia paresthetica: clinical, physiopathological and therapeutic aspects. A study of 12 cases. J Euro Acad Dermatol Venereol. 1999;12(3):215-221.
- 55. Wallengren J, Klinker M. Successful treatment of notalgia paresthetica with topical capsaicin: vehicle-controlled, double-blind, crossover study. J Amer Acad Dermatol. 1995;32(2):287-289.
- 56. Alai NN, Skinner HB, Nabili ST, Jeffes E, Shahrokni S, Saemi AM. Notalgia paresthetica associated with cervical spinal stenosis and cervicothoracic disk disease at C4 through C7. Cutis. 2010;85(2):77-81.
- 57. Fleischer AB, Meade TJ, Fleischer AB. Notalgia paresthetica: successful treatment with exercises. Acta Dermato-Venereologica. 2011;91(3):356-357.
- Lee MW, McPhee RW, Stringer MD. An evidencebased approach to human dermatomes. Clin Anat. 2008;21(5):363-373.
- 59. Muir B, Kissel JA, Yedon DF. Intraosseous ganglion cyst of the humeral head in a competitive flat water paddler: case report. J Can Chiropr Assoc. 2011 ;55(4):294.
- 60. Quintner JL, Cohen ML. Referred pain of peripheral nerve origin: an alternative to the "myofascial pain" construct. Clin J Pain. 1994;10:243-251.
- 61. Dommerholt J, Gerwin RD. A critical evaluation of Quintner et al: Missing the point. J Bodywork Movement Ther. 2015;19(2):193-204.
- 62. Simons DG, Travell JG. Myofascial pain and dysfunction:

the trigger point manual. The upper extremities. Volume 1. Williams & Wilkins; 1983: 335, 345, 369, 378, 428, 432, 441, 448, 457, 464, 623.

- 63. Bove GM, Light AR. The nervinervorum: missing link for neuropathic pain? J Pain. 1997;6:181-190.
- 64. Sauer SK, Bove GM, Averbeck B, Reeh PW. Rat peripheral nerve components release calcitonin generelated peptide and prostaglandin E 2 in response to noxious stimuli: evidence that nervi nervorum are nociceptors. Neurosci. 1999 ;92(1):319-325.
- 65. Mackinnon SE. Pathophysiology of nerve compression. Hand Clin. 2002;18(2):231-241.
- 66. Ellis W. Thoracic outlet syndrome as a disorder of neurogenic inflammation. Vasc Endovasc Surg. 2006;40:251-244.
- 67. Upton AM, Mccomas A. The double crush in nerveentrapment syndromes. Lancet. 1973; 302(7825):359-362.
- Neuhaus V, Christoforou D, Cheriyan T, Mudgal CS. Evaluation and treatment of failed carpal tunnel release. Orthoped Clin North Am. 2012;43(4):439-447.

## Perianal abscess mimicking levator ani syndrome: a case report and approach to the differential diagnosis of anorectal pain.

Paul Mastragostino, HBSc.Kin, DC, FCCS (C)<sup>1</sup> Alexander D. Lee, BSc, DC, FRCCSS(C)<sup>2</sup> Patrick J. Battaglia, DC, DACBR<sup>3</sup>

Objective: To present the clinical management and comprehensive differential diagnosis of a patient with anorectal pain from a perianal abscess.

Clinical Features: A 41-year-old woman presented with pain localized to her perianal and gluteal region, accompanied by internal and external rectal pain. Prior to presentation, the patient had received a working diagnosis of levator ani syndrome.

Intervention and Outcome: An interdisciplinary management approach was utilized. Diagnostic imaging confirmed the clinical suspicion of a perianal abscess and the patient underwent surgical drainage.

Summary: Anorectal pain is complex and multifactorial and a diagnosis such as an abscess should not be overlooked. This case emphasized that Objectif : Présenter le traitement clinique et le diagnostic différentiel complet d'une patiente atteinte de douleur ano-rectale découlant d'un abcès périanal.

Caractéristiques cliniques : Une femme de 41 ans se présente avec une douleur dans la région périanale et fessière, accompagnée de douleur rectale interne et externe. Avant la présentation, la patiente a reçu un diagnostic de travail de syndrome du muscle élévateur de l'anus.

Intervention et résultats : On a utilisé une approche de prise en charge interdisciplinaire. L'imagerie diagnostique a permis de confirmer le soupçon clinique d'abcès périanal et la patiente a subi un drainage chirurgical.

Résumé : La douleur ano-rectale est complexe et multifactorielle; il ne faut pas négliger un diagnostic tel qu'un abcès. Ce cas souligne que les praticiens doivent

<sup>1</sup> Department of Graduate Education and Research Programs, Canadian Memorial Chiropractic College, Toronto, ON

<sup>2</sup> Clinical Faculty, Canadian Memorial Chiropractic College, Toronto, ON

<sup>3</sup> Radiology Department, Logan University, Chesterfield, MO

Corresponding Author: Paul Mastragostino Department of Graduate Education and Research Programs, Canadian Memorial Chiropractic College Toronto, Ontario, Canada, M2H 3J1 e-mail: paul.mastragostino@gmail.com

Consent: The patient has provided written consent to having their personal health information, including radiographs, published.

The authors have no competing interests to declare. © JCCA 2017

practitioners must be diligent in their evaluation and management of patients with anorectal pain, including recognizing situations that require further imaging and interdisciplinary management.

(JCCA. 2017;61(2):145-152)

KEY WORDS: chiropractic, perianal abscess, levator ani syndrome, anorectal pain, differential diagnosis

#### Introduction

Anorectal pain can be a debilitating condition for patients and a diagnostic challenge for health practitioners. Chronic or recurrent pain in the anal, rectal, or pelvic region has been reported to occur in 7% to 24% of the population.<sup>1</sup> Anorectal pain is very heterogeneous and can range from mild discomfort secondary to myofascial pain, to incapacitating pain with life-threatening consequences, such as in the case of abscess formation.<sup>2,3</sup> Nonspecific symptoms and a poor understanding of etiology compounds the difficulties surrounding management of patients with anorectal pain and may lead to chronic and persistent symptoms.<sup>2</sup> There are cases where chronic pelvic and anorectal pain cannot be explained by a structural or other specified pathology and are termed functional chronic pain disorders.<sup>4</sup> These conditions are poorly understood and are often under recognized pain syndromes associated with significant impairment, decreased quality of life, and psychological distress.<sup>1</sup> Although practitioners should be mindful of functional disorders in patients with chronic anorectal pain, this diagnosis is one of exclusion after structural causes are ruled out. Diagnostic considerations for structural pathology that are commonly associated with chronic anorectal pain includes cryptitis, fissure, abscess, hemorrhoids, solitary rectal ulcer, inflammatory bowel disease, and rectal ischemia.1 Accurate diagnosis can often be made based on a thorough history, physical examination, and necessary ancillary work-up including diagnostic imaging and laboratory investigation.5

We present a case of a patient with a perianal abscess initially misdiagnosed as myofascial pain of the levator ani muscle. This case report emphasizes the importance faire preuve de diligence lors de leur évaluation et prise en charge des patients qui souffrent de douleur anorectale, y compris reconnaître des situations nécessitant une imagerie et une prise en charge interdisciplinaire supplémentaires.

(JCCA. 2017;61(2):145-152)

MOTS CLÉS : chiropratique, abcès périanal, syndrome du muscle élévateur de l'anus, douleur ano-rectale, diagnostic différentiel

of a broad differential diagnosis for chronic anorectal pain. Additional diagnostic evaluation and interdisciplinary management is also discussed.

#### **Case Presentation**

A 41-year-old woman presented to an academic chiropractic clinic with complaints of severe sacrococcygeal and gluteal pain, accompanied by burning and stabbing internal and external rectal pain. The symptoms had been intermittent for 6 months, lasting 3-4 days at a time, but had become constant the week prior to her visit. There was no history of trauma or other mechanism for her pain. Aggravating factors included prolonged sitting, shifting her weight while sitting, flatulence, and bowel movements. She denied any hematochezia or hematuria associated with the onset of her pain.

Her medical history was remarkable for ulcerative colitis, diagnosed at age 29, in addition to anxiety and depression. Noted medications included Pentasa (Mesalamine) and monthly intravenous immunoglobulin (IVIG) for her colitis. She had visited her gastrointestinal specialist the preceding month for an endoscopy, the results of which were reportedly unremarkable. She was diagnosed by her specialist at that time with a suspected levator ani syndrome (i.e. myofascial pain of the levator ani muscle) and was instructed to seek physical therapy for management of her symptoms.

During the initial examination at the chiropractic clinic, the patient was in visible pain and had difficulty ambulating. There was no observed swelling, discolouration, or deformity visualized over the gluteal or sacrococcygeal region. Lumbar range of motion was severely reduced in all directions due to pain. A lower limb neurological examination was unremarkable. Gluteal pain was reproduced with palpation of the sacrotuberous ligaments bilaterally, and digital pressure over the L4 and L5 vertebral segments during prone examination. Additionally, tenderness was elicited with palpation of the sacrum and external coccyx, as well as gluteal musculature bilaterally. During the physical examination, paroxysmal pain lasting several seconds, and not provoked by movement or testing maneuvers, was reported.

Due to the severity of her pain and the erratic nature of her symptoms, radiographs of the sacrum and coccyx were ordered, demonstrating enlargement of the pre-sacral soft tissues (Figure 1). There was no effacement of the colon wall or underlying bone destruction on the radiograph. The patient was subsequently referred to her family physician for blood tests as well as pelvic and abdominal ultrasounds to further evaluate pelvic soft tissues and rule out a potential space occupying lesion, such as from a sacral neoplasm.

She was also treated by a chiropractic intern on three separate occasions over the course of three weeks to provide symptomatic relief while she was awaiting further diagnostic testing. Ischemic compression was applied to the sacrotuberous and sacrospinous ligaments and the surrounding hip and pelvic musculature. Grades 1 and 2 mobilizations were performed at the sacroiliac joint. After the second visit the patient reported a subjective decrease in the duration and intensity of her symptoms, with minimal pain on bowel movements. On the third visit the patient described a severe exacerbation of her rectal and gluteal pain, return of pain with bowel movements, and an incidence of bleeding during a bowel movement.

Blood tests revealed mild anemia, as well as elevated neutrophils (8.2 [ref. 2.0-7.5] xE9/L) and high-sensitive C-reactive protein (6.2 [ref. < 3] mg/L), suggestive of inflammation or infection. An abdominal ultrasound was normal. A pelvic and transvaginal ultrasound revealed non-specific mild thickening of a segment of the sigmoid colon with retention of the normal wall layers. The patient was then referred for a pelvic MRI, performed two weeks following her initial assessment at the chiropractic clinic. The MRI revealed evidence of a near circumferential horse shoe shaped abscess in the intersphincteric plane, measuring 1cm at its widest area; the opening approximately 3cm from the anus (Figure 2). A left-sided



#### Figure 1.

Conventional lateral radiograph of the sacrum and coccyx demonstrating enlargement of the pre-sacral soft tissues (asterisk). The posterior colonic wall (arrows) is uneffaced and there are no destructive sacral lesions. Although initially interpreted as evidence for a presacral mass, the enlarged soft tissues in this patient were secondary to fat deposition. This is an established complication of chronic ulcerative colitis.<sup>6</sup>

Bartholin cyst was noted which may have been secondary to the underlying inflammatory process producing ductal obstruction. There was no pre-sacral mass or fluid collection, but rather a notable amount of fat surrounding the colon at this location consistent with the history of chronic inflammatory bowel disease.<sup>6</sup>

Based on these findings, a diagnosis of perianal abscess was made by the gastroenterologist. The patient was subsequently scheduled for urgent surgery approximately three and a half weeks post-MRI. Incidentally, a pre-surgical screen of the patient revealed a pyoderma gangrenosum lesion initiating over her right shin; most likely secondary to her chronic inflammatory bowel condition.

At the hospital, she underwent an examination under



Figure 2.

Axial T2-weighted fast spin-echo sequences. (A) Perirectal fluid collection consistent with a horseshoe shaped abscess is outline by the arrows. The centrally located rectum (asterisk) is also seen. (B) A Bartholin gland cyst (double asterisk) can be seen on the left side of the distal vagina. Smaller discrete fluid collections are noted in a similar location on the patient's right side. Pelvic inflammation from the abscess may have caused duct obstruction and cyst formation.

anesthesia which identified an internal opening of the abscess. Pus was expressed through the internal opening. No external opening or fistula was identified and no incision was required. The next day IVIG was administered to the patient. Her peri-anal pain remained minimal, however she did experience some stool leakage/incontinence the day following her examination. At discharge the patient was ambulating, tolerating an oral diet, and her peri-anal pain had generally resolved. Her right shin pain at the pyoderma gangrenosum lesions was under control with opioids, appeared stable, and was covered with a non-adherent dressing and gauze.

The patient returned to the chiropractic teaching facility for a follow-up appointment at one-month post-surgery. She had not experienced any episodes of incontinence since returning home from her surgery. She denied any recent fever, night pain, or night sweating; however, she reported a daily occurrence of blood in her stool since the surgery, for which she was being monitored for by her gastroenterologist. Moreover, she described complaints of persistent gluteal pain and associated sacral pain, aggravated by prolonged sitting, and lumbar stiffness, worse in the morning. On physical examination, she demonstrated full lumbar spine ranges of motion with segmental restrictions noted at the lumbar spine and sacroiliac joints. Point tenderness was reported with palpation of the left sacrotuberous and sacrospinous ligaments, and gluteal musculature. Apart from her pelvic complaints, the gangrenous lesion on her right shin was healing adequately.

The patient was treated by a chiropractic intern over the course of three consecutive visits over three weeks. Her therapy entailed spinal manipulation of the sacroiliac joints and lumbar spine, soft tissue therapy to the surrounding hip and gluteal structures, and exercises emphasizing core stability. She was contacted by telephone two months following her last chiropractic visit to inquire about her status. She reported that her symptoms had significantly improved to the extent where she was absent of pain for three to four weeks and was no longer experiencing bleeding with bowel moments.



Figure 3. Common anorectal conditions. Reproduced with permission from The Royal Australian College of General Practitioners from Daniel WJ. Anorectal pain, bleeding and lumps. Aust Fam Physician 2010;39(6):376-81. Available at www.racgp. org.au/afp/2010/june/ anorectal-pain,-bleedingand-lumps

#### Discussion

The differential diagnosis for chronic anorectal pain is extensive. Common organic causes include cryptitis, anal fissure, perianal abscess (with or without fistula), hemorrhoids, solitary rectal ulcer, inflammatory bowel disease, and rectal ischemia (Figure 3).<sup>2,7</sup> A thorough and carefully focused history and physical examination should be used to identify signs and symptoms suggestive of these pathologies. Clinicians should also be cognizant that in spite of a rigorous evaluation, previous literature has suggested that in approximately 85% of patients presenting to a specialist with chronic anorectal or pelvic pain, an organic disease explanation will not be found.<sup>1</sup> This may prompt the consideration of functional anorectal disorders.

In the present case the patient's medical history was remarkable for ulcerative colitis, a known risk factor for the development of anorectal complications.<sup>8</sup> It is estimated that approximately 15-20% of patients with ulcerative colitis will experience anorectal complications, the most common being anal fissures, seen in approximately 12% of patients, and anorectal abscesses and fistulas, seen in 5% of patients.<sup>8</sup> An anorectal abscess typically presents with pain that is constant and throbbing in character and exacerbated by ambulation and straining. This is consistent with the present patient as she experienced constant pain that fluctuated in waves of intensity, and she had difficulty ambulating due to pain. Swelling, ery-thema, fever, and a fluctuant mass are additional signs and symptoms that may be present.<sup>8-10</sup>

Abscesses are classified according to their anatomical location in the potential anorectal spaces formed by the inner and outer layers of the pelvic floor musculature: perianal (most common), ischiorectal, intersphincteric, and supralevator (least common).<sup>9-12</sup> The ability of these abscesses to track circumferentially through the anorectal spaces around the anus or the rectum can result in the formation of a horseshoe abscess<sup>9-11</sup>, as was the circumstance in the present case. Timely aggressive surgical treatment under anesthesia, as done in this case, is advised to avoid prolonged morbidity and ensure low re-

currence rates.<sup>13</sup> Consequences of delayed or inadequate treatment of an anorectal abscess may be severe or even fatal. If not drained, an abscess may spread and result in fistula formation, bacteremia and sepsis, and/or tissue necrosis.<sup>8,12,13</sup> Given the susceptibility of colitis patients to anorectal complications, structural origins for chronic anorectal pain, such as an abscess, should be prioritized accordingly when considering differential diagnoses with these patients.

For many patients with chronic anorectal pain, the source of the pain cannot be definitively explained by a specific pathophysiological mechanism<sup>1</sup>, and these cases are commonly defined as functional anorectal disorders.<sup>4,7,14-16</sup> The absence of conclusive diagnostic biological markers for these functional gastrointestinal conditions has led to the development of the Rome diagnostic criteria for functional gastrointestinal disorders, that recognizes two forms of functional anorectal pain: chronic proctalgia and proctalgia fugax.<sup>4</sup> A diagnosis of chronic proctalgia is made based on a history of chronic or recurring episodes of rectal pain, each lasting 20 minutes or longer, and exclusion of other causes of rectal pain. This criterion is fulfilled for the last 3 months with symptom onset at least 6 months prior to diagnosis.<sup>17</sup> Chronic proctalgia is further characterized into two subtypes, levator ani syndrome and unspecified functional anorectal pain, based on the presence or absence of reported tenderness during posterior traction on the puborectalis musculature.<sup>17</sup> This taxonomy reflects the dominant hypothesis regarding the pathophysiological basis of levator ani syndrome, which is chronic tension or spasticity of the pelvic floor musculature.<sup>1,3,16</sup> These pain syndromes pose many diagnostic challenges for clinicians<sup>15</sup>, largely owing to considerable overlap of their symptoms, frequent coexistence, unknown etiology and pathogenesis, as well as unpredictable prognosis<sup>18</sup>. Consequently, a comprehensive differential diagnostic process in which worrisome considerations are systematically excluded should be the foundation for an accurate diagnosis.1,18

Obtaining a detailed history of a patient with chronic anorectal and/or perianal pain is a vital component of the clinical interaction. This is especially relevant for clinicians whose scope of practice may limit the performance of certain diagnostic techniques or procedures on these patients, such as chiropractors operating in a predominantly musculoskeletal environment. Billingham and col-

1) 2)	Where does it hurt? When does it hurt?	7)	Does anything make the pain better?		
3)	When did the pain start?	8)	Does anything make the pain worse?		
4)	How long does the pain last?	9)	Is the pain associated with other symptoms,		
5)	What is the character of the pain?	10)	such as rectal bleeding? Do bowel movements		
6)	Does the pain radiate or travel?		make the pain better or worse?		
Figure 4.					



leagues<sup>9</sup> advocate for a system-based approach in the assessment of anorectal pain that incorporates specific questions necessary for guiding the physical examination and the decision-making process (Figure 4). The authors note that these questions are not exhaustive or inclusive, and supplementary questioning may be necessarily applied to the individual symptoms. It is critical to inquire about pain with defecation, as bowel movements are unlikely to be associated with functional anorectal pain (such as levator ani syndrome)<sup>2,4</sup>, and in fact may even be relieving for patients with levator muscle spasms<sup>9</sup>.

Anorectal pain that begins intensely during a bowel movement that may persist for minutes to hours after defecation is characteristically associated with an anal fissure<sup>2,3,9</sup>; whereas, pain that is constant and is not altered markedly by bowel movements is more typical of anal abscesses and thrombosed hemorrhoids9. A small amount of bright red blood on toilet paper or in the bowl during defecation is often associated with hemorrhoids; however, depending on the hemorrhoid severity, pain may not be a main feature.<sup>19</sup> Moreover, if recurrent abdominal pain or discomfort is reported by the patient, along with changes in bowel habits (frequency and/or consistency of stools) and relief following defecation, a diagnosis of irritable bowel syndrome should be taken into account.<sup>3,4</sup> A diagnosis of functional anorectal pain is also less likely if the pain is associated with eating or menses.<sup>4</sup>

A comprehensive examination that includes inspection and palpation is necessary in most instances to substantiate a diagnosis in cases of anorectal pain.<sup>24,9</sup> The examination should begin with observation of the patient's gait and sitting habits, to identify guarding of a particular body region or side<sup>20</sup>, followed by inspection of the perianal region for signs of structural disease. Specific signs may include the presence of sentinel piles or skin tags, fistulous opening, pain while gently parting the anterior or posterior perianal tissue, in addition to the detection of excoriations, scars, anal strictures or indurations.<sup>2,3,9</sup> The patient must be fully informed of the practitioner's intent and provide explicit consent before the examination is carried out. Offering to have a guardian or chaperone (e.g. the clinic's administrative assistant) present for the examination will help facilitate patient comfort.

Following the external inspection, a digital examination should be included to evaluate sphincter tone, height and symmetry, as well as to palpate for masses and areas of tenderness.<sup>2,9</sup> For some clinicians, performing a digital rectal examination may not be possible due to licensing restrictions; in which case co-management of these patients with an appropriate health professional must be established to ensure a complete examination of the area is carried out so as to guide management and the need for further diagnostic testing. Lab tests (such as complete blood count and erythrocyte sedimentation rate), anoscopy/proctoscopy, flexible sigmoidoscopy, and perianal imaging with ultrasound, MRI or CT scan may be necessary, contingent on the results of the history and examination.<sup>2,4,14</sup>

Although not the primary focus of this report, specific therapeutic interventions for patients with functional and chronic anorectal and pelvic pain disorders may include manual or manipulative techniques. Frequently recommended treatments for patients with conditions such as levator ani syndrome and coccydynia include digital massage of the levator ani musculature and/or mobilization of the coccyx.<sup>21-26</sup> These procedures are typically performed intra-rectally to allow for better access to the intended structures; however, practitioners must be cautious that carrying out these interventions falls within their professional scope of practice to do so. In addition to the precautions that must be taken from a legal perspective, one must be mindful of the potential harms that may be done from administering certain manual therapies in the presence of unidentified structural pathology.

#### Summary

This case demonstrates the critical need for clinicians to employ broad differential diagnoses when encountered with patients who report anorectal or perianal pain. The evaluation of these patients can be challenging due to a diversity of causative factors. The patient in this case presented to an academic chiropractic clinic with severe chronic anorectal and perianal pain that was initially thought to be due to a functional anorectal disorder, levator ani syndrome. Her symptoms were ultimately the result of a perianal abscess which was treated operatively. Functional pain syndromes, such as levator ani syndrome, are common and debilitating conditions; however, these disorders should be regarded as diagnoses of exclusion and considered only after a thorough evaluation has been performed to rule out structural causes of pain.

#### Acknowledgment

The authors thank Dr. Karen Ngo for her contribution to this manuscript.

#### References

- 1. Chiarioni G, Asteria C, Whitehead WE. Chronic proctalgia and chronic pelvic pain syndromes: new etiologic insights and treatment options. World J Gastroenterol. 2011; 17: 4447-4455.
- 2. Garg H, Singh S, Bal K. Approach to the diagnosis of anorectal disorders. J IMSA. 2011; 24: 89-90.
- Lacy BE, Crowell MD, DiBaise JK. Functional and motility disorders of the gastrointestinal tract. New York: Springer; 2015.
- 4. Bharucha AE, Wald AM. Anorectal disorders. Am J Gastroenterol. 2010;105: 786-794.
- 5. Moghaddasi M, Aghaii M, Mamarabadi M. Perianal pain as a presentation of lumbosacral neurofibroma: a case report. J Neurol Surg Rep. 2014; 75:e191-e193.
- 6. Gore RM. CT of inflammatory bowel disease. Radiol Clin North Am. 1989;27(4):717.
- Wald A, Bharucha AE, Cosman BC, Whitehead WE. ACG clinical guideline: management of benign anorectal disorders. Am J Gastroenterol. 2014; 109: 1141-1157.
- Way LW, Doherty GM. Current surgical diagnosis & treatment. 11th ed. Lange Medical Books/McGraw-Hill; 2006.
- Billingham RP, Isler JT, Kimmins MH, Nelson JM, Schweitzer J, Murphy MM. The diagnosis and management of common anorectal disorders. Curr Probl Surg. 2004; 41: 586-645.
- 10. Whiteford MH. Perianal abscess/fistula disease. Clin Colon Rectal Surg. 2007; 20: 102-9.

Perianal abscess mimicking levator ani syndrome: a case report and approach to the differential diagnosis of anorectal pain.

- 11. Parks AG, Gordon PH, Hardcastle JD. A classification of fistula-in-ano. Br J Surg. 1976; 63: 1-12.
- Schubert MC, Sridhar S, Schade RR, Wexner SD. What every gastroenterologist needs to know about common anorectal disorders. World J Gastroenterol. 2009; 15:3201-3209.
- 13. Ramanujam PS, Prasad ML, Abcarian H, Tan AB. Perianal abscesses and fistulas. Dis Col Rect. 1984; 27: 593-597.
- Bharucha AE, Trabuco E. Functional and chronic anorectal and pelvic pain disorders. Gastroenterol Clin North Am. 2008; 37: 685-696.
- 15. Wesselmann U, Burnett AL, Heinberg LJ. The urogenital and rectal pain syndromes. Pain. 1997; 73: 269-294.
- Whitehead WE, Wald A, Diamant NE, Enck P, Pemberton JH, Rao SS. Functional disorders of the anus and rectum. Gut. 1999; 45(suppl 2): II55-II59.
- Rome F. Guidelines Rome III diagnostic criteria for functional gastrointestinal disorders. Journal of gastrointestinal and liver diseases: JGLD. 2006;15(3):307-312.
- Armañanzas L, Arroyo A, Ruiz-Tovar J, López A, Santos J, Moya P, Gómez MA, Candela F, Calpena R. Chronic idiopathic anal pain. Results of a diagnostic-therapeutic protocol in a colorectal referral unit. CIR ESP. 2015; 93: 34-38.
- 19. Daniel WJ. Anorectal pain, bleeding and lumps. Aust Fam Physician. 2010; 39: 376-81.

- 20. Hull M, Corton MM. Evaluation of the levator ani and pelvic wall muscles in levator ani syndrome. Urol Nurs. 2009; 29: 225-231.
- 21. Chiarioni G, Nardo A, Vantini I, Romito A, Whitehead WE. Biofeedback is superior to electrogalvanic stimulation and massage for treatment of levator ani syndrome. Gastroenterology. 2010; 138: 1321-1329.
- 22. Thiele GH. Tonic spasm of the levator ani, coccygeus and piriformis muscle: relationship to coccygodynia and pain in the region of the hip and down the leg. Trans Am Proctol Soc. 1936; 37: 145-155.
- 23. Thiele GH. Coccygodynia and pain in the superior gluteal region and down the back of the thigh: Causation by tonic spasm of the levator ani, coccygeus and piriformis muscles and relief by massage of these muscles. JAMA. 1937; 109: 1271-1275.
- 24. Thiele GH: Coccygodynia: cause and treatment. Dis Colon Rectum. 1963; 6: 422-436.
- 25. Maigne JY, Chatellier G, Le Faou M, Archambeau M. The treatment of chronic coccydynia with intrarectal manipulation: a randomized controlled study. Spine. 2006; 31: E621-627.
- Maigne JY, Chatellier G. Comparison of three manual coccydynia treatments: a pilot study. Spine. 2001; 26: E479-483.

## Innovative application of Cox Flexion Distraction Decompression to the knee: a retrospective case series

Luigi Albano, BSc, DC1

Objective: The purpose of this study is to introduce the application of Cox flexion distraction decompression as an innovative approach to treating knee pain and osteoarthritis.

Methods: Six months of clinical files from one chiropractic practice were retrospectively screened for patients who had been treated for knee pain. Twenty-five patients met the criteria for inclusion. The treatment provided was Cox flexion distraction decompression. Pre-treatment and post-treatment visual analog pain scales (VAS) were used to measure the results. In total, eight patients presented with acute knee pain (less than three months' duration) and 18 patients presented with chronic knee pain (greater than three months) including two patients with continued knee pain after prosthetic replacement surgery.

Results: For all 25 patients, a change was observed in the mean VAS scores from 7.7 to 1.8. The mean number of treatments was 5.3 over an average of 3.0 weeks. Acute patient mean VAS scores dropped from Objectif : Le but de cette étude est de présenter l'application de la décompression par flexion-distraction de Cox en tant qu'approche novatrice de traitement de la douleur au genou et de l'arthrose.

Méthodologie : On a inspecté six mois de dossiers cliniques d'un cabinet de chiropratique rétrospectivement pour trouver des patients traités pour une douleur au genou. Vingt-cinq patients respectaient les critères d'inclusion. Le traitement fourni était la décompression par flexion-distraction de Cox. On a utilisé des échelles visuelles analogues (EVA) avant et après le traitement pour mesurer les résultats. Au total, 8 patients présentaient une douleur aiguë au genou (depuis moins de 3 mois) et 18 patients présentaient une douleur chronique au genou (depuis plus de 3 mois), dont 2 patients avec une douleur toujours présente après la chirurgie de remplacement de prothèse.

Résultats : Pour les 25 patients, on a observé un changement des résultats moyens de l'EVA de 7,7 à 1,8. Le nombre moyen de traitements était de 5,3 sur une moyenne de 3,0 semaines. Les résultats moyens de l'EVA des patients atteints de douleur aiguë ont chuté de 8,1 à 1,1 après 4,8 traitements sur 2,4 semaines. Les résultats

<sup>1</sup> Private practice, Windsor, ON

Corresponding author: Luigi Albano Walkerville Chiropractic, 1275 Walker Road, Windsor, ON N8Y 4X9 Tel: 519-258-7979 E-mail: dr.luigialbano@gmail.com © JCCA 2017 8.1 to 1.1 within 4.8 treatments over 2.4 weeks. Chronic patient mean VAS scores dropped from 7.5 to 2.2 within 5.4 treatments over 3.3 weeks. No adverse events were reported.

Conclusion: This study showed clinical improvement in patients with knee pain who were managed with Cox flexion distraction decompression applied to the knee.

#### (JCCA. 2017;61(2):153-161)

KEY WORDS: knee osteoarthritis, Cox Flexion Distraction Decompression, knee pain, manual therapy, manipulation, chiropractic

#### Introduction

The prevalence of knee pain has increased substantially over a 20 year period, independent of age and body mass index (BMI).<sup>1</sup> It affects approximately 30-40% of adults by age 65.<sup>2</sup> Total knee replacement utilization in the United States more than doubled from 1999 to 2008.<sup>3</sup> Studies have shown that increasing physical activity in people with osteoarthritis (OA) reduces pain and depression.<sup>4</sup> If a patient has symptoms of OA, such as knee pain with associated walking disability, he or she may be at increased risk of premature death from cardiovascular disease.<sup>5</sup> Chiropractic treatment for knee OA is typically multimodal, involving manipulation, mobilization, soft tissue techniques, physical therapy modalities, nutritional counseling, exercise, and orthoses for the knee or foot.<sup>6</sup>

Cox flexion distraction decompression (FDD) of the spine is an evidence-based, non-surgical spinal care treatment modality, a form of spinal manipulation in which the human spine is placed in distraction (a type of measured, controlled traction of the spine) delivered on a specialized spinal manipulation instrument (the Cox table).<sup>7</sup> The main difference between Cox FDD and traction is that the treatment is manually applied, according to patient tolerance, with oscillation of the applied forces, all the while maintaining the joint under decompression (i.e. tensile loading plus mechanical stress). This can be performed with or without passive stretch to the specific joint through various ranges of motion (ROM). Its effects on the spine are well researched and documented. Cox FDD

moyens de l'EVA des patients atteints de douleur chronique ont chuté de 7,5 à 2,2 après 5,4 traitements sur 3,3 semaines. Aucun incident indésirable n'a été déclaré.

Conclusion : Cette étude démontre une amélioration clinique chez les patients atteints de douleur au genou qui ont été traités avec la décompression par flexiondistraction de Cox appliquée au genou.

#### (JCCA. 2017;61(2):153-161)

MOTS CLÉS : arthrose du genou, décompression par flexion-distraction de Cox, douleur au genou, thérapie manuelle, manipulation, chiropratique

has demonstrated a reduction of intradiscal pressure of up to –192mmHg in the lumbar spine and –502mmHg in the cervical spine.<sup>8,9</sup> It is a non-invasive joint therapy for patients that allows for continued, reasonable daily activity or minimal convalescence. Recent research has demonstrated that articular cartilage has the intrinsic ability to repair itself when the joint is exposed to distraction with mechanical stimulation.<sup>10,11</sup>

The application of flexion distraction specifically to the knee with and/or without passive knee flexion has not been well documented. The purpose of this study is to introduce the application of Cox FDD as an innovative approach to the treatment of knee pain and OA. This study was designed in retrospect to treatment. Chart review of 25 patients is presented.

#### Methods

A retrospective search of patient files over six months was performed from one chiropractic facility. Ethics approval was obtained by the Research Ethics Board of the Canadian Memorial Chiropractic College. Charts that were included in this study were patients with only knee pain and having received Cox FDD applied to the knee with and/or without passive knee flexion. VAS scores were used to assess treatment effectiveness. Thirty-two charts were selected. Three were excluded on the basis of having less than three treatments, and four charts were excluded for not having final VAS scores, resulting in 25 patient records. There were no exclusions based on diagnosis.


Figure 1. Initial setup.



Figure 2. Flexion Distraction.

The application of treatment followed similar guidelines as used in the Cox Technic spinal protocols.8 The treatment involved the patient seated at the Cox Table with the affected tibiofemoral joint comfortably resting between the lumbar and dorsal sections of the table (Figure 1). The dorsal section of the table was placed at an angle between 0-15° below horizontal for comfort and to decrease hamstring tension. The caudal section was disengaged to allow for flexion. To create knee joint distraction, the chiropractor applied downward forces above the knee and at the superior aspect of the distal tibiofibular joint (Figure 2). The table was distracted to the "taut point" which is reached when the patient's knee is distracted to the point of the barrier of elasticity. This is the starting point of Cox FDD. The chiropractor controlled the amount of distraction of the device using a foot switch, applied according to patient tolerance. The knee was then distracted and brought to flexion and extension as tolerated by the patient in an oscillatory manner that was smooth and rhythmical for a minimum of 10-15 repetitions (Figures 2, 3, and 4). Each repetition lasted 2-4 seconds. Total treatment time with Cox FDD was approximately 1 minute. Most patients received adjunctive (laser treatment) applied to the knee joint after each Cox FDD session was completed. No adverse events were reported.

#### Results

The age range of the study participants was between 20 and 80 years, with an average age of 57.5. Seventeen of the eligible charts were considered as chronic with having pain greater than three months and eight were considered acute. Of those, 11 were female and 6 were male. Fourteen patients (56.0%) had previous knee surgery, all of which were in the chronic knee pain group: five meniscectomies, two total knee arthroplasties and seven of unknown surgical type. The acute group was comprised



Figure 3. *Returning to neutral with distraction.* 



Figure 4. *Back to neutral*.

Group	Average Age (years)	Initial VAS	Post VAS	Average VAS Change	Average No. Treatment	Average Time Span (weeks)
Chronic (n=17)	58.1	7.5	2.2	-5.3	5.4	3.3
Acute (n=8)	49.1	8.1	1.1	-7.0	4.8	2.4
Overall (n=25)	57.5	7.7	1.8	-5.9	5.3	3.0

Table 1.Change in VAS scores for chronic and acute patients.

of two females and six males. None of the acute patients had previous knee surgery.

The reduction in VAS scores for both groups are shown in Table 1. The total average VAS scores for both groups dropped from 7.7 to 1.8 (a reduction of 5.9) in an average of 5.3 treatments over 3.0 weeks. Within the chronic group, the average reduction in VAS scores was from an initial VAS of 7.5 to 2.2 (a decrease of 5.3 points) in 5.4 treatments over 3.3 weeks. The average reduction in VAS within the acute group was from an initial VAS of 8.1 to 1.1 (a reduction of 7.0 points) in 4.8 treatments over 2.4 weeks. There were no patients that reported an increase in pain. One patient did not report any change. None of the acute cases required any treatment three months post-treatment. Most of the chronic cases required ongoing maintenance treatments at a frequency of one treatment per month. In the chronic group, eight out of 17 (47.1%) patients had undergone previous knee surgery ranging from arthroscopic meniscectomy (n = 6) to total knee arthroplasty (n = 2).

The Cox 8 Table allows for real-time force measurements during treatment (Figure 5). Hand pressure, distraction force, table angle and distraction distances are displayed and recorded in real-time as a visual aid. The force applied along the y-axis during treatment was within 20-40 lbs and table distraction distance was within 16-55mm. The treatment flexion angle ranged between  $0^{\circ}$ – 9.1°.



Figure 5. *Cox 8 table forces graph applied to the knee*.

The primary clinical diagnoses of the patients in both groups were OA, collateral and cruciate ligament sprains, meniscus tears and sprains, and post-surgical continued knee pain for partial and total knee arthroplasty. In many of the cases there was a combination of these diagnoses.

#### Discussion

#### Retrospective Case Series Findings

Both the acute and chronic knee pain groups in this case series responded well to treatment. Almost all of the patients reported a decrease in pain as measured on the VAS with Cox FDD. Notably, most patients reported immediate relief after their first treatment, that lasted from several hours up to two days. At last follow-up, almost all patients reported better mobility, knee joint strength and stability. The last follow-up was performed at 1 to 3 months after treatment. No adverse reactions were reported by any of the patients.

The reduction in VAS scores, number of treatments and duration of care was similar for both groups. Follow-up treatment was evaluated for both groups. Thirteen of the chronic cases returned for follow-up treatments within four weeks and continued with maintenance treatment every three to four weeks and/or as needed. In reviewing the acute group, no patients returned for further treatment after three months.

### Previous Studies of Manual Therapy for OA of the Knee

The current study demonstrates a reduction in pain level and treatment duration that are similar with other studies involving manual therapy, exercise and/or surgery. Van den Dolder and Roberts demonstrated a reduction between -8 to -10 mm versus the control group on 100mm VAS.12 Treatment consisted of transverse friction to the lateral retinaculum, patellofemoral stretches, and the application of sustained medial glide during repeated flexion and extension of the knee. Maher et al.<sup>13</sup> showed a statistically significant improvement in ROM but no longterm changes in pain levels using passive knee flexion. Treatment involved the patient laying prone with the knee flexed and the distal femur secured to the table with a stabilization belt and the therapist applied a traction force to the knee. Pollard et al.8 demonstrated a reduction in mean VAS scores from 3.3 to 1.9 in their treatment group. The

intervention group received myofascial mobilization and an impulse thrust procedure. Deyle *et al.*<sup>14,15</sup> demonstrated in two randomised controlled trials, improvement in self-reported pain and function when combining manual therapy with exercise versus exercise alone. The treatment applied was soft tissue mobilization and stretching. Khademi-Kalantari *et al.*<sup>16</sup> demonstrated significant relief of knee pain with sustained knee joint traction. Finally, Dwyer *et al.*<sup>17</sup> demonstrated improvement but no statistical improvement between groups when comparing manual and manipulative therapy (joint mobilization, manipulation, and soft tissue treatment) with and without rehabilitation (monitored and/or home program).

Six weeks of continuous knee joint distraction has also been shown to postpone the need for total knee joint arthroplasty in patients younger than age 65.<sup>18</sup> Furthermore, Van der Woude *et al.*<sup>18</sup> have shown that knee articular cartilage has the potential to regenerate, in some cases doubling in thickness with continuous knee joint distraction. Interestingly, these outcomes were maintained even at 5 year follow-up.<sup>18</sup> This could help to partially explain the clinical results observed in the current study.

#### Knee Physiology

Similarities exist in the physical properties of the knee and spinal joints. The knee meniscus and intervertebral discs are responsible for load transmission, force distribution, shock absorption, and articular cartilage protection.<sup>19</sup> Both structures rely on collagen fibrils to resist tensile forces. Articular cartilage and the extracellular matrix (ECM) are maintained and produced by chondrocytes, specialized cells derived from mesenchymal stem-cells. Chondrocytes produce the cartilage matrix and its components such as proteoglycans (PGs) and glycosaminoglycans (GAGs), that provide the tissue with hydration and its high capacity to withstand compressive loads.20 The most abundant PG in articular cartilage and the ECM is aggrecan which is composed of a core protein, hyaluronan (HA), and several side chains of GAGs. The most abundant GAG side chains are made up of multiple repeating units of chondroitin-sulphates, keratin-sulphates and dermatan-sulphates. The knee meniscus is predominantly a fibrocartilaginous structure reinforced by highly ordered collagen fibers in a complex orientation. Compressive forces in the intervertebral discs are predominately handled by the nucleus pulposus which is also made up of PGs within a loose framework of collagen fibers. Changes to the viscosity of HA causes densification of tissue and can modify the function of fascia, nerve receptors, muscle layers (epimysium and perimysium gliding) and hydrodynamic properties of connective tissue.<sup>21</sup> It is suggested by the authour that the use of Cox FDD to the knee may cause conformational changes to the synovial capsule and fluid, meniscus, articular cartilage, tendons and entheses, due to the decreased pressure induced by treatment similar to that which occurs in the intervertebral discs.

Increased levels of catabolic enzymes that degrade the ECM occur in patients with OA and rheumatoid arthritis (RA).<sup>22</sup> Arachidonic acid metabolites (PGs, leukotrienes, etc.) and cytokine levels are also increased after inflammatory insult from injury, infection, or in degenerative diseases such as OA and RA. These metabolites signal the biosynthesis of specialized pro-resolving mediators (SPMs) from omega-3 essential fatty acids including eicosapentanoic acid and docosahexanoic acid.<sup>23</sup> SPMs resolve inflammation (catabasis) as opposed to non-steroidal anti-inflammatory medications which block certain steps of inflammation. SPMs initiate healing, contain or limit inflammation, prevent and/or reduce the severity of inflammation, and reduce tissue destruction.<sup>24,25</sup> SPMs include lipoxins, resolvins, protectins and maresins and are known to act as potent regulators of neutrophil infiltration, cytokine and chemokine production, and clearance of apoptotic neutrophils by macrophages which promote the return of tissue homeostasis.25 Stretching of connective tissue reduces the migration of neutrophils and increases SPM resolvin concentrations.24

Passive neurodynamic mobilization has been shown to promote nerve function by limiting or altering intraneural fluid accumulation, preventing the adverse effects of intraneural edema.<sup>26</sup> The use of mobilization techniques, such as those used in the current study, may promote healing of the soft tissues by stimulating the functions of the nervous system to improve adaptability and decrease tissue sensitivity, thereby helping to alleviate symptoms.<sup>27,28</sup>

Mechanotransduction is the process by which biomechanical signals (physical forces) regulate or affect cellular activity and behaviour.<sup>29,30</sup> Paluch *et al.*<sup>29</sup> describes it as how cells sense physical forces and translate them into biochemical and biological responses. Biomechanical signals include compression, stretch (decompression or tension), and shear forces. These forces are converted into chemical signals at the cell surface, acting on cell surface adhesion-receptors and calcium ion channels. They are converted into chemical energy at the cell membrane. Integrins and cadherins are transmembrane proteoglycans that channel mechanotransductive forces and stimuli along the cytoskeletal filaments to distant sites within the cytoplasm and nucleus.<sup>22,23</sup> Genetic transcription of chondrocytes increase the production of aggrecan when influenced by mechanotransduction.22,23,30 Mechanotransduction stimulates mesenchymal stem cells, found throughout joint tissues, to differentiate into chondrocytes. Chondrocytes are also mechanosensitive and under joint distraction they produce increased levels of PGs, GAGs, and increase ECM.<sup>29-31</sup> It has been shown that mechanical stimulation of chondrocytes antagonizes interleukin-1ß and tumor necrosis factor- $\alpha$ .<sup>22</sup> Mechanotransduction has also been shown to reduce the levels of ECM degrading enzymes in OA and RA. Cox FDD, as used in the current study, applies tensile and compressive loads to the ligaments, tendons, menisci, articular cartilage and entheses of the knee all while under reduced joint pressure from distraction.

#### Cox Flexion Distraction Decompression

Cox FDD was developed by Dr. James M. Cox, DC, DACBR over 40 years ago<sup>32</sup>. Cox Technic is an evidence based non-surgical, chiropractic spinal manipulation that is applied using the Cox Table, by a certified practitioner. Several National Institute of Health funded studies have demonstrated the effectiveness of the technique in its application to the spine.<sup>9,32</sup> This is the first study to document the treatment on the knee. Cox and Bakkum demonstrated the treatment applied to the hip joint in treating gemelli-obturator internus complex (GOIC).<sup>33</sup> Federally funded research has shown that Cox Technic applied to the spine:

- 1. Decreases intradiscal pressure in the lumbar spine up to -192mmHg.<sup>34</sup>
- 2. Decreases intradiscal pressure in the cervical spine up to -502mmHg.<sup>35</sup>
- 3. Increases intervertebral disc height.<sup>34</sup>
- Increases intervertebral foraminal area up to 28%.<sup>34</sup>

Cramer *et al.*<sup>36</sup> demonstrated that spinal joint fixation leads to degenerative changes of the facet joints. Distraction of the intervertebral disc increases intervertebral disc

height, increases perfusion of nutrients, regenerates the extracellular matrix and reverses degeneration.<sup>37-39</sup> Distraction of the knee can regenerate the articular cartilage and increase the tibiofemoral joint space.<sup>11,12,18</sup> The application of flexion distraction specifically to the knee with and/or without passive knee flexion has not been documented and may yield similar effects as that observed in the spine. The results of this study suggest that Cox FDD may be useful in treating patients with knee pain and OA. This study had also helped to devise a potential protocol for clinical treatment.

In the most recent published guidelines for non-surgical management of the knee, manual therapy was not included. The reason provided was that there was insufficient evidence for inclusion.<sup>40</sup> A similar conclusion was made in a systematic review by French *et al.*<sup>41</sup> The Cox 8 table is capable of recording the forces used, flexion angles and distraction distance for each patient. This information allows for quantifiable, standardized treatment with reproduction of these parameters as well as tracking of any changes, information that may be useful in future investigations

#### Study Limitations

The results of this retrospective study must be gauged with scrutiny, based on the limitations of this study. The decision to apply Cox FDD to the knee was based on necessity and not investigative study. Bias in this study is a major caveat since it was performed in one location by the same practitioner, and patients underwent Cox FDD without a control group. As such, it is unknown if the treatment results in the current study were as a result of the treatment provided or the natural course of the knee pain disorders. Moreover, the only distinguishing characteristics between the different types of knee pain in this study were based on the classification of acute versus chronic knee pain. Specific diagnoses of the source of the knee pain were not part of the inclusion criteria. The groups were not further categorized or compared based on individual clinical diagnosis. The only measure of any changes in pain level was by the use of VAS which is completely subjective. Objective tests and standardized questionnaires such as ROM, WOMAC (Western Ontario McMaster Universities Arthritis Index) and Stair Climb Test (x-step SCT) would have provided more reliable measurable data. In addition, follow-up review for the chronic group in this study was

based on patients returning for care for their knee pain or other conditions. All of the acute cases were contacted by telephone or were interviewed during treatment for other conditions unrelated to the knee by the same clinician. Telephone follow-up does not provide for actual physical observation. As well, it skews the pain level by not utilizing the VAS references for the patient.

All of the patients in this study were also given advice regarding bracing, exercise and nutrition (an anti-inflammatory/gluten free diet) . Unfortunately record-keeping of compliance with these recommendations was not maintained as the patients were not provided or sold any products by the treating practitioner.

Currently, a 20-patient randomized controlled study is underway in collaboration with the University of Windsor Faculty of Human Kinetics which uses ROM, WO-MAC and the Step Test, and patients will be evaluated before and after treatment. The author also applies Cox FDD treatment to the hip, shoulder and ankle in his clinical practice. Investigation of the effects on these joints has not been performed but similar clinical results to the knee have been observed. Future studies are also planned in order to investigate the effects of Cox FDD therapy on knee joint space, specifically the meniscus and articular cartilage, measured via weight-bearing x-ray and MRI.

#### Conclusion

The results of this study suggest that Cox FDD of the knee joint may offer benefit for patients with knee pain and/or OA. The use of the Cox 8 table may allow for more standardized and reproducible treatments. The outcomes of this study nevertheless necessitate further research in the form of larger, prospective observational and/or controlled studies to confirm similar results.

#### Acknowledgments

I thank Dr. James M. Cox, DC, DACBR and Julie Cox-Cid for their encouragement and recommendation to investigate and publish the findings of this study.

#### Funding sources and potential conflict of interest

No funding sources were offered or provided for this study. Dr Luigi Albano is a chiropractor in private practice and is a certified Cox Technic provider.

#### References

- 1. Nguyen US, Zhang Y, Zhu Y, Niu J, Zhang B, Felson DT. Increasing prevalence of knee pain and symptomatic knee osteoarthritis. Ann Intern Med. 2011; 155(11): 725–732.
- 2. van den Dolder PA, Roberts DL. Six sessions of manual therapy increase knee flexion and improve activity in people with anterior knee pain: a randomized controlled trial. Aust J Physiother. 2006; 52: 261-264.
- 3. Losina E, Thornhill TS, Rome BN, Wright J, Katz JN. The dramatic increase in total knee replacement utilization rates in the United States cannot be fully explained by growth in population size and the obesity epidemic. J Bone Joint Surg Am. 2012; 94(3): 201-207.
- 4. Penninx BWJH, Rejeski WJ, Pandya J, Miller ME, Di Bari M, Applegate WB, Pahor M. Exercise and depressive symptoms: a comparison of aerobic and resistance exercise effects on emotional and physical function in older persons with high and low depressive symptomatology. J Gerontol B Psychol Sci Soc Sci. 2002; 57(2): P124-P132.
- Nüesch E, Dieppe P, Reichenbach S, Williams S, Iff S, Jüni P. All cause and disease specific mortality in patients with knee or hip osteoarthritis: population based cohort study. BMJ. 2011;342:d1165
- 6. Law A. Diversified chiropractic management in the treatment of osteoarthritis of the knee: a case report. J Can Chiropr Assoc. 2001; 45(4): 232-240.
- Gudavalli MR, Cox JM, Baker JA, Cramer GD, Patwardhan AG. Intervertebral disc pressure changes during the Flexion-Distraction procedure for low back pain. Abstract from the Proceedings of the International Society for the Study of the Lumbar Spine, Singapore 1998.
- Pollard H, Ward G, Hoskins W, Hardy K. The effect of a manual therapy protocol on osteoarthritic knee pain: a randomised controlled trial. J Can Chiropr Assoc. 2008; 52(4): 229-242.
- Cox JM. Low back pain: mechanism, diagnosis and treatment. Seventh Edition. Lippincott, Williams and Wilkins, 2011: 20-29, 362-383.
- Lafeber F, Intema F, van Roermund P, Marijnissen A. Unloading joints to treat osteoarthritis, including joint distraction. Curr Opin Rheumatol. 2006 18:519-525.
- 11. Wiegant K, van Roermund P, Intema F, Cotofana S, Eckstein F, Mastbergen S, *et al.* Sustained clinical and structural benefit after joint distraction in the treatment of severe knee osteoarthritis. Osteoarthritis Cartilage. 2013 21:1660-1667.
- 12. Van den Dolder PA, Roberts DL. Six sessions of manual therapy increase knee flexion and improve activity in people with anterior knee pain: a randomised controlled trial. Aust J Physiother. 2006;52: 261-264.
- 13. Maher S, Creighton D, Kondratek M, Krauss J, Qu X. The effect of tibio-femoral traction mobilization on passive

knee flexion motion impairment and pain: a case series. J Manipulative Physiol Ther. 2010; 18(1):29-36.

- Deyle GD, Henderson NE, Matekel RL, Ryder MG, Garber MB, Allison SC. Effectiveness of manual physical therapy and exercise in osteoarthritis of the knee. Ann Intern Med. 2000;132:173-180.
- 15. Deyle GD, Allison SC, Matekel RL, Ryder MG, Stang JM, Gohdes DD, *et al.* Physical therapy treatment effectiveness for osteoarthritis of the knee: a randomized comparison of supervised clinical exercise and manual therapy procedures versus a home exercise program. Phys Ther. 2005; 85: 1301–1317.
- 16. Khademi-Kalantari K, Aghdam SM, Baghban AA, Rezayi M, Rahimi A, Naimee S. Effects of nonsurgical joint distraction in the treatment of severe knee osteoarthritis. J Bodywork Movement Ther. 2014; 18(4): 533-539.
- 17. Dwyer L, Parkin-Smith GF, Brantingham JW, Korporaal C, Cassa TK, Globe G, Bonnefin D, Tong V. Manual and manipulative therapy in addition to rehabilitation for osteoarthritis of the knee: assessorblind randomized pilot trial. J Manipulative Physiol Ther. 2015;38(1): 1-21.
- Van der Woude JT, van Heerwaarden RJ, Wiegant K, van Roermund PM, Saris DB, Mastbergen SC, Lafeber FP. Six weeks of knee joint distraction: sufficient for cartilage tissue repair. Ann Rheum Dis. 2015;74:371.
- 19. Beaufils P, Verdonk R. The Meniscus. Heidelberg, Dordrecht. Springer 2010.
- 20. Fox AJS, Bedi A, Rodeo SA. The basic science of human knee menisci. Sports Health. 2012; 4(4): 340-351.
- 21. Stecco A, Gesi M, Stecco C, Stern R. Fascial components of the myofascial pain syndrome. Curr Pain Headache Rep. 2013;17:352.
- 22. Leong DJ, Hardin JA, Cobelli NJ, Sun HB. Mechanotransduction and cartilage integrity. Ann N Y Acad Sci. 2011; 1240:32-37.
- Berrueta L, Muskaj I, Olenich S, Butler T, Badger GJ, Colas RA, Spite M, Serhan CN, Langevin HM. Stretching impacts inflammation resolution in connective tissue. J Cell Physiol. 2016; 231(7): 1621-1627.
- Serhan C. Novel pro-resolving lipid mediators in inflammation are leads for resolution physiology. Nature. 2014 Jun 5; 510(7503): 92-101.
- 25. Duvall MG, Levy BD. DHA- and EPA-derived resolvins, protectins, and maresins in airway inflammation. Eur J Pharmacol. 2016; 785: 144-155.
- 26. Kroeber M, Unglaub F, Guehring T, Nerlich A, Hadi T, Lotz J, Carstens C. Effects of controlled dynamic disc distraction on degenerated intervertebral discs: an in vivo study on the rabbit lumbar spine model. Spine. 2005; 30(2): 181-187.
- 27. Gilbert KK, Smith MP, Sobczak S, James CR, Sizer PS, Brismée JM. Effects of lower limb neurodynamic

mobilization on intraneural fluid dispersion of the fourth lumbar nerve root: an unembalmed cadaveric investigation. J Man Manip Ther. 2015; 23(5): 239-245.

- 28. Jeong UC, Kim CY, Park YH, Hwang-Bo G, Nam CW. The effects of self-mobilization techniques for the sciatic nerves on physical function and health of low back pain patients with lower limb radiating pain. J Phys Ther Sci. 2016; 28(1): 46-50.
- 29. Paluch EK, Nelson CM, Biais N, Fabry B, Moeller J, Pruitt BL, Wollnik C, Kudryasheva G, Rehfeldt F, Federle W. Mechanotransduction: use the forces. BMC Biology. 2015; 13:47.
- Wang N, Tytell JD, Ingber DE. Mechanotransduction at a distance: mechanically coupling the extracellular matrix with the nucleus. Nature Reviews Molecular Cell Biology. 2009; 10(1): 75-82.
- Tiku ML, Sabaawy HE. Cartilage regeneration for treatment of osteoarthritis: a paradigm for nonsurgical intervention. Ther Adv Musculoskel Dis. 2015; 7(3): 76-87.
- 32. Cox JM. Blog #6 What Is Cox® Flexion Distraction Decompression Spinal Manipulation? http://www.coxtechnic.com/blog/ March 9, 2016.
- Cox JM, Bakkum BW. Possible generators of retrotrochanteric gluteal and thigh pain: the gemelliobturator internus complex. J Manipulative Physio Ther. 2005; 28(7): 534-538.
- 34. Gudavalli MR, Cox JM, Baker JA, Cramer GD, Patwardhan AG. Intervertebral disc pressure changes during the flexion-distraction procedure for low back pain. Abstract from the Proceedings of the International Society for the Study of the Lumbar Spine, Singapore 1998.

- 35. Gudavalli MR, Potluri T, Carandang G, Havey RM, Voronov LI, Cox JM, Rowell RM, Kruse RA, Joachim GC, Patwardhan AG, Henderson CNR, Goertz C. Intradiscal pressure changes during manual cervical distraction: a cadaveric study. Evid Based Compl Alt Med. 2013: 2013: 954134.
- 36. Cramer GD, Fournier JT, Henderson CNR, Wolcott CC. Degenerative changes following spinal fixation in a small animal model. J Manipulative Physiol Ther. 2004; 27(3): 141-154.
- 37. Hee HT, Chuah YJ, Tan BHM, Setiobudi T, Wong HK. Vascularization and morphological changes of the endplate after axial compression and distraction of the intervertebral disc. Spine. 2011; 36(7): 505-511.
- Hee, HT; Zhang, JT; Wong, HK. Effects of cyclic dynamic tensile strain on previously compressed inner annulus fibrosus and nucleus pulposus cells of human intervertebral disc-an in-vitro study. Jour Orth Research 28 (4) Apr 2010. p.503-9.
- 39. Guehring T, Omlor GW, Lorenz H, Engelleiter K, Richter W, Carstens C, Kroeber M. Disc distraction shows evidence of regenerative potential in degenerated intervertebral discs as evaluated by protein expression, magnetic resonance imaging, and messenger ribonucleic acid expression analysis. Spine. 2006; 31(15): 1658-1665.
- 40. McAlindon TE, Bannuru RR, Sullivan MC, *et al.* OARSI guidelines for the non-surgical management of knee osteoarthritis. Osteoarthritis Cartilage. 2014; 22(3): 363-388.
- 41. French HP, Brennan A, White B, Cusack T. Manual therapy for osteoarthritis of the hip or knee: a systematic review. Man Ther. 2011; 16(2): 109-117.

## High-grade spondylolytic spondylolisthesis

Peter C. Emary, DC, MSc<sup>1</sup> Stefan A. Eberspaecher, DC<sup>2</sup> John A. Taylor, DC, DACBR<sup>3</sup>

Case reports of high-grade spondylolisthesis have been rarely published in the chiropractic literature. Documented here is a case involving a 28-yearold woman who presented to the World Spine Care clinic in the Dominican Republic with minimal neuromusculoskeletal symptoms despite a grade 4 spondylolytic spondylolisthesis. The key imaging and etiological features of this clinical disorder are presented.

(JCCA. 2017;61(2):162-166)

KEY WORDS: spondylolisthesis, spondylolysis, pars interarticularis, chiropractic

#### **Case Presentation**

A 28-year-old female presented to the World Spine Care (WSC) clinic in Moca, Dominican Republic with a 10-year history of intermittent lower back pain (LBP) and

Il y a eu peu d'études de cas de spondylolisthésis de haut degré publiées dans la littérature chiropratique. On consigne ici un cas concernant une femme de 28 ans qui s'est présentée à la clinique World Spine Care en République dominicaine avec des symptômes neuro-musculo-squelettiques minimaux malgré un spondylolisthésis spondylolytique de stade 4. Les principales caractéristiques d'imagerie et d'étiologie de ce trouble clinique sont présentées.

(JCCA. 2017;61(2):162-166)

MOTS CLÉS : spondylolisthésis, spondylolyse, isthme interarticulaire vertébral, chiropratique

parasthesia into the right lower leg and foot. The pain, which was rated as a six out of 10, was described as a "dull ache" and was generally worse when rising from bed first-thing in the morning. Standing up after prolonged

<sup>1</sup> Private Practice, Cambridge, ON

Corresponding author: Peter C. Emary

Consent: The patient has provided written consent to having her personal health information, including diagnostic images, published.

The authors have no competing interests to declare. © JCCA 2017

<sup>&</sup>lt;sup>2</sup> World Spine Care, Dominican Republic Clinic Supervisor, Patronato Cibao de Rehabilitación, Moca, Provincia Espaillat, República Dominicana

<sup>&</sup>lt;sup>3</sup> Professor and Coordinator of Diagnostic Imaging, Chiropractic Department, D'Youville College, Buffalo, NY, World Spine Care Consultant

<sup>&</sup>lt;sup>1</sup> Private Practice, 201C Preston Parkway, Cambridge, ON N3H 5E8, Canada

e-mail: drpeter@parkwaybackclinic.ca



Figure 1. Lateral lumbar radiograph (a) and midsagittal T2-weighted magnetic resonance (MR) image (b) reveal six lumbar-type vertebrae. The L6 vertebral body is trapezoidal shaped and has translated 80-90% anteriorly in relation to the S1 vertebral segment. A large gap is present in the region of the L6 pars interarticularis and has resulted in posterior translation of the spinous process and laminae of L6. The L6-S1 disc is obliterated and prominent buttressing of the anteriorinferior aspect of the L6 vertebral body is present. The L5-L6 disc appears well hydrated, but is narrowed and bulging posteriorly. The posterior elements of L5 approximate the posterior aspect of the sacral base resulting in prominent central stenosis at this level.

sitting was also provocative. Exercise at the gym, walking, and performing yoga were described as palliative. Recent diagnostic imaging investigations had revealed a high-grade lumbosacral spondylolisthesis (Figures 1-3). The patient had consulted numerous surgeons and other medical specialists and was told that she would be unable to have children because of her spondylolisthesis and would eventually be confined to a wheelchair if she did not have surgery. There were no signs or symptoms of cauda equina syndrome. The patient was in good general health and did not take medications. Because chiropractic treatment had provided relief in the past, she visited the WSC Moca clinic for a second opinion.

On examination, the patient was of normal weight and her gait was unremarkable. The lumbosacral lordosis was increased. Range of motion of the lumbar spine was full and pain-free, with the exception of mild lower back pain



Figure 2. Anteroposterior lumbopelvic radiograph reveals the characteristic "Bowline of Brailsford" or "Inverted Napolean Hat" sign. The arrows are pointing to the anterior cortex of the vertebral body and transverse processes of L6. This vertebral body is tipped forward significantly such that it resembles an axial projection.



Figure 3. Axial (b) T2-weighted MR image through the lumbosacral region illustrates the relationship between the L5 vertebral body (L5), the sacral base (S1), and the spinal canal and thecal sac (oval). The spinal canal is not significantly narrowed at this level. The sagittal MR image on the left (a) indicates the plane of the axial section depicted on the right (b). Note that the L6 vertebral body is translated anteriorly and inferiorly and is outside the plane of the axial image and therefore does not appear on the axial image.



Figure 4. Dynamic lateral lumbar radiographs obtained in partial flexion (a) and full flexion (b). The curved dotted line represents the rounded-off superior surface of the sacral base (the S1 superior vertebral body surface) and the straight dotted line represents the inferior L6 vertebral body endplate surface. The L6 segment translates approximately 5 mm anteriorly and inferiorly in relation to S1 indicative of instability.

on passive end-range extension. Bilateral Kemp's test<sup>1</sup> provoked mild LBP but the straight leg raise, slump, and femoral nerve stretch tests were negative. Neurologic examination of the lower limbs, including motor, reflex, sensory, vibratory, and Rhomberg's testing, was normal except for absent Achilles tendon reflexes (graded as 0) bilaterally. Mid-thigh and calf muscle circumferences were symmetric bilaterally with no evidence of atrophy. A non-tender midline 'step defect' was palpated at the lumbosacral junction and mild pain was elicited with the application of posterior-to-anterior pressure over this region. Palpation also revealed hypertonicity of the quadratus lumborum and lumbosacral erector spinae muscles, bilaterally. Milgram's test<sup>1</sup> (i.e. bilateral active straight leg raise test, performed with the patient supine) severely provoked the chief LBP complaint and elicited patient apprehension. Further radiographic imaging including dynamic views of the lumbar spine were obtained. The flexion radiographs revealed instability of the spondylolisthesis (Figure 4). Based on clinical and imaging findings, a diagnosis of chronic unstable grade 4 spondylolytic spondylolisthesis was made.

#### Discussion

Table 1 lists the radiologic grading and etiological classification systems for spondylolisthesis.<sup>2,3</sup> The key imaging features for high-grade spondylolytic spondylolisthesis are listed in Table 2. According to the Meyerding classification<sup>2</sup>, high-grade spondylolistheses are characterized by an anterior vertebral slippage of 50% or greater (i.e. grades 3, 4, and 5). The incidence of isthmic (spondylolytic) spondylolisthesis in the general adult population ranges between 3.7% and 8%;9 however, the epidemiology of high-grade spondylolisthesis is unknown. To date, the evidence concerning the long-term prognosis and optimal (i.e. conservative versus surgical) management of patients with high-grade spondylolisthesis also remains insufficient and controversial.<sup>4-9</sup> Moreover, clinical symptoms in such patients often do not correlate with the degree of slip.<sup>10</sup> Several authors advocate for surgical intervention (e.g. fusion, with or without slip reduction) regardless of patient symptoms, in order to prevent further slippage and or symptom progression.<sup>7,8</sup> Others have suggested however that non-surgical management can be considered, particularly in asymptomatic or minimally symptomatic cases.<sup>5-8</sup> Regarding natural history, the risk of slip progression is greater in skeletally immature children and adolescents than it is in adults.<sup>8,10</sup> As such surgery is rarely indicated for this reason in adults.8

In the current case, the patient underwent a trial of conservative therapy, including spinal manipulation (to the thoracic and upper lumbar spine), soft-tissue trigger-point therapy and Active Release Techniques<sup>®</sup> (to the lumbosacral and thoracolumbar paraspinal muscles), and homebased spinal stabilization exercises (i.e. cat-camel mobilizations, bird-dogs, side-bridges, and crunches). The patient was also encouraged to continue with her weekly exercise activities (e.g. walking, gym exercise, and yoga). After nine visits (over five weeks) the patient had no functional limitations and her pain severity was reduced to a two out of 10. Furthermore, there were no adverse treatment effects during the course of her care.

Indications for surgery in patients with spondylolisthesis are listed in Table 3. With a greater than 10% reported risk of complications including neurologic injury with surgical intervention<sup>6,8,11</sup>, some authors have indicated that non-surgical treatment may be suitable for patients in the absence of functional limitations or neurologic impairment.<sup>5-8</sup> In the current case, the patient had clinical

Table 1.Radiologic grading and etiologic classification systemsfor spondylolisthesis

Radiologic (Meyerding <sup>2</sup> ) grading system								
<ul> <li>Grade 1 (slippage of 1% to 25%)</li> <li>Grade 2 (slippage of 26% to 50%)</li> <li>Grade 3 (slippage of 51% to 75%)</li> <li>Grade 4 (slippage of 76% to 100%)</li> <li>Grade 5 (slippage of &gt;100%) (i.e. spondyloptosis)</li> </ul>								
Etiologic (Wiltse <sup>3</sup> ) classification								
<ul><li>Type 1: Dys</li><li>Type 2: Isth</li></ul>	plastic ( mic I s a t	Congenital defect of the neural arch Pars interarticularis defect (3 subtypes) a) Stress (fatigue) fracture (most common type, present in this case) b) Elongated pars c) Acute pars fracture						
• Type 3: Deg	generative I	Facet joint osteoarthrosis						
• Type 4: Trai	umatic A	Acute fracture of the vertebral arch other than the pars						
• Type 5: Path	nological I	insufficiency fracture as a result of pone weakening diseases						
• Type 6: Iatro	ogenic I	Fracture secondary to spinal surgery such as laminectomy						

# Table 2.Key imaging features of high-grade spondylolytic(isthmic Type 2a) spondylolisthesis

# Key imaging features Spondylolysis: pars interarticularis stress (fatigue) fracture that usually occurs during childhood or early adolescence Most common at L5 Anterior displacement of the vertebral body and corresponding posterior displacement of the neural arch resulting in characteristic palpable "step defect" (see Figure 1) Bowline of Brailsford on frontal radiograph (see Figure 2) Trapezoidal shape of listhetic vertebral body and corresponding "rounding" of the subjacent sacral base (see Figure 4) (These are signs of chronic, longstanding vertebral body displacement.) Vertebral body translation >4.5 mm observed on dynamic flexion-extension radiographs suggests radiologic instability (see Figure 4)

# Table 3. Indications for surgery in patients with spondylolisthesis

Surgical indications8

- Slip progression
- High-grade spondylolisthesis with significant lumbosacral kyphotic deformity and sagittal imbalance
- Neurologic deficit
- Low back pain unresponsive to conservative treatment
- Radicular pain and nerve root compression documented on
- imaging studies unresponsive to conservative treatment

and radiologic evidence of instability but no significant functional limitations, postural deformities, gait abnormalities, or progressive neurologic signs or symptoms. Her LBP symptoms also responded favourably to a shortcourse of conservative treatment. She was advised to return for chiropractic treatment as needed, or for re-evaluation should any neurologic signs or symptoms develop. As the patient was planning on getting married later in the year, the news that her spondylolisthesis would have no bearing on her ability to have children after all came as a welcome surprise.

#### Key Messages

- Published cases involving high-grade spondylolisthesis are rare within the chiropractic literature
- Indications for surgery in such patients include slip progression, significant deformity and postural imbalance, neurologic deficit and/or cauda equina syndrome, and imaging confirmed radiculopathy unresponsive to conservative treatment
- Conservative therapy may be considered as a first line treatment option in patients with high-grade spondylolisthesis who present with minimal neuromusculoskeletal symptoms

#### References

- 1. Evans RC. Illustrated Essentials in Orthopedic Physical Assessment. St. Louis: Mosby; 1994.
- Meyerding HW. Spondylolisthesis. Surg Gynecol Obstet. 1932; 54:371-377.
- Wiltse LL, Newman PH, Macnab I. Classification of spondylosis and spondylolisthesis. Clin Orthop Relat Res. 1976; 117:23-29.
- DeWald CJ, Vartabedian JE, Rodts MF, Hammerberg KW. Evaluation and management of high-grade spondylolisthesis in adults. Spine. 2005; 30(6 Suppl):S49-S59.
- Bourassa-Moreau É, Mac-Thiong JM, Joncas J, Parent S, Labelle H. Quality of life of patients with high-grade spondylolisthesis: minimum 2-year follow-up after surgical and nonsurgical treatments. Spine J. 2013; 13(7):770-774.
- Lundine KM, Lewis SJ, Al-Aubaidi Z, Alman B, Howard AW. Patient outcomes in the operative and nonoperative management of high-grade spondylolisthesis in children. J Pediatr Orthop. 2014; 34(5):483-489.
- 7. Xue X, Wei X, Li L. Surgical versus nonsurgical

treatment for high-grade spondylolisthesis in children and adolescents: a systematic review and meta-analysis. Medicine. 2016; 95(11):e3070.

- Kasliwal MK, Smith JS, Kanter A, Chen CJ, Mummaneni PV, Hart RA, Shaffrey CI. Management of high-grade spondylolisthesis. Neurosurg Clin N Am. 2013; 24(2):275-291.
- Kreiner DS, Baisden J, Mazanec DJ, Patel RD, Bess RS, Burton D, *et al*. Guideline summary review: an evidencebased clinical guideline for the diagnosis and treatment of adult isthmic spondylolisthesis. Spine J. 2016; 16(12):1478-1485.
- Haun DW, Kettner NW. Spondylolysis and spondylolisthesis: a narrative review of etiology, diagnosis, and conservative management. J Chiropr Med. 2005; 4(4):206-217.
- 11. Fu KM, Smith JS, Polly DW Jr, Perra JH, Sansur CA, Berven SH, *et al*. Morbidity and mortality in the surgical treatment of six hundred five pediatric patients with isthmic or dysplastic spondylolisthesis. Spine. 2011; 36(4):308-312.

# A rare case of Eagle syndrome and diffuse idiopathic skeletal hyperostosis in the cervical spine

Peter C. Emary, DC, MSc<sup>1</sup> Marshall Dornink, DC<sup>2</sup> John A. Taylor, DC, DACBR<sup>3</sup>

Published case studies involving diffuse idiopathic skeletal hyperostosis (DISH) along with symptomatic ossification of the stylohyoid ligament, or "Eagle syndrome," are rare. In this report, we document a case of bilateral Eagle syndrome and advanced DISH in the cervical spine of an 80-year-old man who presented with severe neck stiffness and intermittent asphyxia. The key imaging and clinical features of this disorder are also described.

(JCCA. 2017;61(2):167-170)

KEY WORDS: Eagle syndrome, diffuse idiopathic skeletal hyperostosis, airway obstruction, chiropractic

Les études de cas publiées concernant l'hyperostose squelettique idiopathique diffuse (HSID) avec une ossification symptomatique du ligament stylohyoïdien, ou « syndrome d'Eagle », sont rares. Dans le présent rapport, nous consignons un cas de syndrome d'Eagle bilatéral et de HSID avancée du rachis cervical chez un homme de 80 ans présentant une raideur grave de la nuque et une asphyxie intermittente. Les principales caractéristiques d'imagerie et cliniques de ce trouble sont également présentées.

JCCA. 2017;61(2):167-170)

MOTS CLÉS : syndrome d'Eagle, hyperostose squelettique idiopathique diffuse, obstruction des voies respiratoires, chiropratique

Corresponding author: Peter C. Emary Private Practice, 201C Preston Parkway, Cambridge, ON N3H 5E8, Canada e-mail: drpeter@parkwaybackclinic.ca

Consent: The patient has provided written consent to having his personal health information, including radiographs, published.

The authors have no competing interests to declare. © JCCA 2017

<sup>&</sup>lt;sup>1</sup> Private Practice, Cambridge, ON

<sup>&</sup>lt;sup>2</sup> Private Practice, West Seneca, NY

<sup>&</sup>lt;sup>3</sup> Professor and Coordinator of Diagnostic Imaging, Chiropractic Department, D'Youville College, Buffalo, NY



Figure 1. Anteroposterior open mouth radiograph of the upper cervical spine reveals thick, diffuse ossification of both stylohyoid ligaments (\*).



Figure 2. Lateral cervical spine radiograph. The extensive ossification of the stylohyoid ligaments, identified by asterisks (\*) extends from the skull base to the hyoid bone. A pseudoarticulation between the abnormal ossification and the hyoid bone is evident. Also observed is an 8 mm thick vertically oriented shield of ossification adjacent to the anterior vertebral bodies and disc spaces from C2 through C7 typical of DISH. The intervertebral disc spaces are typically preserved and the facet joints are normal in DISH as illustrated in this case. Of note, there is a 1 cm circular calcified mass located anterior to the C6-7 level most likely representing a benign thyroid cyst.

#### **Case Presentation**

An 80-year-old white male presented to a chiropractic clinic with a 3-year history of insidious and progressing neck stiffness and "cramping," increased difficulty turning his head from side-to-side (e.g. while driving), and a "choking" sensation when flexing his neck. He also complained of a constant pressure in his throat as well as difficulty swallowing his saliva, to the point where he felt like he was "choking on his spit at times." He had no problems with normal breathing, speaking, or swallowing food and he denied any symptoms suggestive of cervical radiculopathy or myelopathy. The only difficulty he reported with eating was an "inability to look down at the plate to see his food." On examination, his cervical spine range of motion was severely limited (by 80-90%) in all directions. Flexing his neck forward beyond five degrees would elicit asphyxia. Bilateral upper limb neurologic examination, including motor, reflex, and sensory testing, was unremarkable. Cervical spine radiographs revealed complete bilateral ossification of the stylohyoid ligaments as well as advanced ossification of the anterior longitudinal ligament (flowing hyperostosis) throughout the cervical and upper thoracic spine, abutting and displacing the prevertebral soft-tissues (Figures 1-3). Based on these findings, the patient was diagnosed with Eagle syndrome and diffuse idiopathic skeletal hyperostosis (DISH).

#### Discussion

Symptomatic elongation of the styloid process and/or ossification of the stylohyoid ligament was first described by Eagle<sup>1,2</sup> in the late 1940s and is known as *Eagle syndrome*. Elongated styloid processes or ossified stylohy-

	Table 1.		
Key imaging and	l clinical features	of Eagle	syndrome

Key imaging features

• Ossification of the stylohyoid ligaments is best seen on lateral cervical and anteroposterior upper cervical radiographs

Clinical features<sup>1-4</sup>: dysphagia (i.e. difficulty swallowing), sensation of a foreign body in the throat, otalgia (i.e. pain radiating to the ear), constant dull nagging ache in the throat, carotodynia (i.e. pain in the distribution of the carotid arteries), temporomandibular joint pain, glossopharyngeal neuralgia, and facial pain or headache



Figure 3. Anteroposterior and oblique cervical spine radiographs also reveal the ossified stylohyoid ligaments. The right stylohyoid ligament appears bipartite with the apparent formation of a central pseudoarticulation dividing it.

oid ligaments can be palpated intraorally along the tonsillar pillars and radiographic imaging usually confirms the diagnosis.<sup>1-3</sup> Anatomically, these bony structures can compress or irritate the surrounding neurovascular soft-tissues in the neck, namely the internal carotid artery and the trigeminal, facial, glossopharyngeal, and vagus nerves. Patients may present with an array of symptoms although many with elongated styloid processes or ossified stylohyoid ligaments will also remain asymptomatic.<sup>3,4</sup> Table 1 lists the key imaging and clinical features for Eagle syndrome and Table 2 lists the key imaging features of DISH. Symptomatic patients with Eagle syndrome can be treated surgically or non-surgically.<sup>3,5,6</sup>

Table 2.Key imaging features of DISH

Key imaging features

- Flowing hyperostosis
- Preservation of disc spaces
- Preservation of facet joints (no arthrosis or ankylosis)
- Displacement or compression of prevertebral soft-tissues and airway

note, however, that elongated styloid processes or ossified stylohyoid ligaments may pose a relative contraindication to thrust manipulation of the cervical spine.<sup>7</sup> For instance, an association between styloid process length and risk of cervical carotid artery dissection has been reported.<sup>8</sup> Surgical treatment for Eagle syndrome typically involves styloid process shortening or resection.<sup>3,5</sup> The etiology of Eagle syndrome is still controversial but it is thought to result from post surgical (a particular)

Chiropractors and other manual therapy providers should

but it is thought to result from post-surgical (e.g. tonsillectomy) or traumatic (e.g. styloid fracture) scarring.<sup>1,3</sup> There is some evidence to suggest that there may also be a correlation between ossification of the stylohyoid ligaments and ligamentous ossification of the cervical spine in patients with DISH.<sup>4</sup> This combination of findings is unique, however, in that very few cases involving both Eagle syndrome and DISH have been described in the literature.<sup>4,9</sup> As such, further investigation is needed in order to validate this association.

In the current case, the patient consulted his primary care physician and no further diagnostic testing or surgical treatment was recommended. A course of conservative care, including neck mobilizations (i.e. cervical flexion-distraction / decompression therapy) and instru-

ment-assisted soft-tissue massage (i.e. Graston® Technique), was implemented. The soft-tissues treated included the cervical paraspinals, scalenes, upper trapezius, and levator scapulae muscles, bilaterally. After seven visits (over 10 weeks), the patient reported a mild decrease in neck pain and stiffness but continued to have symptoms of "choking" and asphyxia, particularly with neck flexion. It is unknown if these symptoms were as a result of Eagle syndrome, DISH, or a combination of both. Several cases of dysphagia and airway obstruction in relation to DISH and/or Eagle syndrome have been reported.9,10 In the current case, an upper G-I fluoroscopic (barium swallow) study could have been performed in order to more definitively ascertain the cause of the patient's oropharyngeal symptoms. Regardless, the clinical findings of neck pain, throat irritation, mild dysphagia, and ossified stylohyoid ligaments were compatible with a diagnosis of Eagle syndrome. For more information and additional examples of Eagle syndrome, visit Radiopaedia.org.<sup>11</sup>

#### Key Messages

- Published cases involving Eagle syndrome and DISH of the cervical spine are rare
- The differential diagnosis of patients who present with dysphagia, a sensation of a foreign body in the throat, pain in the distribution of the carotid arteries, and/or neuralgia involving cranial nerves 5, 7,9, and 10 should include Eagle syndrome
- Patients with this disorder can be treated surgically or non-surgically, however there is a relative contraindication to thrust manipulation of the cervical spine

#### References

- Eagle WW. Elongated styloid process; further observations and a new syndrome. Arch Otolaryngol. 1948; 47(5):630-640.
- 2. Eagle WW. Symptomatic elongated styloid process; report of two cases of styloid process-carotid artery syndrome with operation. Arch Otolaryngol. 1949; 49(5):490-503.
- 3. Fusco DJ, Asteraki S, Spetzler RF. Eagle's syndrome: embryology, anatomy, and clinical management. Acta Neurochir (Wien). 2012; 154(7):1119-1126.
- Guo B, Jaovisidha S, Sartoris DJ, Ryu KN, Berthiaume MJ, Clopton P, Brossman J, Resnick D. Correlation between ossification of the stylohyoid ligament and osteophytes of the cervical spine. J Rheumatol. 1997; 24(8):1575-1581.
- Ceylan A, Köybaşioğlu A, Celenk F, Yilmaz O, Uslu S. Surgical treatment of elongated styloid process: experience of 61 cases. Skull Base. 2008; 18(5):289-295.
- Han MK, Kim DW, Yan JY. Non surgical treatment of Eagle's syndrome – a case report. Korean J Pain. 2013; 26(2):169-172.
- Green BN, Browske LK, Rosenthal CM. Elongated styloid processes and calcified stylohyoid ligaments in a patient with neck pain: implications for manual therapy practice. J Chiropr Med. 2014; 13(2):128-133.
- Raser JM, Mullen MT, Kasner SE, Cucchiara BL, Messé SR. Cervical carotid artery dissection is associated with styloid process length. Neurology. 2011; 77(23):2061-2066.
- 9. Unlu Z, Orguc S, Eskiizmir G, Aslan A, Bayindir P. Elongated styloid process and cervical spondylosis. Clin Med Case Rep. 2008; 1:57-64.
- Verlaan JJ, Boswijk PF, de Ru JA, Dhert WJ, Oner FC. Diffuse idiopathic skeletal hyperostosis of the cervical spine: an underestimated cause of dysphagia and airway obstruction. Spine J. 2011; 11(11):1058-1067.
- 11. Radiopaedia.org. Eagle syndrome. Available from: https://radiopaedia.org/articles/eagle-syndrome [Accessed 28 November 2016].

#### **Book Review**

The Basic Science of Pain. An Illustrated and Clinically Orientated Guide Edited by Philip Peng & Oscar de Leon-Casasola. Philip Peng Education Series, 2016. Electronic book, 183 pages, \$9.99 CAD. ISBN: 978-0-9917509-3-1.

Educating chiropractic students and practicing clinicians on the multidimensional nature of pain can be a difficult task. In this electronic book (available on iPad, iPhone, and Mac), the editors, along with distinguished researchers and clinicians, aimed to present the basic science of pain and its clinical application in a concise and interactive manner. The target readers of this book are trainees in pain-related programs, clinicians who treat pain, and others who are interested in better understanding the multidimensional nature of pain.

The book has four chapters: ascending pathways, descending mechanisms, visceral pain, and brain imaging. The chapters are split into basic science and clinical perspective sections. The basic science sections describe important concepts, such as the difference between nociception and pain. These sections clearly outline the numerous mechanisms and locations where nociception can be facilitated or inhibited and how this may contribute to the pain experience. The clinical perspective sections include topics such as: available analgesics, the impact of placebo and nocebo effects, how psychosocial factors can modulate pain, how common interventions such as yoga and meditation may be helpful, and referred pain mechanisms and clinical importance. Clinical conditions that chiropractors commonly see are discussed, such as low back pain and headache. The clinical perspective sections also nicely highlight emerging basic science research, current uncertainties, and allude to novel pain treatments that may arise in the future.

As this is an electronic book, words can be searched, defined, and easily underlined or highlighted. Notes can also be made, which automatically transfer between devices that contain the book and are connected to the Internet. Further, this book enhances learning through illustrations, interactive content, and videos to supplement the text. This multimedia platform makes complex concepts easily digestible, such as the difference between central sensitization, long-term potentiation, and wind-up. Although this book aims to focus on the basic science of pain, an expansion of the content related to the clinical evaluation of patients experiencing pain may increase practicing clinicians' receptivity to this book. A weakness noted in this book was the occasional lack of flow and consistency in terminology between the chapters and sections with different authors. However, this did not significantly impact the overall take-home messages of the book. Another weakness is that the book can only be obtained through iTunes on devices meeting specific iBooks requirements, limiting accessibility to some potential users.

Overall, this book fulfilled its aim. It is suitable for chiropractic students eager to learn more about pain, as well as chiropractic educators looking to update their knowledge of pain to enhance their teaching. Although this book is not specific to chiropractic practice, it is highly relevant to all practicing chiropractors as it may facilitate a better understanding and treatment of the sensory, emotional, cognitive, and social components of patients' pain. This well-referenced electronic book is a valuable pain resource and is highly recommended, especially given its practicality and low cost.

Peter Stilwell, BKin, DC, MSc Dalhousie University, 5869 University Ave. PO Box 15000 Halifax, NS B3H 4R2 E-mail: peterstilwell@dal.ca Phone: 902-817-2280 © JCCA 2017