



CANADIAN CHIROPRACTIC ASSOCIATION

Chair Dr. Gerald Olin, 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 Bussi eres, DC, FCCS(C), PhD
Faculty of Medicine, McGill University, Montr eal, Qu ebec
D epartement chiropratique, Universit e du Qu ebec   Trois-Rivi eres, Trois-Rivi eres, Qu ebec

Dana J. Lawrence, DC, MMedEd, MA
Parker University, Dallas, Texas

Assistant Editors Gregory N. Kawchuk, DC, PhD
University of Alberta, Edmonton, Alberta

Mohsen Kazemi, RN, DC, MSc, PhD, FRCCSS(C), FCCPOR(C)
Faculty of Clinical Education, Graduate Studies and Research,
Canadian Memorial Chiropractic College, Toronto, Ontario

Brynne Stainsby, BA, DC, FCCS(C)
Canadian Memorial Chiropractic College, Toronto, Ontario

Production Co-ordinator John Doiron

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

TYPESETTING

Thistle Printing
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, 2020

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 publication by the Canadian Chiropractic Association. The JCCA is published 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 64 No 1 ISSN 0008-3194 (Print) and ISSN 1715-6181 (Electronic)

Editorial

- 6 JCCA April 2020 Chiropractic Sciences Special Issue: Inaugural Edition
Brynne E. Stainsby, BA, DC, FCCS(C)

Original Articles

- 7 Lessons learned from cases of rib fractures after manual therapy: a case series to increase patient safety
Daphne To, BSc, DC
Anthony Tibbles, BSc, DC, FCCS(C)
Martha Funabashi, BSc, MSc, PhD
- 16 “I stay in bed, sometimes all day.” A qualitative study exploring lived experiences of persons with disabling low back pain
Sharli-Ann Esson, BSc, MHSc
Pierre Côté, DC, PhD
Robert Weaver, MA, PhD
Ellen Aartun, MSc, PhD
Silvano Mior, DC, FCCS(C), PhD
- 32 Patients with cancer. Is there a role for chiropractic?
Maria Tsampika Laoudikou, MChiro
Peter W. McCarthy, PhD
- 43 Clinical management of benign joint hypermobility syndrome: a case series
Patricia Adele Boudreau, BSc, DC
Igor Steiman, BSc, DC, MSc, FCCS(C)
Silvano Mior, DC, FCCS(C), PhD
- 55 Health-related quality of life and balance confidence among participants in a senior community-based exercise (SWIFT) program compared to age matched controls: a cross-sectional study
Erin Cogle, BHSc, DC
Margaret Black, RN, BScN, MEd, EdD
Sheilah Hogg-Johnson, MMath, PhD
Philip Decina, DC, FCCS(C)
Anthony Tibbles, BSc, DC, FCCS(C)
Silvano A. Mior, DC, PhD, FCCS(C)
- 65 Locus of control in patients with Huntington disease: a cross-sectional study
Brynne E. Stainsby, BA, DC, FCCS(C)
Silvano Mior, DC, FCCS(C), PhD
Mark Guttman, MD, FRCPC
- 76 Superficial venous thrombosis of the upper limb presenting to a chiropractic clinic: a case report
Paul Mastragostino, HBSec.Kin, DC, FCCS(C)
Chadwick Chung, BSc, DC, FCCS(C)

Contents

JCCA Vol 64 No 1 ISSN 0008-3194 (Print) and ISSN 1715-6181 (Electronic)

- 82 Potential publication bias in chiropractic and spinal manipulation research listed on clinicaltrials.gov
Breanne M. Wells, DC, MS
Dana Lawrence, DC, MMedEd, MA
- 88 Tibialis anterior herniation – a rare clinical entity: a case report and review of the literature
Ali Masoumi, BSc, DC, MSc, RCCSS(C) resident
Gerry Ramogida, BSc, DC

Editorial Board

Alan H Adams, DC
Texas Chiropractic College
Pasadena, Texas

Carlo Ammendolia, DC, PhD
University of Toronto

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

Brian Budgell, DC, PhD
CMCC

Jason Busse, DC, PhD
McMaster University

Carol Cancelliere, DC, PhD
Toronto, Ontario

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
New York Chiropractic College
Seneca Falls, NY

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, MEd, 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

Scot Howitt, BA, CK, MSc, DC, FRCCSS(C),
FCCPOR
CMCC

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

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

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

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 A. 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

Mathieu Piché, DC, PhD
UQTR

Katharine Pohlman, DC, PhD
Parker University
Dallas, Tx

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

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

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

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

Carol Ann Weis, DC, MSc
CMCC

Jessica J. Wong, DC, MPH, FCCSC
CMCC

JCCA April 2020 Chiropractic Sciences Special Issue: Inaugural Edition

Brynne E. Stainsby, BA, DC, FCCS(C)¹
Assistant Editor



(JCCA. 2020;64(1):6)

KEY WORDS: chiropractic, clinical

MOTS CLES : chiropratique, clinique

It is my great honour and privilege to present the first Chiropractic Sciences issue of the JCCA. This issue includes practical case reports and important original re-

search papers. I hope the content presented in this issue helps to inform your clinical practice and potentially future research endeavours.

The growth of research and scholarly activity in chiropractic in Canada has been fueled by dedicated researchers, Chiropractic Sciences Fellows, faculty members, residents, and students. I would like to thank Dr. Kent Stuber for his support of this initiative, and his support in creating this issue. I would also like to thank all of the contributing authors and peer reviewers who have helped make the JCCA Chiropractic Sciences issue possible.

As I write this editorial, the world has never felt more uncertain. In saying that, this issue brings me hope - I take comfort in knowledge, trust in the science and am grateful to the great professionals who are working so hard to advance health care for our patients and all Canadians.

I encourage you to get involved in research. Be inquisitive and ask questions. If you have an interesting case, set of data or research ideas or questions that you would like to further investigate and need any help, please do not hesitate to contact me, one of the JCCA's Editorial Board members, or a member of the College of Chiropractic Sciences (Canada).

¹ Canadian Memorial Chiropractic College

Corresponding author: Brynne E. Stainsby, 6100 Leslie Street, Toronto, ON M2H 3J1

E-mail: bstainsby@cmcc.ca

Tel: 416-482-2340

© JCCA 2020

The author has no disclaimers, competing interests, or sources of support or funding to report in the preparation of this manuscript.

Lessons learned from cases of rib fractures after manual therapy: a case series to increase patient safety

Daphne To, BSc, DC¹

Anthony Tibbles, BSc, DC, FCCS(C)¹

Martha Funabashi, BSc, MSc, PhD¹

Objective: To identify commonalities among cases of rib fractures after spinal manipulative therapy (SMT); discuss chiropractors' case management perspectives; and propose strategies for prevention and/or management of future cases.

Methods: Semi-structured interviews were conducted with chiropractors who identified cases of rib fractures after SMT at a chiropractic institution's teaching clinics. Patient characteristics, incident characteristics, and chiropractors' perspectives were collected and analysed.

Results: Three chiropractors were interviewed, each identifying one case. Patient ages ranged from 57-77; two were female; two had osteopenia; two cases involved thoracic SMT; and one involved lumbar SMT. Chiropractors agreed that verifying and updating potential contributing factors for rib fractures,

Objectif : Établir les points communs entre des cas de fractures des côtes après des manipulations vertébrales (MV); examiner des points de vue de chiropraticiens sur la prise en charge de cas; proposer des stratégies de prévention et/ou de prise en charge des cas à venir.

Méthodologie : On a fait des entrevues semi-structurées avec des chiropraticiens travaillant à la clinique d'un établissement d'enseignement de la chiropratique et ayant identifié des cas de fractures de côtes après des MV. Les caractéristiques des patients, les caractéristiques des incidents et les points de vue des chiropraticiens ont été recueillis et analysés.

Résultats : Trois chiropraticiens ont été interrogés, chacun ayant identifié un cas. Les patients étaient âgés de 57 à 77 ans; deux étaient de sexe féminin; deux souffraient d'ostéopénie; deux cas avaient été traités par manipulations thoraciques et un cas par manipulations lombaires. Les chiropraticiens ont convenu qu'il était important de vérifier et de mettre à jour les facteurs contributifs potentiels de fractures des côtes, d'informer le patient, en toute transparence, avant

¹ Canadian Memorial Chiropractic College

Corresponding author: Daphne To, Canadian Memorial Chiropractic College, 6100 Leslie Street, Toronto, Ontario M2H 3J1, Canada

E-mail address: dto@cmcc.ca

© JCCA 2020

The authors have no disclaimers, competing interests, or sources of support or funding to report in the preparation of this manuscript. The involved patients provided consent for case publication.

transparent communication prior to SMT and/or after the adverse event (AE) occurrence, and enhancing student education on AE management were important.

Conclusion: Important lessons can be learned from AEs, despite their infrequent occurrences. A more open and constructive patient safety environment is needed within the chiropractic profession.

(JCCA. 2020;64(1):7-15)

KEY WORDS: adverse events, chiropractic, patient safety, quality improvement, spinal manipulative therapy

Introduction

Patient safety continues to be a leading global health care challenge.^{1,2} The World Health Organization (WHO) defines patient safety as the absence of preventable harm to a patient during the process of health care³ and prioritises the safety of every patient in order to provide high quality health services².

The Institute of Medicine's report "To Err is Human: Building a Safer Health System"⁴ and the Government of Canada's "Building a Safer System: A National Integrated Strategy for Improving Patient Safety in Canadian Health Care"⁵ have emphasised the importance of creating an open and constructive patient safety environment in order to develop strategies to reduce preventable adverse events (AEs). It promotes shifting from a blaming culture to a safety culture that learns from AEs, in order to maximise the potential to avoid future AEs.⁶ While strategies for prevention and improving the quality and safety of health care delivery have been shown to improve patient safety in hospital settings⁷ and in family physician practices^{8,9}, little has been reported within the chiropractic profession.

Manual therapy, which includes spinal manipulative therapy (SMT), is used by various health care providers, including chiropractors. Spinal manipulative therapy is commonly used to treat several musculoskeletal conditions and has been recommended by clinical practice guidelines for the management of spinal pain.¹⁰⁻¹⁴ It is estimated that up to 50% of patients who receive manual

d'effectuer des MV et après la survenue d'un événement (ÉI) et d'améliorer la formation des étudiants sur la prise en charge des ÉI.

Conclusion : Les ÉI, bien qu'ils soient rares, peuvent nous permettre de tirer d'importantes leçons. Une attitude plus ouverte et plus constructive envers la sécurité du patient s'impose chez les chiropraticiens.

(JCCA. 2020;64(1):7-15)

MOTS CLÉS : événements indésirables, chiropratique, sécurité du patient, amélioration de la qualité, manipulation vertébrale

therapy experience some form of AE.¹⁵⁻¹⁷ Although most AEs experienced after manual therapy are mild and transient¹⁶, some AEs may have a greater impact on a patient's well-being, function, and quality of life¹⁸.

Rib fractures are often identified as a risk to SMT treatment on clinical consent forms¹⁹; however, to our knowledge, there are limited reports of rib fractures following SMT within the scientific literature. Although most rib fractures are generally benign, heal on their own, and can be managed with conservative therapy, there is the potential risk of serious complications, such as hemothorax or pneumothorax, that can have a substantial impact on patients' morbidity and mortality.²⁰ Since SMT has been clinically perceived as a risk for rib fractures, it is important to explore the occurrences of rib fractures after SMT in more detail. By better understanding the characteristics of the rib fractures after SMT, prevention and mitigation strategies can potentially be developed to increase the safety of this popular intervention.

Therefore, the objectives of this study were to: (1) identify commonalities among cases of rib fractures after SMT; (2) discuss chiropractors' perspectives in case management; and (3) propose strategies for prevention and/or mitigation of future cases. Specifically, our case series will provide an overview of cases of rib fractures after SMT and propose prevention and mitigation strategies. This can contribute to the development of strategies to reduce the occurrences of rib fractures after SMT, contributing to enhancing SMT safety.

Methods

This study was a case series involving supervising chiropractors at the teaching clinics of a chiropractic institution, exploring their experiences and perspectives gained from cases of rib fractures observed after SMT.

Participants

All chiropractors involved in a supervisory role at the Canadian Memorial Chiropractic College (CMCC) teaching clinics were invited to identify eligible cases and to participate in this study. Supervising chiropractors at clinics located within the institution as well as those located at externally hosted institutions were invited to participate. Cases were eligible for this study if the following inclusion criteria were met: the supervising chiropractor volunteered to participate in the study; SMT was provided at the teaching clinic at the time of the rib fracture diagnosis; diagnostic imaging (based on a radiologist's report) was used to confirm the diagnosis of a rib fracture; and cases occurred within the last seven years. All participating chiropractors signed a written informed consent. All patients of CMCC's teaching clinics provided written informed consent for the use of their information for research purposes. This study was approved by the research ethics board at CMCC (1905B01).

Data collection

Participating chiropractors who volunteered to participate in the study were asked to review the electronic medical record (EMR) (IndiviCare, Indivica Inc., Toronto, Canada) of the patient they identified as having experienced a diagnosed rib fracture following SMT. A pre-defined standardised data collection form was used to collect data on patient characteristics and incident characteristics. Variables included in the data collection form were consistent with variables used in a previous study investigating AEs following SMT (SafetyNET)²¹ and variables used in fracture risk prediction tools²². Specifically, variables related to patient characteristics included: patient demographics (including age, sex, weight, body mass index, bone mineral density, physical activity level, co-morbidities, use of medications and supplements, and potential red flags for fractures), and the diagnosis for which the patient was receiving chiropractic treatment for. Variables on incident characteristics included: 1) plan of management (including type and location of SMT, frequency and duration of

care, other treatment modalities); 2) details of the rib fracture (including imaging modality and results, level and location of fracture, time to onset of symptoms, time to diagnosis on imaging, and patient description of event); and 3) rib fracture resolution (including healing time, complications, and return to treatment).

After case review and completion of the standardised data collection form, semi-structured interviews were then conducted with the participating chiropractors. The principal investigator followed a list of pre-determined open-ended questions which included questions on the chiropractors' perceptions of potential contributing factors, their suggestions for prevention of future cases, recommendation/advice to their colleagues, and how the event may have changed their practice. Data on patient and incident characteristics that were previously identified through the data collection form were used to enhance the interview and allow for further details and discussion. The semi-structured interviews were conducted in person by the principal investigator in a quiet room at the chiropractors' offices and lasted approximately 30 to 60 minutes. Details of the interviews were recorded with written notes.

Data analysis

Information from the standardised data collection forms and semi-structured interviews were transferred to a spreadsheet on Microsoft Excel (Microsoft, Redmond, USA). Commonalities between the three cases were identified through visual inspection and interpretation of the data in the spreadsheet by the principal investigator. The data were reviewed by a second investigator and any disagreements were resolved through discussion. This case series followed the CARE Guidelines for clinical case reporting where possible.²³

Results

A total of four chiropractors, with an average of 23.5 years of practice, identified rib fracture cases and volunteered to participate in this study: three of them identified one patient case meeting the inclusion criteria and one chiropractor identified two cases. Due to differences in institutional policies for research use of clinical data at an externally hosted institution, two cases were excluded. Patient characteristics of the three cases are described in Table 1. Incident characteristics of the three cases are described in Table 2.

Table 1.
Patient characteristics. BMI (body mass index); BMD (bone mineral density);
N/A (not applicable, due to unavailable data)

	Age (years)	Sex	BMI (kg/m ²)	Activity level	Smoking	Alcohol	BMD (T-score)	
							Lumbar spine	Femoral neck
Case 1	77	Female	N/A	Low	No	No	-2.1	-2.3
Case 2	60	Female	21.2	Low	Yes	No	-1.5	-2.4
Case 3	57	Male	25.1	Moderate	No	No	N/A	N/A

Case 1:

Case one is of a 77-year-old female with a sedentary lifestyle. Bone mineral density (BMD) T-scores in the lumbar spine and femoral neck were indicative of low BMD (osteopenia).^{24,25} She did not smoke or consume alcohol. She was on medications for the management of hypertension, hyperlipidemia, and hypothyroidism. She had a history of a motor vehicle collision with multiple fractures. She was being treated with chiropractic care, including SMT, for non-specific spinal pain in the cervical, thoracic, and lumbar spines for several years with no previous reports of AEs.

Her treatment plan included multimodal therapy including SMT, spinal mobilisations, and soft tissue therapy to the cervical, thoracic, and lumbar spines. She received SMT targeted at T3-T6 in the supine position with a bilateral posterior contact.²⁶ She reported hearing a loud

“pop” and felt immediate pain on her left side. She did not return to chiropractic treatment for the following three weeks due to scheduling conflicts; however, she reported constant pain at the left lateral chest wall over that three-week period, as well as pain with breathing and sleeping on her left side.

When she returned to the chiropractic clinic after three weeks, a physical examination was conducted, including rib springing and sternal compression, which reproduced mild pain. Vibration testing over the ribs was inconclusive. The patient was referred for an x-ray, which demonstrated a recent rib fracture at the left 5th and 6th ribs in the axillary region. Treatment was modified to exclude SMT to the thoracic and lumbar spines. No complications from the rib fractures were reported. The patient reported symptom resolution in seven weeks and continued to re-

Table 2.
Incident characteristics. SMT (spinal manipulative therapy); N/A (not applicable, due to unavailable data)

	SMT			Symptom onset	Fracture location	Complications	Time to symptom resolution
	Type	Side	Level				
Case 1	Supine; posterior contact	Bilateral	T3-T6	Immediate	Ribs 5 and 6; left side, axillary region	None	7 weeks
Case 2	Prone; hypothenar transverse contact	Left	C7-T1	Immediate	Ribs 4 and 5; left side, anterolateral region	None	12 weeks
Case 3	Side posture; lumbar roll	Left	L3-L5	Immediate	Rib 9; left side, anterior region	None	N/A

ceive chiropractic care. She perceived the experience of the rib fracture as mild.

Case 2

The second case is of a 60-year-old female with a sedentary lifestyle. Bone mineral density T-scores in the lumbar spine and femoral neck were indicative of low BMD (osteopenia).^{24,25} She was a daily smoker and did not consume alcohol. She was on medications for the management of hypertension, depression, pain, and osteopenia (including anti-resorptive therapy, calcium, and vitamin D). She had a history of previous falls with fractures. She was being treated with chiropractic care for non-specific spinal pain in the cervical, thoracic, and lumbar spines for several years with no previous reports of AEs.

Her treatment plan included multimodal therapy including mobilisations and soft tissue therapy to the cervical, thoracic, and lumbar spines. On the day of the rib fracture incident, she was not treated by her usual chiropractic intern and an unintentional error was made where she received SMT targeted at C7-T1 in a prone position using a left unilateral hypothelar transverse contact.²⁶ She reported immediate pain on her left side over the ribs around the axillary region.

A physical examination was conducted immediately after she reported the described pain. Percussion, vibration, and palpation over the left 6th, 7th, and 8th ribs reproduced the chief complaint. Rib springing and thoracic spine ranges of motion produced vague pain over the lower left ribs. She was referred for an x-ray of the ribs, which was taken one week later. The x-ray demonstrated a healing rib fracture at the left 4th and 5th ribs in the anterolateral aspect of the ribs. She reported pain with coughing, sleeping on her left side, and moving from a supine or side-lying position to an upright position.

Treatment was modified to include soft tissue therapy to the intercostal muscles and low-level laser therapy over the affected ribs. As SMT was not part of the patient's original treatment plan, the supervising chiropractor reinforced the importance of easily accessing this information in the patient's file to potentially prevent future unintentional errors. No complications from the rib fractures were reported. The patient reported symptom resolution in 12 weeks and continued to receive chiropractic care. She perceived the experience of the rib fracture as mild.

Case 3:

The third case is of a 57-year-old male who was regularly engaged in moderate level physical activity. Bone-mineral density scores were not available. He did not smoke or consume alcohol. He was not taking any medications for the management of any health conditions. He had a history of a traumatic fall with multiple fractures. He was being treated with chiropractic care for non-specific low back pain; he had received five treatments according to the current treatment plan with no previous reports of AEs.

His treatment plan included multimodal therapy including SMT and soft tissue therapy to the lumbar spine. He received SMT targeted at L3-L5 on the left in the side posture position.²⁶ He reported feeling immediate sharp pain over his left ribs and pain with breathing.

The patient was immediately referred for x-ray, which demonstrated a non-displaced anterior rib fracture of the left 9th rib. He was advised to apply ice by the chiropractor. He was contacted over the phone nine days later and reported improvement in pain. No complications from the rib fracture were reported. The patient was lost to follow-up; therefore, data on symptom resolution and patient perception of the rib fracture could not be recorded.

Chiropractors' perspectives on lessons learned

Three main themes emerged from the semi-structured interviews with the chiropractors who participated in the study: 1) verifying and updating potential contributing factors associated with rib fractures; 2) transparent communication prior to SMT and/or after the occurrence of an AE; and 3) the opportunity for enhancing student education on AE management.

Verifying and updating potential contributing factors associated with rib fractures

In patients with identified risk factors for osteopenia or osteoporosis who may be at an increased risk of fracture (e.g. sex, age, sedentary lifestyle, smoking, alcohol intake, prolonged use of glucocorticoid medication), the chiropractors in this study thought that it was not only important to identify those risk factors at the initial assessment, but also to continuously verify and update them in order to continually choose treatment options to mitigate risk to the patient. Additionally, some chiropractors in this study emphasised that osteopenia and osteoporosis are

relative, not absolute, contraindications to SMT.²⁷ Lastly, the chiropractors in this study suggested that modifications to SMT, such as using non-thrust interventions, may be appropriate in patients who may be at risk for or who have been diagnosed with osteopenia or osteoporosis.

Transparent patient communication

Open and transparent communication with the patient, both prior to providing SMT and/or after the occurrence of an AE, was also identified as an important strategy by the participating chiropractors. Prior to SMT, the chiropractors in this study emphasised the importance of informed consent as a process where the treatment's benefits, risks, and alternatives should be discussed with the patient so that the patient can have an active and informed involvement in the decision-making process. After the occurrence of an AE, participating chiropractors believed that it was important to understand the patient's perception of the event, as the patient's perception may not be the same as the chiropractor's perception.

Opportunity to enhance student education on AE management

As all of the participating chiropractors in this study are involved in a supervisory role at a teaching clinic, they perceived these experiences as an opportunity to enhance students' education on patient safety and AEs. Some of the chiropractors in this study believed that it is important to introduce the concept of patient safety and AEs early in the chiropractic curriculum in order to expose students to an environment where they feel comfortable and supported to talk about safety concerns and AEs, thus promoting an open and trusting patient safety culture focused on learning from AEs instead of blaming.

Discussion

This case series provided an overview of three cases of rib fractures after SMT. In two of the cases, the patients were over 60 years of age, female, had BMD T-scores in the osteopenic range, and were sedentary. In one case, the patient was under the age of 60, male, had unknown BMD T-scores, and was moderately active. In all three cases, the patients were treated with SMT, however the type and location of SMT were different in all cases. In all three cases, the patient felt immediate pain and continued to report aggravation of symptoms with sleeping on the

affected side and with breathing. There were no known complications in any of the cases.

The limited availability of scientific evidence regarding rib fractures following SMT precludes the comparison of our findings to those previously reported in the literature. Two of the cases in this study, however, described characteristics that are similar to the risk factors included in fracture risk prediction tools commonly used to evaluate fracture risk of patients.²⁸ These factors include age, previous fracture, smoking, and low BMD.

Verifying and updating potential contributing factors associated with rib fractures

In 2000, osteoporosis resulted in more than 9.0 million fractures annually worldwide, contributing to the growing global health burden associated with low BMD.^{29,30} Fracture risk assessment tools for low BMD patients have been developed to include risk factors such as sex, age, history of fracture, prolonged glucocorticoid use, rheumatoid arthritis, cigarette smoking, and alcohol intake.²² As patients presenting to chiropractors may return over time for the management of their condition, it is important to always have the most updated information about a patient's overall health, including information on the patient's most recent BMD examination results. For patients with diagnosed osteopenia or osteoporosis, some chiropractors in this study emphasised the importance of making this diagnosis clearly visible and accessible in the patient's file. In doing so, any provider providing care to the patient can easily identify the presence of osteopenia or osteoporosis, which may affect the patient's treatment plan.

According to the WHO, osteopenia and osteoporosis are relative contraindications to SMT.²⁷ The WHO defines a relative contraindication as "*one where the treatment may place the patient at undue risk unless the presence of the relative contraindication is understood and treatment is modified so that the patient is not at undue risk*".²⁷ The chiropractors in this study felt that students training in SMT should understand what a relative contraindication means for the management of their patient. Students should be able to effectively communicate and discuss relative risks to their patients during the informed consent process by presenting the treatment's potential benefits, risks, and alternatives, allowing them to actively engage in a process of informed and shared decision making.

Although previous studies described that during SMT^{31,32}, forces are applied and transmitted through the patient, no studies have quantified forces applied to the thoracic or lumbar spines in a clinical setting. Additionally, there are no studies quantifying the SMT force-time characteristics required to fracture ribs of varying bone mineral densities. Regardless of the applied SMT force-time characteristics, however, evidence suggests that both thrust (SMT) and non-thrust (spinal mobilisation) interventions may lead to reductions in pain and improvements in function outcomes in individuals with chronic neck and low back pain.^{33,34} As such, treatment modifications including non-thrust interventions instead of thrust interventions was suggested by participating chiropractors for patients with identified osteopenia or osteoporosis, or in those who may be at risk, in order to reduce the occurrences of rib fractures.

Transparent patient communication

Chiropractors in this study also emphasised the importance of the informed consent process prior to SMT. Specifically, it has been described that informed consent should be an ongoing process, and that it is perceived by patients as such.³⁵ Similar to what was suggested in the previous theme (*Verifying and updating potential contributing factors associated with rib fractures*), patients who present with potential contributing factors associated with any AE should receive all relevant information in order to make an informed decision by weighing the risks of the treatment to its potential benefits. Nevertheless, participating chiropractors thought that all patients, even those without apparent contributing factors, should be appropriately informed about treatment risks. Additionally, as part of the informed consent process, alternative treatment options should also be explained to the patient. Treatment options could include not only alternative manual therapy techniques (e.g. spinal mobilisation instead of SMT), but also referrals for collaborative, interdisciplinary approaches to management based on the needs of the patient (e.g. co-management with physicians or pharmacists for management with medication; with other rehabilitative professionals for fall prevention; and/or with nutritionists for management of diet). Lastly, based on the case in which a rib fracture occurred after lumbar SMT, the chiropractor emphasised the importance of including rib fracture as a potential risk in the informed consent process

when providing manual therapy to both the thoracic and lumbar spines.

Patient perception of the event after the occurrence of an AE was highlighted as an important consideration. While some patients may perceive the event as very serious, others may perceive it as mild. In this study, patients involved in two of the three cases were very understanding of the situation, were not upset or angry, perceived the rib fracture as a mild AE, and subsequently returned to chiropractic care. In the third case, data on the patient perception was not available. After an AE, the chiropractors in the study stressed the importance of communicating with the patient about the events that occurred, what the patient should expect to feel, and potential complications, as this communication demonstrates accountability and professionalism.

Opportunity to enhance student education on AE management

In order to create a culture of learning from AEs when they occur (as opposed to a blaming/shaming culture), curricular changes may be needed. Specifically in the chiropractic curriculum, general concepts around patient safety and AEs (e.g. epidemiology, use of safety checklists) could potentially be implemented early in the curriculum. This could then be followed by practising cases and using simulation training, which would allow for students to identify potential contributing factors associated with particular AEs, practise different manual therapy techniques in addition to SMT, and practise the informed consent process in order to facilitate shared decision making. Indeed, structured educational programs including didactic and practice-based learning have been used in medical residency programs to educate medical residents on patient safety and quality of care.³⁶ Furthermore, simulation training for acute care nurses has been demonstrated to achieve improved patient safety outcomes.³⁷ By enhancing education on AEs and patient safety, chiropractic institutions have the potential to teach the next generation of chiropractors to be comfortable with talking about patient safety, which would significantly advance patient safety within the chiropractic profession.

Limitations

Not all cases of rib fractures that occurred after SMT may have been included in this study, as participating chiro-

practitioners had to actively volunteer for this study. In addition, details surrounding the patient and incident characteristics were based on previously documented clinical notes and memory recall. Therefore, there is the potential for unclear documentation, missing data, and memory decay, with no method for verifying the information. It is important to note that this study was not designed to establish risk factors associated with rib fractures or AEs, nor was it designed to establish causality of observed AEs. Lastly, no standardised qualitative technique was used for inquiry or to analyse the data and the perspectives of interns and patients were not collected. Future studies should use a systematic qualitative technique to identify themes or develop a taxonomy on lessons learned and risk mitigation strategies.

Future research

Future studies are needed to establish the SMT force-time characteristics necessary to cause a rib fracture in patients with varying characteristics, including varying BMD levels. A standardised method for systematically collecting AE data is also needed so that potential risk factors can be identified, significantly contributing to advancing patient safety related to SMT. Patients' and providers' expectations and perceptions towards AEs should also be further explored to expand on risk prevention and mitigation strategies.

Conclusion

This case series reviewed chiropractors' perspectives on cases of rib fractures after SMT, including their thoughts on potential contributing factors based on patient and incident characteristics, as well as their suggestions on enhancing patient safety and developing prevention and mitigation strategies. The chiropractors in this study stressed the importance of verifying and updating potential contributing factors that may be associated with rib fractures over the course of treatments, as well as open and honest communication with the patient as suggested prevention and mitigation strategies. They also viewed their experience in managing AEs as an opportunity to enhance student education in order to improve the overall patient safety culture. Our study indicates that important lessons can be learned from AEs, despite their infrequent occurrences. As patient safety is a global healthcare challenge, chiropractors need to be leaders in creating an open

and constructive patient safety environment within their profession.

References

1. Brasaitte I, Kaunonen M, Suominen T. Healthcare professionals' knowledge, attitudes and skills regarding patient safety: a systematic literature review. *Scand J Caring Sci.* 2015;29(1):30-50.
2. World Health Organization. *Patient Safety: Making health care safer.* Geneva:2017.
3. World Health Organization. *Patient safety.* 2019; <https://www.who.int/patientsafety/en/>. Accessed September 1, 2019.
4. Kohn L, Corrigan J, Donaldson M. *To err is human: Building a safer health system.* Washington, D.C.: National Academy Press; 2000.
5. Wade J. *Building a Safer System : A National Integrated Strategy for Improving Patient Safety in Canadian Health Care.* The National Steering Committee on Patient Safety;2002.
6. Rafter N, Hickey A, Condell S, et al. Adverse events in healthcare: learning from mistakes. *QJM.* 2015;108(4):273-277.
7. Anderson JE, Kodate N, Walters R, Dodds A. Can incident reporting improve safety? Healthcare practitioners' views of the effectiveness of incident reporting. *Intl J Qual Health Care.* 2013;25(2).
8. Howell AM, Burns EM, Bouras G, Donaldson LJ, Athanasiou T, Darzi A. Can patient safety incident reports be used to compare hospital safety? Results from a quantitative analysis of the English National Reporting and Learning System Data. *PLoS One.* 2015;10(12): e0144107.
9. Bhasale AL, Miller GC, Reid SE, Britt HC. Analysing potential harm in Australian general practice: an incident-monitoring study. *Med J Austr.* 1998;169(2).
10. Beliveau PJH, Wong JJ, Sutton DA, et al. The chiropractic profession: a scoping review of utilization rates, reasons for seeking care, patient profiles, and care provided. *Chiropr Man Therap.* 2017;25: 35.
11. Mior S, Wong J, Sutton D, et al. Understanding patient profiles and characteristics of current chiropractic practice: a cross-sectional Ontario Chiropractic Observation and Analysis Study (O-COAST). *BMJ Open.* 2019;9(8): e029851.
12. French SD, Charity MJ, Forsdike K, et al. Chiropractic Observation and Analysis Study (COAST): providing an understanding of current chiropractic practice. *Med J Aust.* 2013;199(10): 687-691.
13. Bussieres AE, Stewart G, Al-Zoubi F, et al. Spinal manipulative therapy and other conservative treatments for low back pain: a guideline from the Canadian Chiropractic Guideline Initiative. *J Manipulative Physiol Ther.* 2018;41(4): 265-293.

14. Côté P, Wong JJ, Sutton D, et al. Management of neck pain and associated disorders: A clinical practice guideline from the Ontario Protocol for Traffic Injury Management (OPTIMa) Collaboration. *Eur Spine J.* 2016;25(7): 2000-2022.
15. Carnes D, Mars TS, Mullinger B, Froud R, Underwood M. Adverse events and manual therapy: a systematic review. *Man Ther.* 2010;15(4): 355-363.
16. Swait G, Finch R. What are the risks of manual treatment of the spine? A scoping review for clinicians. *Chiropr Man Therap.* 2017;25: 37.
17. Walker BF, Hebert JJ, Stomski NJ, et al. Outcomes of usual chiropractic. The OUCH randomized controlled trial of adverse events. *Spine.* 2013;38(20): 1723-1729.
18. Carlesso LC, Cairney J, Dolovich L, Hoogenes J. Defining adverse events in manual therapy: an exploratory qualitative analysis of the patient perspective. *Man Ther.* 2011;16(5): 440-446.
19. Canadian Chiropractic Protective Association. Consent to Chiropractic Treatment. September 2015.
20. Talbot BS, Gange CPJ, Chaturvedi A, Klionsky N, Hobbs SK, Chaturvedi A. Traumatic rib Injury: patterns, imaging pitfalls, complications, and treatment. *Radiograph.* 2017;37(2): 628-651.
21. Pohlman KA, O'Beirne M, Thiel H, et al. Development and validation of providers' and patients' measurement instruments to evaluate adverse events after spinal manipulation therapy. *Eur J Integr Med.* 2014;6(4): 451-466.
22. Compston JE, McClung MR, Leslie WD. Osteoporosis. *Lancet.* 2019;393(10169): 364-376.
23. Gagnier JJ, Kienle G, Altman DG, et al. The CARE Guidelines: Consensus-based clinical case reporting guideline development. *Glob Adv Health Med.* 2013;2(5): 38-43.
24. World Health Organization. Assessment of fracture risk and its application to screening for postmenopausal osteoporosis: report of a WHO study group. Geneva 1994.
25. Lewiecki EM, Watts NB, McClung MR, et al. Official positions of the international society for clinical densitometry. *J Clin Endocrinol Metab.* 2004;89(8):3651-3655.
26. Bergmann TF, Peterson DH. Chiropractic technique: Principles and procedures. 3rd ed: Mosby 2010.
27. World Health Organization. WHO guidelines on basic training and safety in chiropractic. Geneva 2005.
28. Kanis JA, McCloskey EV, Johansson H, Oden A, Ström O, Borgström F. Development and use of FRAX in osteoporosis. *Osteoporosis Int.* 2010;21: 407-413.
29. Johnell O, Kanis JA. An estimate of the worldwide prevalence and disability associated with osteoporotic fractures. *Osteoporosis Int.* 2006;17(12):1726-1733.
30. Sánchez-Riera L, Carnahan E, Vos T, et al. The global burden attributable to low bone mineral density. *Ann Rheum Dis.* 2014;73(9):1635-1645.
31. Herzog W, Conway PJ, Kawchuk GN, Zhang Y, Hasler EM. Forces exerted during spinal manipulative therapy. *Spine.* 1993;18(9): 1206-1212.
32. Herzog W, Kats M, Symons B. The effective forces transmitted by high-speed, low-amplitude thoracic manipulation. *Spine.* 2001;26(19): 2105-2110.
33. Coulter ID, Crawford C, Hurwitz EL, et al. Manipulation and mobilization for treating chronic low back pain: a systematic review and meta-analysis. *Spine J.* 2018;18(5): 866-879.
34. Coulter ID, Crawford C, Vernon H, et al. Manipulation and mobilization for treating chronic nonspecific neck pain: a systematic review and meta-analysis for an appropriateness panel. *Pain Phys.* 2019;22(2): E55-E70.
35. Winterbottom M, Boon H, Mior S, Facey M. Informed consent for chiropractic care: comparing patients' perceptions to the legal perspective. *Man Ther.* 2015;20(3): 463-468.
36. Ford EC, Nyflot M, Spraker MB, Kane G, Hendrickson KRG. A patient safety education program in a medical physics residency. *J Appl Clin Med Phys.* 2017;18(6): 268-274.
37. Lewis KA, Ricks TN, Rowin A, Ndlovu C, Goldstein L, McElvogue C. Does simulation training for acute care nurses improve patient safety outcomes: a systematic review to inform evidence-based practice. *Worldviews Evid Based Nurs.* 2019.

“I stay in bed, sometimes all day.” A qualitative study exploring lived experiences of persons with disabling low back pain

Sharli-Ann Esson, BSc, MHSc¹

Pierre Côté, DC, PhD^{1,3}

Robert Weaver, MA, PhD^{4,5}

Ellen Aartun, MSc, PhD¹

Silvano Mior, DC, FCCS(C), PhD^{1,3,6}

Aim: To explore the lived experiences of persons with low back pain (LBP) and disability within the context of the International Classification of Function, Disability and Health (ICF) framework.

Methods: Qualitative study using focus group methodology. We stratified LBP patients into two low (n=9) and one high disability (n=3) groups. Transcript-based thematic analysis was conducted through an interpretivist lens.

Results: Four themes emerged: Invisibility, Ambivalence, Social isolation, and Stigmatization

Objectif : Étudier les expériences vécues par les personnes souffrant de lombalgies et de handicaps dans le cadre de la Classification internationale du fonctionnement, du handicap et de la santé (CIF).

Méthodologie : Étude qualitative utilisant des groupes de discussion comme méthodologie. Des patients souffrant de lombalgies ont été répartis en deux groupes : l'un Déficit léger (n = 9) et l'autre Déficit grave (n = 3). Une analyse thématique fondée sur des notes a été effectuée selon un point de vue interprétatif.

Résultats : Quatre thèmes ont été dégagés : invisibilité, ambivalence, isolement social, stigmatisation et marginalisation. Les participants ont expliqué

¹ Centre for Disability Prevention and Rehabilitation at Ontario Tech University and Canadian Memorial Chiropractic College (CMCC)

² Canada Research Chair in Disability Prevention and Rehabilitation, Ontario Tech University

³ Faculty of Health Sciences, Ontario Tech University

⁴ Department of Health and Exercise Science, Rowan University

⁵ Department of Medicine, Cooper Medical School of Rowan University

⁶ Department of Research, Canadian Memorial Chiropractic College

Corresponding author:

Silvano Mior, Canadian Memorial Chiropractic College, 6100 Leslie St., Toronto, Ontario, M2H 3J1

E-mail: smior@cmcc.ca

© JCCA 2020

The authors have no disclaimers or competing interests to report in the preparation of this manuscript.

Funding: Norwegian Research Foundation “*Et liv i bevegelse*” (ELIB), the Canadian Institutes of Health Research and the Canada Research Chair Program.

and marginalization. Participants described how environmental factors affected how they experienced disability and how their awareness of people's attitudes affected personal factors and participation in social activities. High disability participants experienced challenges with self-care, employment, and activities. The invisibility of LBP and status loss contributed to depressive symptoms.

Conclusion: LBP patients experience physical, social, economic and emotional disability. Our findings highlight the interaction between domains of the ICF framework and the importance of considering these perspectives when managing LBP patients with varying levels of disability.

(JCCA. 2020;64(1):16-31)

KEY WORDS: low back pain, disability, biopsychosocial model, ICF framework, qualitative research

Introduction

Lower back pain (LBP) is a leading cause of disability worldwide.^{1,2} It is one of the most prevalent chronic disorders and imposes a substantial economic burden globally.³ Approximately 80% of adults will experience LBP at some point in their lives.¹ LBP manifests itself as stiffness, tension or achiness confined between the costal margin and the inferior gluteal folds; with or without sciatica.⁴ The pathophysiological causes of LBP are often unidentifiable.⁵ This creates challenges to its effective treatment and management, especially because patients experience LBP in different ways.⁶ Others suggest that this unidentifiable pathology along with unclear diagnoses and often the lack of visible proof can cause LBP sufferers to be labeled as hypochondriacal, malingerers and even mentally ill.⁷⁻⁹ This may lead to disbelief or a dismissal of the seriousness and authenticity of disability associated with LBP.^{10,11}

In addition to the physical effects experienced by LBP patients, there are personal, societal and psychological ramifications associated with the condition.^{12,5} In some

de quelle façon les facteurs environnementaux se répercutaient sur leur façon de vivre avec le handicap et comment leur conscience des attitudes des gens se répercutaient sur les facteurs personnels et la participation aux activités sociales. Les participants souffrant d'un grave handicap éprouvaient des difficultés en ce qui a trait aux soins personnels, à l'emploi et aux activités. L'invisibilité de la lombalgie et la perte du statut contribuaient aux symptômes dépressifs.

Conclusion : Les patients souffrant de lombalgies ont un handicap physique, social, économique et émotionnel. Les résultats de notre étude mettent en évidence l'interaction entre les domaines du cadre de la CIF et soulignent l'importance de prendre en compte ces perspectives dans la prise en charge des patients souffrant de lombalgies causant divers degrés d'incapacité.

(JCCA. 2020;64(1):16-31)

MOTS CLÉS : lombalgies, handicap, modèle biopsychosocial, cadre de la CIF, recherche qualitative

cases, asocial behaviour and negative self-image are additional consequences of living with LBP.¹³ Furthermore, increased work absenteeism, lower productivity, status loss, and depressive symptoms often accompany chronic LBP.^{1,14} However, limited qualitative data is available which describes LBP patients' daily experiences with LBP associated disability from a biopsychosocial perspective.^{12,15,16} Thus, it is important to understand the everyday lived experiences of people with LBP and explore how psychosocial factors impact pain and disability, in order to effectively address them in their care plan.

The ICF Framework

In consideration of the biopsychosocial attributes of LBP, we framed our qualitative study using the International Classification of Functioning, Disability and Health (ICF) framework as a point of reference for our data collection.¹⁷ The ICF is helpful to conceptualize the positive and negative aspects of functioning from a biological, individual, and social perspective.¹⁷ The framework emphasizes the role of the environment by stressing the im-

portance of understanding the context in which the person lives and its interactions with health conditions and personal factors. The ICF includes five interacting domains: i) *body functions*: physiological functions of body systems (including psychological functions); ii) *body structures*: organs and limbs; iii) *activity*: execution of a task or action (including cognitive functions); iv) *participation*: involvement in a life situation; and v) *environmental factors*: physical, psychological, social, and attitudinal environment in which people live (barriers to or facilitators of functioning) (Figure 1). The ICF framework is the international reference for the conceptualization and evaluation of disability.¹⁷ It is in line with the UN Convention on the Rights of Persons with Disabilities and provides a common and universal language to understand disability and human functioning across communities.^{18,19} The ICF framework provides a structured guide for the conceptualization, collection and organization of data ne-

cessary to arrive at a comprehensive understanding of an individual's lived experience, within the context of their health condition. Because disability denotes "*the negative aspects of the interaction between an individual (with a health condition) and that individual's contextual factors*"¹⁷ a clinician engaged in patient care must seek to understand the individual's environmental and personal factors, if appropriate care is to be delivered.

We used the ICF framework to guide our analysis and address our objective of exploring the lived experiences of persons with low back pain and disability. Our study is part of an international, collaborative project between the Ontario Tech University and the ICF Research Branch (a cooperation partner within the WHO Collaborating Centre for the Family of International Classifications in Germany at the German Institute for Medical Documentation and Information (DIMDI)). The aim of this international collaborative project is to identify the aspects of

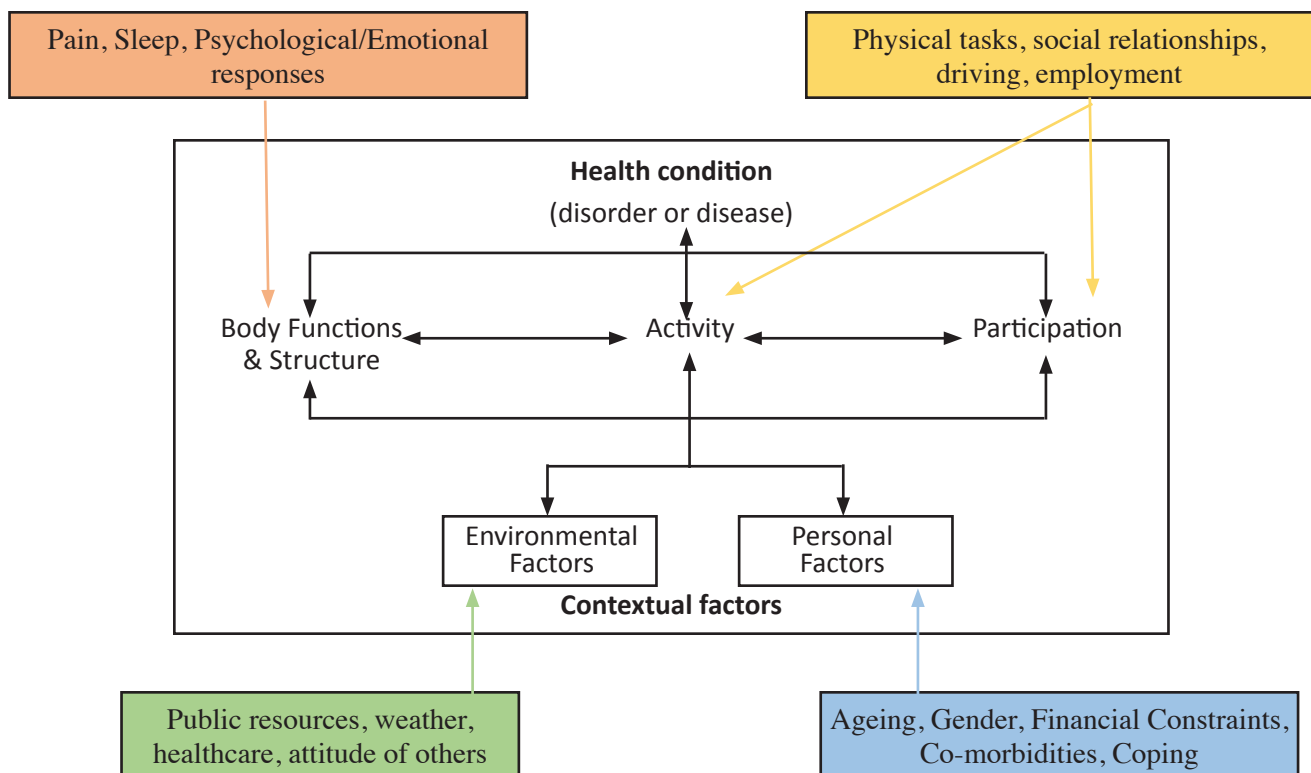


Figure 1. Domains of the International Classification of Function, Disability and Health Model and related emergent sub-categories.

functioning that are most important to participants and subsequently develop an ICF assessment schedule, a standardized measurement instrument, specifically designed for manual medicine for the reporting of functioning. Our study investigates aspects of functioning among patients with LBP in Ontario, Canada.

Materials and Methods

Study design

We used a qualitative design to explore the everyday experiences of persons with LBP. We used Interpretive Phenomenological Analysis (IPA), situated within the interpretivist paradigm, to understand participants' experiences. IPA reveals complex and dynamic relationships and places value on the subjectivity of participants' experiences.¹³

We used focus groups to elicit these everyday experiences. Focus groups offer a forum that enables participants in similar circumstances to share their experiences, and often facilitate disclosure of additional and more nuanced responses regarding their own experiences. Focus groups provide richness in the data that reflects the synergy between participants and explores their perceptions of an issue.²⁰ Ethics approval was obtained through the Research Ethics Boards of Ontario Tech University (REB # 14050) and CMCC (REB # 1629014).

Participants and recruitment

Participants were recruited from three Canadian Memorial Chiropractic College (CMCC) teaching clinics in the Greater Toronto Area (GTA) in Ontario, Canada. Participants were eligible to participate if they met the following criteria: 1) 20-65 years of age; 2) reported LBP; 3) were receiving chiropractic care for their LBP; and 4) spoke English.

Participants were recruited through advertisements placed in clinic reception rooms and by clinicians informing their patients about the study. CMCC staff clinicians introduced the study to patients and identified interested patients. The first author contacted interested patients and provided them with study information and the informed consent package. Focus groups were scheduled at the convenience of participants. Each focus group was conducted in a private room within the clinic, and situated in a convenient location for participants.

Focus group allocation

We used the World Health Organization Disability Assessment Schedule (WHODAS) to stratify participants into low disability focus groups (LDFG) and high disability focus groups (HDFG). The WHODAS is a 12-item, self-administered questionnaire designed to assess difficulty experienced doing regular, everyday tasks.²¹ The WHODAS is directly derived from the ICF and evaluates six domains of disability the "activity and participation" dimension of the ICF: cognition; mobility; self-care (hygiene, dressing, eating & staying alone); getting along (interacting with other people); life activities (domestic responsibilities, leisure, work & school); and participation (joining in community activities). The WHODAS is considered to be a valid and reliable measure of disability and thus was appropriate for stratifying our sample.^{22,23} The WHODAS is useful to measure disability in chronic low back pain patients and significantly positively correlated with the Roland Morris Disability Questionnaire, the Patient Health Questionnaire-9 item, the Screener and Opioid Assessment for Patients with Pain-Revised, the Current Opioid Misuse Measure (COMM) and the Opioid Risk Tool (ORT).²⁴ We used a pre-determined cut point of 36 out of a possible 60 points to allocate participants into LDFG and HDFG. A score above 36 is suggestive of a person having higher levels of disability severity. Previous studies used similar methods of stratification using this questionnaire.²⁵⁻²⁷

We anticipated recruiting 32 participants, with eight people in each of 4 groups, with an equal distribution of male and female participants. However, we presumed difficulty in recruiting equal distributions due to clinic population and would accept a 5:3 ratio of participants in each focus group.

Data collection

We used a script to guide questioning of participants. The focus group interview script was designed to elicit responses related to the ICF framework. Further probative questions explored answers to the questions in the event that what was said was not understood or required further clarification (Appendix 1). The script was reviewed in advance by the research team and pretested in a sample focus group to ensure clarity and comprehension. Each focus group was led by a trained facilitator (SE) and assisted by a co-investigator (EA). Focus groups were scheduled at

Table 1.
Demographic characteristics of participants in this study.

Characteristics	Low disability focus group #1 [LDFG1]	Low disability focus group #2 [LDFG2]	High disability focus group [HDFG3]	Total
<u>Gender</u>				
Male	2	2	1	5
Female	3	2	2	7
<u>Age Group</u>				
20-35	0	1	1	2
36-50	1	1	0	2
51-65	4	2	2	8
<u>Demographics</u>				
Employed	2	1	0	3
Unemployed/student	2	3	1	6
Retired	1	0	2	3
<u>Total</u>	5	4	3	12

different times to accommodate participants availability. The focus groups lasted approximately 90 minutes each.

Each session was audio-recorded and subsequently transcribed verbatim with participants’ consent. The recordings were transcribed by an experienced transcriptionist. Each transcript was checked for accuracy by cross referencing the audio file with the transcribed document. Errors in content and sentence structure were corrected and extraneous sounds/comments noted. Finally, confidentiality of statements made by each focus group participant in transcripts was assured by providing pseudonyms. Transcripts were not returned to participants for review.

Analysis

We used the NVivo11 Software (QSR International Pty Ltd. Version 11, 2015) to organize and analyze the transcripts. There was broad agreement among team members regarding the essential meaning of the core elements of the ICF framework. The framework became part of the scaffolding used during the coding process. These elements provided the foundation for our thematic analysis, where emergent themes were identified and conceptually expanded. The first author imported transcripts into NVivo software and reviewed, identifying, organizing, and coding key passages in NVivo nodes. Team members discussed and resolved ambiguities in the coding process as they arose and until consensus was reached. Once agreement was reached coded nodes were linked to

components of the ICF framework. The framework was used to scaffold themes emerging from the data. Once preliminary themes were identified, the team further discussed how they interrelated within the context of the ICF framework until consensus was reached regarding the soundness of the emergent themes.

Results

We enrolled twelve participants in the study - seven women and five men, who participated in one of three focus groups. The two LDFG included five and four participants, respectively. The HDFG included three participants. In addition to their varying degrees of disability, participants also had varying ages, ethnicities, and socio-economic backgrounds, including students, employed, unemployed and retired individuals (Table 1).

ICF Domains

Based on the five a priori domains from the ICF framework, participant experiences were coded accordingly. Our findings suggest that the domains of “activity” and “participation” bear similarities that make it difficult to distinguish between them. Similar findings have been also reported by others²⁸⁻³⁰; therefore, we merged these two domains (Figure 1).

Body Function and Body Structure

Participants described various challenges associated with

body structure and body function. These included the exacerbation of, difficulties sleeping and varied emotional responses stemming from their condition and pain. In both the low and high disability focus groups, participants provided conflicting accounts about the location of their pain. Some participants suggested that their LBP was confined to one area – typically the small of the back, while others explained that their pain was not localized but rather travelled from one area to the next, making it difficult to predict when or where the pain would arise.

“When I first started getting the pain I would say it was somewhat localized and then it started spreading and now I can’t even tell the difference anymore because it is throughout my entire body.” Allan [HDFG3]

Difficulty falling asleep and interrupted sleep are common experiences amongst persons living with disability.³¹ Participants in the LDFG reported falling asleep was not difficult but they struggled to sleep restfully or remain asleep, often having to change positions to relieve their pain or discomfort:

“For me I have really rough nights sleeping so like every hour or so I have to wake up and stretch and move around. So, in the morning the same thing, it is about a half an hour of stretching and moving around before I can actually function.” Corrina [LDFG1]

In contrast, HDFG participants reported struggling not only with falling asleep but remaining asleep. Allan’s account clearly exemplifies these challenges.

“I would say both because it is almost impossible to find a comfortable position where you say, ‘OK I am not in pain in this position so I will stay here.’ You find yourself tossing and turning all night long trying to find a position that works and usually you don’t and 9 times out of 10 the only reason you do fall asleep is from restlessness.” Allan [HDFG3]

Participants described how their LBP negatively impacted their motivation to perform daily activities. Emotional responses and concentration on daily tasks varied

by participant group. While LDFG participants experienced few challenges with concentration or maintaining focus, the HDFG participants described a significantly diminished ability to concentrate, having to work much harder than before:

“...I also have a hard time concentrating. So, my concentration when it comes to studying doesn’t last more than like 10-15 minutes. So I have to study in like 10-15 minutes fighting to read and then break 5 minutes... before I would just go to class listen and barely have to study anything or read too much now I find myself doing 10 times more work just to get one section over with.” Allan [HDFG3]

Activity and participation

There were marked contrasts among the participants in the ability to engage in physical actions, which affected their social relationships, driving and employment. Participants in the LDFGs expressed few activity limitations. They were able to differentiate between activities they could manage and those seen as detrimental to their ability to function. Unlike the HDFG participants, the LDFG participants reported being better able to manage their pain by modifying, rather than limiting, their activities. Many enjoyed cycling, yoga and swimming, but avoided high intensity exercises such as running, which they maintained placed severe pressure on their back and legs/knees.

“I went to a trampoline park with my friends...I had to completely stop because of pain in my neck, pain in my back...and I’m like well I’m going to watch you guys...because you know you can’t really do the same level as they can...” Leo [LDFG2]

Conversely, the HDFG participants struggled with even elementary body movements and body positions, and described serious exercise restrictions:

“Lying flat is very, very painful. Bending down like as the day progresses the worse I get and by the end of the day it is nearly impossible to function.” Helena [HDFG3]

Participants in the HDFG noted that their chiropractors

recommended exercises to manage their LBP but felt the chiropractor did not understand the challenges they faced in doing the exercises. This is an example of the dissonance between LBP patients and their healthcare providers which may impact their compliance.³²

"It limits your ability to do things especially exercise. So, it seems like everybody where you go for treatment recommends exercise but they kind of don't understand that it is very hard to do things, especially when you squeeze, the pain just intensifies times 50." Allan [HDFG3]

Most LDFG participants suggested their condition did not negatively impact their social interactions. In contrast, participants in the HDFG described a more dramatic change in social relationships, which included loss of friends and the desire to socialize. These findings are typical of persons living with severe back pain and supports findings in previous literature.^{13,36,37}

"I just don't return calls if they call. I don't think they understand, they don't understand what you are going through." Francine [HDFG3]

The employment status of persons in the LDFGs varied and included retired persons, unemployed persons, students and working persons. Those who worked were aware of their physical capabilities and sought employment accordingly:

"I can't really do certain physical jobs because I am not sure if it is going to tighten up...So I try to stay away from anything like that. The problem is a lot of jobs are going to still require standing anyways." Leo [LDFG2]

HDFG participants reported fewer employment opportunities compared to those in the LDFG. All HDFG participants were unemployed. For one participant, it was a personal choice to become full-time caregiver for a loved one. Another participant was no longer able to assume the labour-intensive demands of their work. Yet another participant quit her job because other co-workers assumed her compensatory movements and gait were related to her being intoxicated. HDFG participants expressed a desire

to return to work but noted their LBP prevented them from long periods of sitting and standing. They viewed seeking new employment as a challenge, fearing the potential employers' reactions after disclosing their LBP.

"It is also hard to try and get another job...So when I go and try and get jobs I would rather be honest...When you say those kinds of things to people about how you really are, it is like OK, right away you look at their face and you're like 'I know I didn't get this job.'" Allan [HDFG3]

Environmental factors

Environmental factors that impacted participants included public resources, healthcare and the attitudes of others. Communal spaces and transit were the primary public resources discussed by participants. Many of the participants in the LDFG lived within the downtown core and took advantage of the many available community resources:

"They will also fall-proof your house. So that is one of the things that you can get, you have to have a doctor referral to it but they will come in and look at your house and how you have it set-up and then do the fall prevention." Walter [LDFG1]

Participants in the HDFG, who also lived in the downtown core, were significantly less informed about community resources. They knew that some resources were available online but struggled to access them because they did not own a computer, were unaware how to access, or could not afford some resources. A student in the HDFG described classroom design, uncomfortable seating and poor accessibility as a barrier to attending classes. They also noted that although campus buildings were equipped with handicap push buttons to automatically open doors, many simply did not function:

"At my school I would say about 75% of the handicap buttons don't work and if they do work maybe it is only in the summer time because in the winter they get jammed." Allan [HDFG3]

Participants with HDFG relied on elevators or es-

calators to get to higher floors in multi-story buildings. Where neither were available, they relied heavily on the handrails of the stairs:

“So every time I walk into a building I always like to know where the elevator is or escalator or some easier way to get up and if the last resort is the stairs then I have to kind of coach myself into doing it...” Allan [HDFG3]

Public transit was reported as a significant concern among most participants. Buses and streetcars were the most frequently used modes of transportation amongst participants. Participants in both groups expressed caution and care when moving on and off buses and streetcars. The physical design of the vehicles made travel difficult for participants. One participant suggested that bus seats provided no back support and aggravated their pain:

“Yes, their seats are really bad for people with lower back pain. It is like sitting on a metal plate.” Allan [HDFG3]

Participants also described experiences with other transit users, ranging from being helpful by offering a seat to flat-out dismissive. Participants experienced feelings of frustration as their disability often went unnoticed, with few fellow passengers understanding their pain and functional impairment. Whether in interactions with family members or with persons on a bus, LBP sufferers often encounter others’ disbelief of their disability – if they appear fine on the outside, they must be fine on the inside too.³³ Since they “look good” and appear to be able-bodied and fully functional, participants felt their pain was misunderstood and delegitimized.

All participants sought treatment from general practitioners and chiropractors. Participants in both low and high disability focus group were pleased with the treatment they received in the chiropractic clinic. A few participants detailed the empathetic and understanding nature of their chiropractor and positive outcomes of care:

“Actually, my chiropractor now is actually having me...stand straight and you move your hips forward, like a tilt kind of thing, and that’s how you

walk and it’s amazing. The pain is much less over a fairly long period of time you can actually walk properly.” Mallory [LDFG2]

“I do like when the chiropractor does work on me. Basically, they stretch it out first and then put menthol or whatever stuff they put on it. Like this morning I was there and I find that I can move around a lot better once they do that.” Helena [HDFG3]

In addition to chiropractic treatments, participants in the LDFGs were more actively involved in their care and encouraged interprofessional correspondence between those involved in their treatment, including the fitness expert at the gym. HDFG participants were mindful of what they were feeling so they could appropriately articulate them to their chiropractors. Participants also said their chiropractors made suggestions about strategies or equipment they might use to cope with various everyday challenges.

A unique and interesting finding about participants attending for chiropractic care was their opportunity to interact with others in the waiting room. Some participants did not have healthy social lives and seemed to appreciate the friendly environment in the clinics. They often treated their chiropractic appointments as a part of their social calendar.

“Some people that go to the bar and they drink and try to get rid of their stress which actually makes things worse and to socialize. Believe it or not...I actually get a bit of a high in coming in from my treatment. So I am getting the medical help and it is also a social structure too.” Mason [LDFG2]

The attitudes of friends, family and the general community were important to all participants. However, there was a disparity between the groups. Participants in the LDFG shared varying experiences about the attitudes of family members and community members. Some reported that healthy family and social relationships did not much differ from when they did not have LBP.

“Yes mine hasn’t affected things that much with getting together with friends and that, so I am lucky ...” Wendy [LDFG2]

Conversely, participants in the HDFG saw living with LBP as the reason why they experienced daily personal strife. They believed their LBP led to the decline of relationships. They felt their friends and family did not understand what it was like to live with LBP and were often reluctant to discuss the pain they experience, and instead would steer conversations away from pain and disability or even distancing themselves from others.

“I find that people say they will be there for you, they are your friends or whatever and even family, and all of a sudden there will be days or times when I need somebody for even emotional support or physical support to do something, and everybody is busy or they don’t want to come or they don’t want to hear about it.” Helena [HDFG3]

HDFG participants implied their LBP was wholly responsible for their inability to work or effectively function in social settings. As has been reported elsewhere¹³, respondents also were made more aware of their disability when in the presence of those who have not experienced back pain, and they worried about how others perceived them.

Personal Factors

Personal factors that affect participants included age, co-morbidities, and financial constraints; gender impacted frequency of activity. Ageing and comorbidities affected participants’ differently. HDFG participants did not perceive that age impacted their level of disability but felt their comorbidities did. In contrast, LDFG participants were less affected by their co-morbidities and questioned whether their experiences with disability were a result of normal ageing processes rather than LBP:

“I think my emotional state is just understanding that this is a 51-year-old body that has gone through a lot of sports and athletics and knocks and bruises and stuff like that.” Val [LDFG1]

Financial constraints were a recurrent theme among HDFG but not so in the LDFG participants. HDFG participants’ primary concern was with the cost of engaging in certain activities or using resources such as a gym. Instead they emphasized the need to satisfy basic needs such as securing healthy food and shelter.

“Eating is expensive... You buy what is healthy and what is on sale and you try to eat healthy... they say with the inflammation you have to watch what you eat... you have to watch dairy and gluten and all that stuff but again they are expensive stuff.” Francine [HDFG3]

Self-management was the primary coping mechanism for participants in both low and high disability groups. It allowed them temporary relief from their LBP and gave them the opportunity to function more adeptly in everyday situations. They used various temporary modalities to alleviate their pain such as hot/cold packs, topical pain-relieving creams and painkillers. A few participants also mentioned that they found deep breathing exercises and meditation to be effective. Other enablers to functioning included developing creative self-management techniques and interacting with other LBP patients. One participant in the HDFG decreased the discomfort she experienced when travelling on public transit by carrying a backpack stuffed with soft items (scarves, clothes etc.) and used it as a cushion to ease the pressure on her back. Another participant said that receiving advice from other LBP patients and learning about different coping strategies improved her ability to function.

“Hearing what other people are doing, I think community support is a big thing, because everybody knows one piece of the puzzle but nobody knows the whole puzzle.” Corrina [LDFG1]

Interrelated themes

Due to the interrelated nature of the ICF domains, we identified four emergent themes that recurred across all the focus groups and were interwoven among the domains. We summarized participant responses within these respective themes as: Invisibility, Ambivalence, Social isolation, and Stigmatization and marginalization.

Invisibility

Since chronic LBP is not physically visible, non-sufferers often do not validate that the condition is real to sufferers.²³ For example, some participants described the attitudes of transit operators who did not recognize their disability, while using public transportation. They expressed con-

cern that operators often maneuvered buses in a less than smooth manner and often accelerated into traffic before they were seated or in a secure standing position. Corrina recounted her experience using transit buses: *“They will put the ramp down but they are not going to put it down for someone who ‘looks good’” [LDFG1]*. A participant in the HDFG described an encounter while using public transportation, where another passenger asked her to surrender the accessible seat she was occupying to another passenger who appeared to need it. Participants reported feeling frustrated by the lack of recognition of their disability. Even when LBP sufferers tried to explain their symptoms to others, non-LBP sufferers often failed to recognize or believe the suffering and functional impairment of LBP sufferers. Whether through interactions with family members or strangers, the pain and disability LBP sufferers endure remains invisible. Their pain is not viewed as legitimate because they often appear to be able-bodied and fully functional.

Ambivalence

Participants in the HDFG seemed to display feelings of ambivalence about how to live with LBP. They seemed to grapple with whether to accept that they might be less able to do some things they were previously capable of doing or to attempt to normalize their current situation, despite possibly requiring special consideration. Some used assistive devices to improve functioning. However, all participants in the HDFG were adamant about only using these devices temporarily as they strived to maintain their independence. Helena noted,

“I can do without any of those devices. I am better off because once you start using them, it is a crutch and basically your muscles and whatever further deteriorates because you are not using them... My independence with that is no good” [HDFG3].

Some participants reported refusing to use certain assistive devices altogether such as wheelchairs and walkers as they perceived them as symbols of disablement, choosing not to announce their disability to others. This is consistent with previous studies suggesting persons with disabilities often abandoned the use of assistive devices to avoid the judgement of others and prevent their potential social exclusion.^{34,35}

Despite lamenting that others often did not recognize their disability, participants were nonetheless concerned about appearing disabled and the accompanying perceived loss of social status. This contradiction illustrates an internal struggle that LBP patients must manage as they try to renegotiate and redefine the self to accommodate for lost capabilities.

Social isolation

The theme of social isolation spanned many domains of the ICF framework, reflecting the psychological, relational and emotional aspects of LBP sufferers. The emotional toll chronic LBP had on participants negatively impacted their motivation to perform daily activities. Depressive symptoms sometimes lead participants to withdraw and retreat to their homes for extended periods of time.²⁸ Participants described behavioural changes such as loss of self-esteem and social isolation that resulted from feelings of depression. Both LDFG and HDFG participants felt emotionally drained and disliked being dependent on others and assistive devices. In particular, participants in the HDFG felt especially overwhelmed and withdrawn and wanted to avoid the reality of their current situation. Francine stated, *“I stay in bed, sometimes all day which is even worse for the back pain...but if you don’t want to get out, you don’t want to get out...” [HDFG3]*. This withdrawal offers some relief from having to defend or explain a condition, which others may not acknowledge or understand.²⁹

Across the focus groups, participants expressed varying experiences related to social relationships. Most LDFG participants suggested that their condition did not negatively impact their social interactions; however, they did acknowledge small changes in their relationships. For example, one participant identified a change in the interests she previously shared with friends. When the interests were no longer shared, friendship ties became frayed:

“I mean I had work friends but only at work. Once you leave work, they go home you know and didn’t really have time to talk...My friends are not interested in what I want to do ok so I would like to see people more interested in what I want to do and I will join them” [Corrina, LDFG1].

Their accounts illustrate the strain on relationships that

can occur when the primary subject of conversation revolves around chronic pain and may eventually become bothersome to friends, who may not understand this pain. Respondents felt that friends sometimes shied away from them to avoid such conversation or interaction.

In contrast, participants in the HDFG described a more dramatic change in social relationships, which included loss of friends and loss of the desire to socialize. Allan notes, *"You will probably lose all your friends, they will become tired of always having to lag behind"* [HDFG3]. Francine illustrates the lost desire to socialize and the perceived dissonance between LBP sufferers and non-sufferers: *"I just don't return calls if they call. I don't think they understand, they don't understand what you are going through"* [HDFG3]. These changes appear consistent with persons living with severe back pain.^{36,37,13}

Stigmatization and marginalization

Stigmatization, and the marginalization that often accompanies it, became apparent in the focus groups as participants discussed their physical activities as well as employment, or lack thereof. Employed participants in the LDFG were aware of their physical capabilities and limitations, and sought employment within these confines:

"If I am looking for work I can't really do certain physical jobs because I am not sure if it (his back) is going to tighten up... So I try to stay away from anything like that" [Leo, LDFG2].

Unlike their counterparts, participants in the HDFG described considerably fewer employment opportunities. At the time of the focus group session, all participants in the HDFG were unemployed. Some expressed a desire to return to work but noted that their LBP caused diminished sitting and standing capabilities. The idea of seeking new employment became a challenge, as participants feared the reaction of potential employers once they disclosed their condition:

"It is also hard to try and get another job... So when I go and try and get jobs I would rather be honest... When you say those kinds of things to people about how you really are, it is like OK! Right away you look at their face and you're like 'I know I didn't get this job'" [Allan, HDFG3].

When asked about what would enable them to function in the workplace, participants in the HDFG said that it was important for employers to be empathetic towards their need for frequent breaks. They feared that their LBP would not be recognized and that employers might think they did not take their jobs seriously.

The discomfort, shame, and stigma associated with the negative responses of others towards LBP sufferers has also been directly linked to depressive symptoms and isolated behavior.¹⁰ Some participants felt that family members had other concerns and chose not to discuss their LBP. In this regard, the disinterest of family members caused feelings of marginalization. Val noted,

"...you are at the dining room table with your family, there is always other people's issues that are more important and more pressing kind of thing, than just 'oh, you just have lower back pain; Whatever!'" [LDFG1].

This finding supports previous work by Smith and Osborn¹³ who found that social situations often intensified the psychological dilemma faced by LBP patients as they become self-conscious and are fearful of the judgement of others.

Discussion

Our findings suggested both commonalities and divergence between LDFG and HDFGs. The ICF conceptualizes activity and participation as two distinct categories. However, numerous researchers have argued that the domains of activity and participation within the ICF model are difficult to distinguish.³⁰⁻³² Our findings suggest these two domains bear many similarities and often supplement each other. Therefore, the domains of activity and participation were merged and reported together to show individual limitations and the resulting restrictions that LBP patients experience.

The ICF framework and its diverse domains enabled us to capture an array of experiences identified by LBP sufferers in LDFGs and HDFG. Persons in the LDFGs had higher levels of functionality but living with LBP required them to modify several of the activities of daily living. Further, they demonstrated increased awareness of the events and activities they could and could not safely and easily participate in. In most low-disability cases,

familial relationships and friendships were only minimally affected. Nonetheless, several participants expressed some emotional responses and depressive symptoms which they associated with living with LBP.

HDFG participants also experienced emotional challenges living with LBP, but their social isolation and depressive symptoms appeared to be more extreme. Their physical abilities were more diminished and there was evidence of some fear avoidance behaviour. Their interpersonal relationships with family and friends were significantly strained and, in some cases, completely severed. Participants in the HDFG showed a greater proclivity toward social isolation as a result. They also demonstrated a heightened sensitivity toward and awareness of how their illness was perceived by others and how people behaved toward them. They felt they were no longer able to maintain social relationships or carry out gainful employment. These experiences support findings by Walker²⁹ who developed the theme of loss in their article. Our participants reflected upon the physical, social, and economic losses that may occur as a result of high levels of disability associated with LBP.¹²

Public transportation was a major topic of conversation in our focus groups. Most participants agreed that many of their experiences using public transportation were unpleasant and this provided a clear example of the challenges that LBP sufferers face as a result of living with an invisible condition. The uncomfortable seating and less than smooth rides had physical consequences for LBP patients. However, previous literature has focused primarily on the LBP in transit operators rather than passengers, suggesting that drivers' seats needed to be ergonomically evaluated and adjusted accordingly.⁴¹ Our data suggest an equally important need is to also assess and evaluate the passengers' perspective. A significant portion of the experiences described by the participants pertain to the effects of environmental and personal factors as articulated in the ICF model.

Previous quantitative studies suggest LBP sufferers are subjected to loss of employment, social identity and inequality; experience isolationism, depression, distressing experiences; as well as pain, disability and low well-being.^{3,5,13} However, there are fewer qualitative studies exploring the in-depth understanding of patients' pain experiences with LBP.^{13,28} A recent systematic review identified three overarching themes emergent from

28 qualitative studies on chronic LBP: impact on self; relationships with family and friends, and health providers and organizations; and coping.^{13,28} Yet, few of the included the qualitative studies assessed the effect of age, gender, physicality, temporality and disability on patients' experiences. Our study adds to this gap in the literature by having stratified our focus groups into low and high disability. The two groups described similar experiences, though their salience and consequences varied considerably. This offers an important first step toward understanding the experiences and impact of different levels of LBP and disability. Future research should go beyond the binary distinction used here, to explore how more subtle differences in levels of LBP and disability affect experiences and behaviours of those afflicted.

Our findings confirm that disability associated with LBP has multiple and often simultaneous effects.⁴² For example, participants indicated that physical pain contributed to their inability to complete activities or participate in events which in turn influenced people's attitudes towards them, friendships, and sense of isolation. This supports the reported direct interaction between body function, activities, participation, and environmental factors of the ICF model.⁴³

Our findings highlight the benefits of using a biopsychosocial model, specifically the ICF model, to interpret our data. Our findings support the connections among the domains of the ICF model as manifested in the lives of those afflicted with LBP. The feedback loop between the domains in the framework is reflected in the description of participants' lived experiences in our study. Our findings support the contention that personal factors influence the other domains and humanizes the ICF framework by valuing and respecting the uniqueness of the person.^{11,43} Thus, our study adds to the paucity of literature assessing the potential utility of the ICF in clinical settings.

Strengths and limitations

The use of the ICF framework is a major strength of our study. Its expansive framework has been shown to be useful and generalizable in a variety of scenarios and is applicable to other health conditions and disabilities.²³ Additionally, the connections between our data and data previously collected in other studies that also utilized the ICF framework affirms our decision to use this model.⁴⁴ Other strengths of our study relate to our focus on eliciting

ing participants' everyday experiences living with LBP. We recruited participants with varying demographic profiles and low and high levels of disability. The qualitative approach encouraged participants to share freely and the results are likely to be clinically applicable.

There were limitations in the study as well. First, despite efforts to recruit participants and extend data collection period, we were unable to achieve our predetermined sample estimate per focus group. Second, we were unable to represent fully the similarities and differences between employed and unemployed participants, as most focus group respondents were unemployed, which may suggest that employed people have less time to participate in focus groups. Third, we were only able to conduct one high disability focus group. Our results showed that LBP patients with high disability experienced greater restriction in mobility (transportation), which could be an indication that attending focus groups was more difficult for these persons. We suggest that further research be conducted in this regard. Fourth, the limited age distribution of participants impacts our ability to interpret their lived experiences. Fifth, the sample only captured the perspectives of chiropractic care seekers, and may under-represent LBP sufferers with sub-clinical symptomology, or who seek traditional medical care or no care at all. Finally, we crudely differentiated subjects into low and high disability groups that may not account for more subtle distinctions with regard to LBP severity. Further research might include a middle group to help detect more subtle differences with regard to LBP severity.

Significance / implications

Our study raises awareness about the importance of environmental and personal factors in the ICF framework and their unique interaction with, and influence on persons' lived experiences. This information facilitates clinicians by encouraging them to consider these factors in their understanding of their patients' disability and modifying their management strategies.

Also, our data contributes an important component to an international, collaborative project by providing a unique local Canadian perspective of how LBP patients experience disability. We were able to determine some of the environmental and personal factors on the ICF framework, which LBP patients describe as affecting their disability and functioning. The data will complement quali-

tative data collected in Norway and Botswana. Using similar qualitative methodology, the data collected from different regions make it possible to access results across cultures and nations, strengthening the ability for regional and cultural comparisons. This will aid in the creation of a standardized assessment tool which will contribute to improved patient centered models of care and facilitate clinicians' ability to better assess and document disability in LBP patients within the context of the ICF framework.

Conclusion

Our study supports the notion that LBP is associated with varying social and psychological consequences in sufferers' daily lives that may not be assessed, documented nor addressed in their clinical care. The ICF framework addresses the often-overlooked social factors of the biopsychosocial model but also includes the impact of environmental and personal factors. The findings of our study support the need to measure and address important social factors, often underrepresented in previous work.^{45,46} Furthermore, our findings highlight the inherent interrelatedness of the dimensions of the ICF framework as they manifest in the narratives describing the lived experiences of people who suffer from LBP, while valuing and respecting the uniqueness of the person.

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(17): 1860-1866.
2. Vos T, Barber R M, Bell B, Bertozzi-Villa A, Biryukov S, Bolliger I, Murray C J L, et al. Global, regional, and national incidence, prevalence, and years lived with disability for 301 acute and chronic diseases and injuries in 188 countries, 1990-2013: a systematic analysis for the Global Burden of Disease 2013. *Lancet* 2015; 386(9995): 743-800.
3. Mutubuki EN, Luitjens MA, Maas ET, Huygen FJPM, Ostelo RWJG, van Tulder MW, van Dongen JM. Predictive factors of high societal costs among chronic low back pain patients. *Eur J Pain*. 2019; 24(2): 325-337.
4. Van Tulder M, Koes B, Bombardier C. Low back pain. *Best Pract Res Clin Rheumatol*. 2002; 16(5): 761-775.
5. Hartvigsen J, Hancock MJ, Kongsted A, Louw Q, Ferreira MI, Genevay S, et al. What low back pain is and why we need to pay attention. *Lancet* 2018; 391(10137): 2356-2367.
6. Wong JJ, Côté P, Sutton D A, Randhawa K, Yu H,

- Varatharajan S, et al. Clinical practice guidelines for the noninvasive management of low back pain: A systematic review by the Ontario Protocol for Traffic Injury Management (OPTIMa) Collaboration. *Eur J Pain*. 2017; 21: 201-216.
7. Glenton C. Chronic back pain sufferers--striving for the sick role. *Soc Sci Med*. 2003; 57(11):2243-2252.
 8. Crowe M, Whitehead L, Gagan MJ, Baxter GD, Pankhurst A, Valledor V. Listening to the body and talking to myself – the impact of chronic lower back pain: a qualitative study. *Int J Nurs Stud*. 2009; 47(5): 586-592.
 9. Dima A, Lewith GT, Little P, Moss-Morris R, Foster NE, Bishop FL Identifying patients' beliefs about treatments for chronic low back pain in primary care: a focus group study. *Br J Gen Pract*. 2013; 63(612): e490-e498.
 10. Holloway I, Sofaer-Bennett B, Walker J. The stigmatisation of people with chronic back pain. *Disabil Rehabil*. 2007; 29(18): 1456-1464.
 11. Darlow B1, Dean S, Perry M, Mathieson F, Baxter GD, Dowell A. Easy to harm, hard to heal: patient views about the back. *Spine* 2015; 40(11): 842-850.
 12. Froud R, Patterson S, Eldridge S, Seale C, Pincus T, Rajendran D, Underwood M. A systematic review and meta-synthesis of the impact of low back pain on people's lives. *BMC Musculoskel Dis*. 2014; 15: 50.
 13. Smith JA, Osborn M. Pain as an assault on the self: An interpretative phenomenological analysis of the psychological impact of chronic benign low back pain. *Psychol Health*. 2007; 22(5): 517-534.
 14. Nolet PS, Kristman VL, Côté P, Carroll LJ, Cassidy JD. Is low back pain associated with worse health-related quality of life 6 months later? *Eur Spine J*. 2015; 24(3): 458-466.
 15. Walker J, Sofaer B, Holloway I. The experience of chronic back pain: Accounts of loss in those seeking help from pain clinics. *Eur J Pain*. 2006; 10(3): 199-207.
 16. Snelgrove S, Edwards S, Lioffi C. A longitudinal study of patients' experiences of chronic low back pain using interpretative phenomenological analysis: changes and consistencies. *Psychol Health*. 2013; 28(2): 121-138.
 17. World Health Organization. International Classification of Functioning, Disability and Health: ICF. Geneva: WHO; 2001.
 18. Madans JH, Loeb ME, Altman BM. Measuring disability and monitoring the UN Convention on the Rights of Persons with Disabilities: the work of the Washington Group on Disability Statistics. *BMC Pub Health*. 2011; 11 Suppl 4: S4.
 19. Cieza A, Fayed N, Bickenbach J, et al. Refinements of the ICF Linking Rules to strengthen their potential for establishing comparability of health information. *Disabil Rehabil*. 2016; Mar 17: 1-10.
 20. Morgan D L. Focus Groups. In L. M. Given (Ed.) *The SAGE Encyclopedia of Qualitative Research Methods*. SAGE Publications, Inc., 2008: 353-355.
 21. WHO Disability Assessment Schedule 2.0 (WHODAS 2.0) [Internet]. World Health Organization. World Health Organization; 2019 [cited Jan 30 2020]. Available from: https://www.who.int/classifications/icf/more_whodas/en/
 22. World Health Organization (WHO) (2002). Towards a common language for functioning, disability and health: ICF - The International Classification of Functioning, Disability and Health (ICF). Geneva: WHO. <http://www.who.int/classifications/icf/en/>
 23. Geyh S, Schwegler U, Peter C, Müller R. Representing and organizing information to describe the lived experience of health from a personal factors perspective in the light of the International Classification of Function, Disability and Health (ICF): a discussion paper. *Dis Rehabil*. 2019; 41(14): 1727-1738.
 24. Wawrzyniak KM, Finkelman M, Schatman ME, Kulich RJ, Weed VF, Myrta E, DiBenedetto DJ. The World Health Organization Disability Assessment Schedule-2.0 (WHODAS 2.0) in a chronic pain population being considered for chronic opioid therapy. *J Pain Res*. 2019;12 1855–1862.
 25. Von Korff M, Crane PK, Alonso J, Vilagut G, Angermeyer MC, Bruffaerts R, Ormel J. Modified WHODAS-II provides valid measure of global disability but filter items increased skewness. *J Clin Epidemiol*. 2008; 61(11): 1132-1143.
 26. Silva C, Coleta I, Silva AG, Amaro A, Alvarelhão J, Queirós A, Rocha N. Adaptation and validation of WHODAS 2.0 in patients with musculoskeletal pain. *Revista De Saúde Pública*. 2013; 47(4): 752.
 27. Marx BP, Wolf EJ, Cornette MM, Schnurr PP, Rosen MI, Friedman MJ, Keane TM, Speroff T. Using the WHODAS 2.0 to assess functioning among veterans seeking compensation for posttraumatic stress disorder. *Psychiatric Serv*. 2015; 66: 1312-1317.
 28. Snelgrove S, Lioffi C. Living with chronic low back pain: a metasynthesis of qualitative research. *Chron Illness*. 2013; 9(4): 283-301.
 29. Ashby S, Fitzgerald M, & Raine S. The impact of chronic low back pain on leisure participation: implications for occupational therapy. *Br J Occup Ther*. 2012; 75(11): 503-508.
 30. Naughton F, Ashworth P, Skevington SM. Does sleep quality predict pain-related disability in chronic pain patients? The mediating roles of depression and pain severity. *Pain*. 2007;127(3): 243–252.
 31. Fu Y, Mcnichol E, Marczewski K, Closs SJ. Patient-professional partnerships and chronic back pain self-management: a qualitative systematic review and synthesis. *Health SocCare Commun*. 2015;24(3):247–259.
 32. Hopayian K, Notley C. A systematic review of low back pain and sciatica patients expectations and experiences of health care. *Spine J*. 2014;14(8):1769–1780.
 33. Matthews CK. To tell or not to tell: The management

- of privacy boundaries by the invisibly disabled. In: Annual meeting of the Western States Communication Association, San Jose, CA., 1994.
34. Pape TL, Kim J, Weiner B. The shaping of individual meanings assigned to assistive technology: a review of personal factors. *Disabil Rehabil.* 2002; 24 (1/2/3): 5-20.
 35. Shinohara K, Wobbrock J. In the shadow of misperception: Assistive technology use and social interactions. Paper presented at the CHI 2011, Vancouver BC, Canada May 7-12, 2011: 705-714.
 36. Corbett M, Foster NE, Ong BN. Living with low back pain--Stories of hope and despair. *Soc Sci Med.* 2007; 65(8): 1584.
 37. Walker J, Sofaer B, Holloway I. The experience of chronic back pain: Accounts of loss in those seeking help from pain clinics. *Eur J Pain.* 2006; 10(3): 199-207.
 38. Jette AM, Haley SM, Kooyoomjian JT. Are the ICF activity and participation dimensions distinct? *J Rehabil Med.* 2003; 35(3): 145-149.
 39. Badley EM. Enhancing the conceptual clarity of the activity and participation components of the international classification of functioning, disability, and health. *Soc Sci Med.* 2008; 66(11): 2335-2345.
 40. Resnik L, Plow MA. Measuring participation as defined by the international classification of functioning, disability and health: an evaluation of existing measures. *Arch Phys Med Rehabil.* 2009; 90(5): 856-866.
 41. Okunribido OO, Shimbles SJ, Magnusson M, Pope M. City bus driving and low back pain: a study of the exposures to posture demands, manual materials handling and whole-body vibration. *Applied Ergon.* 2007; 38(1): 29-38.
 42. Arnow BA, Blasey CM, Constantino MJ, Robinson R, Hunkeler E, Lee J, Hayward C. Catastrophizing, depression and pain-related disability. *Gen Hosp Psych.* 2011; 33(2): 150-156.
 43. Geyh S, Peter C, Müller R, Bickenbach JE, Kostanjsek N, Ustun, BT, Stucki G, Cieza A. The personal factors of the International Classification of Functioning, Disability and Health in the literature – a systematic review and content analysis. *Disabil Rehabil.* 2011; 33(13-14): 1089-1102.
 44. Kostanjsek N. Use of The International Classification of Functioning, Disability and Health (ICF) as a conceptual framework and common language for disability statistics and health information systems [Internet]. *BMC public health.* BioMed Central; 2011 [cited Jan 30 2020].
 45. Carr JL, Moffett JAK. Review Paper: The impact of social deprivation on chronic back pain outcomes. *Chron Illness.* 2005;1(2):121–129.
 46. Campbell P, Jordan KP, Dunn KM. The role of relationship quality and perceived partner responses with pain and disability in those with back pain. *Pain Med.* 2012;13(2):204–214.

Appendix 1.
Focus group interview guide (abridged version).

1. In what part of your body is the pain localized?
Probe: location of primary and secondary pain and discomfort
 2. In what part of your body do you feel the pain is coming from?
Probe: Joints, muscles, bones
 3. What sorts of physical problems have you noticed about yourself while living with LBP?
Probes: strength and endurance; movements and posture
 4. What sorts of emotional or mental responses have you noticed about yourself while living with LBP?
Probes: ability to concentrate, if easily distracted, energy levels, ability to fall and stay asleep
 5. If you think about your daily life, what difficulties do you encounter living with LBP?
Probe: impact on day-to-day activities, carrying on with usual work or household activities
 6. Tell us about some of the social activities you are involved in.
Probes: limitations, barriers, impact on others (e.g. friends, family, colleagues); frequency socializing
 7. Think about yourself, your life situation, gender, who you are – how does it affect the way you function?
Probe: experiences with low back pain
 8. Thinking about your environment, e.g. home, working conditions and social settings, what do you think are some things that enable you to function better?
Probe: developed habits or use of devices
 9. How well do you think society understands you? Would you say people are supportive in helping you manage from day-to-day? How?
Probe: attitudes and assistance of those around you
 10. What services and/or resources in the community have you used and found helpful?
Probe: system or people assistance
 11. Reflecting or thinking about your surroundings, e.g. home, working conditions and social settings, is there anything that limits your ability to adequately function? What limits you and how?
Probe: challenges and limitations through the day
 12. Describe any services or resources which you find difficult to use or implement into your everyday life?
Probe: difficulties accessing or using resources or services
-

Patients with cancer. Is there a role for chiropractic?

Maria Tsampika Laoudikou, MChiro¹
Peter William M^cCarthy, PhD¹

People who have a diagnosis of cancer may develop, or already have musculoskeletal conditions, just like any other person. However, discussion about potential benefits of chiropractic treatment to this group has generally been avoided related to the fear of misrepresentation. We aimed to derive a consensus from a group of experienced chiropractors regarding their perception of what chiropractic care offered to patients with cancer. An anonymous, two stage, on-line, Delphi process was performed using experienced chiropractors (n=23: >10 yrs practice experience, who had treated patients with cancer) purposively selected and recruited independently. One opted out of the study, 13 actively engaged in two rounds of questions and verification; agreeing such patients gained benefit from chiropractic care but use of spinal manipulation was not essential. There was no clear consensus regarding a protocol for interaction within any multidisciplinary

Comme tout le monde, les personnes atteintes d'un cancer peuvent développer des troubles musculosquelettiques, si elles n'en ont pas déjà. En règle générale, on évite de discuter des éventuels bienfaits des traitements chiropratiques pour ce groupe de personnes de peur de faire de fausses déclarations. Nous avons cherché à obtenir un consensus auprès d'un groupe de chiropraticiens d'expérience à qui on a demandé ce qu'ils pensaient des traitements chiropratiques administrés aux patients cancéreux. On a mené une enquête Delphi anonyme, en deux étapes et en ligne, auprès de chiropraticiens d'expérience (n =23 : >10 ans d'exercice, ayant déjà traité des patients atteints d'un cancer) choisis et recrutés de manière indépendante. L'un d'entre eux a abandonné l'étude, 13 ont répondu à deux séries de questions et se sont soumis aux vérifications. Les chiropraticiens ont convenu que ces patients bénéficiaient des traitements chiropratiques, mais que le recours aux manipulations vertébrales n'était pas essentiel. Aucun consensus clair ne s'est

¹ Faculty of Life Sciences and Education, University of South Wales, Treforest, Wales, UK

Corresponding author: Maria Tsampika Laoudikou, Life Chiropractic Clinic. 58 East St, Southend-on-Sea, Essex, SS2 6LH, United Kingdom
Tel: 0044 07860805212

© JCCA 2020

The authors have no disclaimers, competing interests, or sources of support or funding to report in the preparation of this manuscript.

List of Abbreviations:

- ECU: European Chiropractic Union,
- WHO: World Health Organization
- CAM: Complementary and Alternative Medicine
- SMT: Spinal Manipulative Therapy
- MSK: Musculoskeletal

team treating the patient. Concerns were raised about misinterpretation of advertising any benefits for cancer patients from chiropractic care. Lack of evidence in this area was acknowledged.

(JCCA. 2020;64(1):32-42)

KEY WORDS: cancer, chiropractic, Delphi, evidence based care, integrated care, manual therapy, multidisciplinary practice, patient management, spinal manipulation

Introduction

Cancer is the second leading cause of death globally, accounting for 8.8 million deaths in 2015.¹ This disease can affect almost any part of the body and has many anatomic and molecular subtypes each requiring specific management strategies. The greatest step forward in the increasing success in treatment of this disease has derived from the improvements in understanding and early detection.^{2,3} The mixture of diversity of presentation, commonality of the condition and the rigors of treatment would make it highly likely that people with such a problem will develop or exacerbate pre-existing musculoskeletal conditions and as a result seek care from a manual therapist at some point in their therapeutic journey.^{4,6} It is critically important, therefore, that a responsible profession has protocols in place to recognize the possibility of diagnosis, facilitate access to the appropriate treatment of the condition by accurate referral or provide musculoskeletal support within part of an integrated care package for those already undergoing treatment.^{2,5}

Treatment of patients with cancer is an emotive subject in complementary and alternative healthcare circles.⁴ Although treatment of the cancer itself is restricted to orthodox healthcare by law in many countries, this has not prevented reports suggesting that other therapeutic modalities can be used to “cure” the disease.⁴ Mostly such claims are based on case reports and literature reviews and refer to a wide range of Complementary and Alternative Medicine (CAM) practice, with very little focus on chiropractic.⁷⁻⁹ However, this situation has creat-

dégagé autour d'un protocole d'interaction entre les membres d'une équipe multidisciplinaire traitant le patient. On s'inquiétait des idées fausses qu'on se fait sur les bienfaits des traitements chiropratiques administrés aux personnes atteintes d'un cancer et on a reconnu le manque de preuves sur cette question.

(JCCA. 2020;64(1):32-42)

MOTS CLÉS : cancer, chiropratique, Delphi, traitements fondés sur des preuves, soins intégrés, thérapie manuelle, pratique multidisciplinaire, prise en charge du patient, manipulation vertébrale

ed a degree of confusion and obfuscation, which has impeded serious discussion of the potential health benefits that CAMs such as chiropractic may have on issues such as the patients' quality of life. An added problem results from the difficulty in quantifying the effects of individual components of any integrated care package as many are probably indirect benefits loosely associated with recovery and remission.⁸⁻¹⁷ A further reason for not raising awareness of offering treatment to this group derives from allegations that CAM practitioners can delay appropriate access to care by failing to diagnose the metastatic disease in its early stages.¹⁸

It is generally accepted that musculoskeletal symptoms are common reasons for patients to present to a chiropractic practice.¹⁹⁻²¹ Indeed, the motivations for the patient with cancer to seek chiropractic care appear to be primarily the presence of neuro-musculoskeletal symptoms.²²⁻³⁶

Occasionally, patients who were unaware that the underlying cause of their symptoms was cancer present to manual therapists, on occasion being appropriately diagnosed and referred.^{22,25,29,30,32,34,36-38} Indeed, it is important to recognize that a number of primary tumours (lung cancer for example) may initially present with musculoskeletal symptoms.²⁷ The diagnosis of cancer for many of the above cases was made through a careful history and physical examination and/ or because the patient was not responsive to care.²²⁻³⁸ It is generally considered that chiropractic education and continued professional development emphasises the importance of the practitioner considering progression of severity and/or frequency of

symptoms as the need to trigger re-examination, which may then warrant further investigation. Additionally, the education of chiropractors includes extensive training in the recognition of diagnostic characteristics of various cancers⁹, including the use of radiographic imaging, which can play an important part in confirming the majority of such diagnoses³⁹⁻⁴¹. A driving force for this emphasis results from the fact that failure to diagnose, make the appropriate referral, or even the delivery of chiropractic manipulation when contraindicated could have potentially fatal consequences for the patient.⁴¹⁻⁴³

Patients undergoing treatment for their cancer usually have to battle both the psychological effects of the diagnosis and the metabolic effects of the therapeutic approaches; both of which are likely to increase the likelihood of musculoskeletal conditions adding to their burden. However, an analysis of CAM use in Washington, based on the claims data of two large insurance companies, revealed a slightly lower proportion of cancer patients (11.6%) sought chiropractic care when compared to those patients without a diagnosis of cancer (12.3%).⁴⁴ Although this change might be considered relatively insignificant, it does appear to be contrary to expectations based on the increased depression and anxiety as well as decreased activity (due to fatigue) that have been associated with having a diagnosis of cancer: all of which have been associated with increased musculoskeletal issues.^{45,46} Indeed, based on this outcome, possibly erroneously, the authors of that article concluded that spinal manipulation may not be relevant to patients undertaking cancer treatment. This perception, whether made by those delivering care or those requiring care, could be damaging to both the chiropractic profession and patients if not subjected to further consideration.

Although historically treatment plans for patients with cancer were focused on the disease, recently the importance of improving the quality of life of the patient has been recognised.⁸ As a proportion of patients with cancer do not have significant pain relief with the treatment received, it would be expected for these people to seek alternative options of pain relief. Hence, in order to quality control this aspect of the therapy, the concept of the cancer rehabilitation team has been developed. This concept aims at helping with the multidimensional problems faced by a patient with cancer⁹; however, interpretations such as those made from the Washington study⁴⁴ could impact on

the inclusion of certain forms of CAM such as chiropractic in any integrated care package.

Currently, little information is available regarding treatment of cancer patients by the chiropractic profession⁹, especially in Europe. The authors are aware of one initiative in the United States where the Cancer Treatment Centers of America (CTCA) promote themselves as being part of an integrative care plan adjusted on the needs of each cancer patient alongside other supportive therapies such as acupuncture and naturopathic medicine. Although their project aims to establish a more evidence informed approach showing how an integrative care plan could be of benefit for patients with cancer; to the authors' knowledge, there is currently no published research underpinning their approach.

We therefore chose to initiate our study of this area by gaining a range of views and maybe consensus from experienced European chiropractors who had treated patients with cancer as part of their general practice. The main issue was whether they considered their treatment to have benefitted these patients. We also wished to determine the degree of engagement with the other clinical disciplines responsible for treating the patient and what approach they might choose including use of manipulation and other therapeutic interventions.

Study Aims

Primary aim: to derive a consensus regarding whether chiropractic treatment was perceived to have any benefit for patients with cancer.

Secondary aim: to determine if there was consensus of approach regarding use of chiropractic in an integrated therapy package, as part of a multidisciplinary clinical team in the treatment of patients with cancer.

Methods

A two-stage Delphi process was performed using a panel constructed from chiropractors who were members of the European Chiropractors Union (ECU). A panellist needed to be a chiropractor with over ten years practice – based experience, during which time the panellist should have treated patients who either have or have had cancer. Members of the panel were purposively selected by a committee member of the ECU independently of the research team. The selection brief was to source chiropractors in practice who complied with the inclusion criteria

and would be interested in participating in this research process. The panel members were unaware of the names and locations of the other panel members.

To comply with current European Union legislation, each potential panel member was asked if they would like to consider being involved in this process, by giving approval to pass their email and practice addresses to the research team. At this point the person was signifying their interest in principle, without having detailed knowledge of the topic under investigation.

The contact details of 23 chiropractors were supplied to the research team who then circulated information detailing the research topic. At this point, the chiropractors who had shown an interest were free to choose to respond to the survey or not. Furthermore, the research team were not able to determine who had responded and who did not, which ensured anonymity for the participants. Both rounds of surveys were delivered to all members of this group who had not opted out (the panel). Informed consent was implied through both a statement in the introductory email text and as warnings given at the start and end of the questionnaire that submission would be considered implied consent to use the submitted data.

Panel members each received a personalised email with the link (active for two weeks) to the questionnaire that used the SurveyMonkey platform. This e-mail also contained reminders concerning the implied consent nature of the questionnaire, anonymity and the right to withdraw their involvement at any point up to the point they submitted their completed questionnaire. We also ensured panel members were aware that they could exit from the study at any time by simply asking to be removed from the email list.

The questionnaire mostly comprised free text option questions. Free text options were chosen to allow the panellists to include their opinions and experiences as well their management strategies regarding chiropractic care of patients with cancer.

The responses were collated and recirculated to the entire panel at the end of each survey, in order to verify that the responses and their synthesis were a true reflection of the panel's views. Verification was performed by uploading the summary document to the online platform (SurveyMonkey) and sending a link to all the panel, giving them the opportunity to add any further comments anonymously, if they so wished.

The questions for the second round were developed based on the responses from the first round, following verification. The aim of the second round was to delve deeper into the topic and clarify some of the issues raised about use of chiropractic treatment on cancer patients. Those questions were also distributed in the form of a survey using the same platform (SurveyMonkey). Access to the second questionnaire was available for four weeks. A similar verification procedure was completed before the final analysis.

Ethical approval was granted by the chiropractic undergraduate research ethics review subgroup (granted devolved responsibility from the Faculty of Life science and Education Ethics Committee, University of South Wales).

Results

Twenty-three chiropractors were contacted to take part in the project as part of the panel by the ECU member. One of them contacted the research team asking more details about the project and decided to opt out before the release of the first questionnaire. Thirteen of the 22 remaining panellists responded to the first questionnaire (59%) with three contributing to the first verification stage. Thirteen of the 22 responded to the second-round questionnaire, with none engaging in the second verification stage. Due to the anonymity of the respondents, it was not possible to determine whether the same 13 responded to both questionnaires or not. Those engaging in the verification did so only to suggest minor changes.

Demographics of the panel:

Although anonymous, limited information was available about the 13 panel members (from responses to direct questions on the questionnaire). Only one had less than 15 years' experience; the majority (7/13) had between 15 and 20 years' experience, with five having more than 20 years' experience. Seven of the panel had studied chiropractic outside the UK. Details on those who chose to not to respond was not available.

Areas of unanimous or general agreement:

Of those choosing to respond, it was unanimously agreed (13/13) that there were benefits that the patient with cancer could derive from chiropractic care. According to the majority of the panel (9/13) the perceived benefits were

similar to those recognised and reported by patients without cancer. The panel unanimously agreed that the role of chiropractic treatment in patients with a diagnosis with cancer should not differ from its role for any other patient. The following were mentioned by at least one of the panel members:

- Chiropractic could help a patient with cancer in terms of their: pain relief, empathy, mobility, energy levels, quality of life, sleeping patterns and function.
- Perceived benefits of chiropractic care in this group of patients were reported to include: pain relief, sleep pattern improvement, immune system improvement, wellbeing, higher energy levels and psychological reinforcement.

The whole panel agreed that a cancer diagnosis should make a difference to a chiropractic treatment plan.

- The range of reasons given for this included: the medication used, possibility of metastasis, possible bone density or ligamentous integrity alterations due to the cancer. Three of the panel stated that post-chemotherapy osteoporosis and cancer diagnosis must be considered a red flag before any treatment protocol be considered.

All the panel members concurred that SMT should not be used on all cancer patients. Although the panel stated that SMT was not considered necessary on all occasions; it was also stated that SMT should not be contraindicated in any plan of management. There were a range of different exclusion criteria offered, the main one being metastasis (6/13 responses). Other contraindications mentioned included stage, type and location of the tumour along with the extent of the area involved, the overall health of the patient, muscle weakness, atrophy and osteoporosis.

Interestingly, three of the five participants that had been in practice for 20 or more years and reported seeing 10 or more patients with cancer a year agreed it was appropriate to adjust areas other than the involved area, or considered first treating the patient without SMT if possible. One of this group reported using only Activator Adjusting Instrument based techniques on this category of patient.

The reasons that a patient with cancer will visit a chiropractor were not considered to be different from those of any other patient namely: musculoskeletal pain/ conditions (12/13). One panellist reported that “*cancer patients seek chiropractic care for neurological complications af-*

fecting eyesight, balance, dizziness, autonomic nervous system complications and weakness”.

Additional comments made at the end of the first round included: “*most patients seek chiropractic treatment after the cancer was diagnosed*” and “*the aim should be the improvement of the function of the patient and that multidisciplinary patient centred approach could benefit patients with cancer*”.

Three of the panellists stated that chiropractors should not treat the cancer but address the neuro-musculoskeletal problems of the patient and help them by improving their function.

A further panellist stated: “*patients with cancer may benefit from chiropractors and a vitalistic approach as long as it is as part of multidisciplinary management. Contraindications must be considered and weeded out very carefully. Specific chiropractic spinal manipulation guidelines must be determined, and all of the healthcare providers must work together in a patient-centred manner*”.

The areas of concern raised by the panel included:

- *a lack of evidence*: 8 panellists considered there was insufficient evidence to support the safety of chiropractic on patients with cancer, whereas 2 considered that there was. Additionally, one panellist outlined that there is enough evidence for safe chiropractic care in special populations like osteoporotic patients as the worry was instability or bone weakening; therefore one could extrapolate that there would be a good safety record for cancer patients as well.
- *a lack of communication with the medical team*: part of the panel acknowledged that they do not communicate with the medical team (7/13). The situation with the remaining respondents (6/13) was not clear.
- *a fear of the misconception that chiropractic cures cancer instead of helping the neuro-musculoskeletal aspect of the symptoms associated with the disease or its treatment*. Throughout their comments the panellists were continually underlining the need of giving a clear message that the chiropractor would not cure the cancer but only help with the MSK symptoms associated with it.
- *a lack of specific chiropractic techniques other than spinal manipulation therapy*. Two of the thirteen

actively engaged panel members suggested soft tissue work, a further two stated there was nothing specific to chiropractic and seven gave no answer. Interestingly two panellists replied that they use SMT if indicated and would apply SMT in other areas of the body if required.

- *chiropractors should not advertise the benefits of their care.* One respondent said that such advertising was not legal in their country of practice, as new rules are limiting medical advertisement, whereas the others could not find any reason to target advertisements towards patients with cancer. In the comment field, two other panellists stated that chiropractors should not advertise any treatments specifically for cancer patients as either cancer patients are to be seen as any other patient with neuro-musculoskeletal problems or because an advertisement like that could “*make things worse*”. Two of the panellists responded in the comment field requesting this section be removed as there was no option not to answer.

Regarding whether chiropractic as a profession should do more to advertise the benefits of chiropractic on patients with cancer, two of the 12 who responded agreed and 10 disagreed. Reasons for disagreeing were that cancer patients are not and should not be a chiropractor’s primary patient (n=1), and there is insufficient evidence to claim that chiropractic could benefit these patients (n=1). Again, the comments focussed on the possibility of the message being misconstrued as being the chiropractor is able to cure cancer, instead of that chiropractic can help the MSK aspect of the patient’s problem.

Treatment modalities used for treating patients who have been given a cancer diagnosis

Regarding whether the presence of a bone tumour could be a contraindication to SMT: 9/13 agreed and 4/13 disagreed with the statement. Ruling out presence of metastases and osteoporotic regions was the main point of concern. Although there was consensus that SMT could be used, low force techniques were considered to be safer (n= 9). Additionally, comments from a panel member (n= 1) indicated there was insufficient information provided in this question, with the decision being dependent on the primary tumour location.

While the panel agreed that the SMT does not appear

necessary in the treatment plan of a patient with cancer (first round question, 13/13 agreed), the same degree of consensus did not exist when the panel were asked to suggest alternative treatment methods and comment on which would be considered specific to chiropractic. Two of the 13 answered that there is nothing specific to chiropractic, five out of 13 suggested soft tissue work, while one responded that the question was not clear. Respondents suggested the following to be alternative chiropractic specific therapies: dietary advice, adjustments of areas not affected by the cancer, use of Activator Adjusting instruments, active mediations, bio resonance, acupuncture, SOT, NUCCA, N.E.T., SSEP, trains of four, electrostimulation, Transcranial Magnetic Stimulation, balance training and eye exercises.

Protocol for treating patients who have been given a cancer diagnosis.

Although a large proportion of the respondents tended to agree on their approach regarding engagement with the medical team, there were some interesting differences within the group.

Many of the respondents (11/13) would not consider contacting the medical team of the patient to request permission to treat. However, one panellist stated they would contact the clinical team regardless of whether the patient was diagnosed with cancer, in chemo- or radio-therapy or in remission.

Approximately half of the respondents (7/13) considered that a clinical relationship between the chiropractor and the oncologist was not necessary, while six of 13 considered it to be necessary. Comments within the responses to this question showed some differences in terms of type of interaction. Two of nine who commented directly, stated that either oncologists are not open to chiropractic care in the country of practice (n= 1), or that the oncologist does not know what a chiropractor is or could do (n= 1).

Comments supportive of a multidisciplinary approach came from six of the 13 panel members. These are best encapsulated in the following statement: all healthcare practitioners working on a patient should have some clinical relationship for the benefit of the patient and that the patients’ optimal management is based on a mutual understanding of each practitioners’ role. Finally, 11/13 of the actively engaged panel agreed that a chiropractor should

offer treatment to a patient who has a current diagnosis of cancer; however, two disagreed.

Discussion

There was unanimous agreement of the panel regarding the perception that patients with cancer can benefit from chiropractic treatment. Interestingly, the main reasons that a patient with cancer seeks chiropractic treatment were considered by the panel to be no different from those of any other patient, namely MSK pain and associated disorders. A better quality of life, pain relief and improved function were reported to be the most common perceived benefits of chiropractic in relation to the panels' experience with cancer patients.

The panel agreed that a cancer diagnosis should make a difference to a chiropractic treatment plan, even if the patient seeks care when in remission. Spinal manipulative therapy was not reported as being used on all cancer patients, with exclusion criteria including the location of the tumour as well as presence of metastases or concurrent osteoporosis. Type of cancer was not mentioned as a factor by any of the panel, however, this might relate to the lack of a specific question.

One of the obvious limitations was that the panellists only had restricted clinical experience of patients with cancer, having only encountered them through their own practices. The potential lack of diversity in terms of the cancer types seen requires consideration when interpreting the comments reported here. The fact that these chiropractors have seen sufficient patients with these conditions to be comfortable discussing their treatment, however, does indicate that chiropractors should expect to see these patients in general practice.

The authors had initially considered a general questionnaire to the profession; however, a Delphi method was considered an appropriate starting place to gain some insight into the issue.

The Delphi method maximizes the benefits of using an expert/knowledgeable panel while minimizing potential disadvantages by implementing anonymity.⁴⁹⁻⁵¹ Furthermore, this method allows everything to be performed by email and does not require the participants to meet or interact directly. The presence of anonymity allowed those participating, the room to air their views without the inhibition that might result when discussing potentially contentious issues in a direct (face-to-face) social

interaction. This was an important consideration in relation to approaching this topic area within members of the chiropractic profession, in order to gather a wide range of views. Furthermore, anonymity allows decisions to be evaluated on their merit, rather than being influenced by the strength of personality (i.e. of the person who had proposed the idea). Anonymity and confidentiality of participants are central to ethical research practice in social research.⁵⁰⁻⁵³

Using the Delphi methodology rather than focus groups allowed information exchange between numerous geographically (and temporarily) dispersed individuals in an iterative process. The belief is that there could be benefits from the exchange of information while retaining a low cost and convenience of accessing the questionnaires. In this case, the method allowed chiropractors from across Europe to answer the questionnaires in their own time and without awareness of other panel members' views. Supplying their responses to a central point and not sharing them prevented any adverse personal interaction. This approach has been criticized for limiting the potentially positive aspects of interaction found in any face-to-face exchange of information, as these often help identify the reasons for any disagreements.⁵⁰ The preliminary basis of this study accepted this minor disadvantage in relation to the major advantage of determining the nature of the issues.

Consensus development methods are being used to help clinical guidelines, which define key aspects of the quality of health care.⁵² However, particularly appropriate indications/suggestions for interventions, such as those revealed in this Delphi study, do not represent any clinical guidelines. Instead, these results should only be considered as a representation of a consensus between members of a small panel of European chiropractors regarding their perspective on chiropractic management of patients with cancer.

Although 23 potential participants were invited, only one actively decided to opt out. Of the remaining 22 who indicated they were interested in participating, slightly more than half (n=13) actively participated in the first round. Reassuringly, this level of participation continued into the second round, however due to the success of the anonymization process we were not in a position to determine whether participation was by the same 13 chiropractors in both rounds. The low response rate during

the verification stages could be considered as reflecting a general agreement with the conclusions, however as this was not an active agreement, this can only be considered tacit approval at best.

Improvements in quality of life, pain relief and function were the most commonly reported perceived benefits of chiropractic in regard to patients with cancer. Importantly making potential patients more aware of these benefits was not considered appropriate. The debate in the profession regarding the “philosophy of chiropractic” seems to have made some chiropractors apprehensive regarding who they will talk to about chiropractic treatment in these patients, with the motivation apparently being a fear of possible misunderstanding about what the chiropractor could do. Indeed, when presenting our preliminary analysis at a major European chiropractic meeting one of the authors found that a number of chiropractic scientists misinterpreted the aim of the research. A small number of the panel expressed concerns about advertising any perceived benefits. Apart from local advertising restrictions and lack of evidence base, the main concern was that these patients should not be considered any differently from patients without a history of cancer, due to the treatment focus being neuro-musculoskeletal.

The panel agreed that chiropractors should view the patient as a “whole person” with needs reaching beyond the management of the disease entity. Indeed, the chiropractic profession has, ever since its inception, embraced such a “holistic” approach toward patient care. The generally accepted primary role of the chiropractor is to assist the patient with pain management and help the patient to increase mobility and function beyond a disease diagnosis.^{9,47} The panel did consider that the use of spinal manipulation might be contraindicated or require careful consideration when treating patients with cancer. When challenged regarding alternative management/treatment tools, the panel reported using a variety of tools, but only a few of them appeared to be chiropractic specific. The key feature was that each patient must be evaluated thoroughly to determine which methods (chiropractic or other) will provide the greatest benefit in the particular case. In some instances, treatment may call for non-force techniques, whereas other situations could be better addressed through use of more standard manipulative procedures.⁸ Interestingly, most of the techniques mentioned by the panel did not appear specific to chiropractic; as a variety of physical

therapists, physiotherapists, osteopaths and sport massage therapists would also consider them part of their toolbox. It was agreed by all the panel who expressed an opinion (n=13) that more evidence would be needed in order for chiropractic adjustments and chiropractic specific techniques to be considered safe to use with such patients.

Although the attitude of health care providers and regulators to chiropractic has been historically negative, the opinion of the consumers has always been positive. It appears the public’s opinion of chiropractors does not suffer because of advertising,⁴⁸ however it has been suggested that approval of the majority of clients can be helped by using a professionally designed and well-conceived advertising campaign. It has been reported that almost 77% of the general public seek and want information regarding the services a chiropractor provides.⁴⁸ This supports the need for clarity and transparency when communicating the identity for chiropractic: as we found here, what a chiropractor considers specific to chiropractic, may not be considered to be specific to chiropractors by those outside the profession.

The vast majority of the panel agreed that chiropractors should treat patients with cancer, which provides a positive answer to the initial question. However, there was a recognition of the need for evidence to indicate whether chiropractic treatment is safe for these patients which was one of the main concerns of the panel. In addition, the panel struggled to find chiropractic specific management techniques, which could raise an issue for further research.

Although anecdotal, there has been the perception of both fear and confusion in the profession regarding the role of the chiropractor in the management of patients with cancer. This was strongly reflected in the comments made by the panel. Therefore, going forward it is apparent that evidence will be needed in order to both allay fears, define roles and facilitate in the engagement of chiropractic as part of an integrated care package for these patients. This suggests there may be a need, at least initially, to create consensus based guidelines (as there is no research available to currently inform such guidelines) that support currently considered best practice and prevent more dubious and unhelpful claims of efficacy.

This research does not present evidence supporting benefits for patients with cancer from chiropractic care, or whether spinal manipulative therapy should be used on

the management of patients with a diagnosis of cancer. However, it does give evidence that experienced chiropractors both treat such patients and recognise a potential role for chiropractic in this population of patients.

Conclusions

Chiropractors treat patients who have cancer, seeking care mainly for neuro-musculoskeletal complaints. Advertising is not considered viable due to potential for adverse interpretation.

Further research is necessary regarding initially how chiropractic could gather data about the relative safety and risks of chiropractic care in such patients. Chiropractors need to establish better inter-professional relationships with the patient's medical and rehabilitation team.

It is important to send a clear message that chiropractors do not cure cancer but only aim to help with the neuro-musculoskeletal signs and symptoms. Therefore, construction and publication of consensus-based guidelines of best practice should be considered a priority.

Acknowledgements

The authors would like to acknowledge the time and contribution made by those chiropractors who responded, without which this study would not have been possible.

References

1. World Health Organization. Global status report on non-communicable diseases 2014. World Health Organization; Geneva, 2014.
2. Centers for Disease Control and Prevention. 24/7: Saving Lives. Protecting People.™ National Center for Chronic Disease Prevention and Health Promotion. Atlanta, GA: Centers for Disease Control and Prevention (US) (2013). Available from: www.cdc.gov/chronicdisease/about/public-health-approach.htm
3. Ferlay J, Soerjomataram I, Dikshit R, Eser S, Mathers C, Rebelo M, Parkin DM, Forman D, Bray F. Cancer incidence and mortality worldwide: sources, methods and major patterns in GLOBOCAN 2012. *Int J Cancer*. 2015 1;136(5):E359-386.
4. Bao Y, Kong X, Yang L, Liu R, Shi Z, Li W, et al. Complementary and alternative medicine for cancer pain: an overview of systematic reviews. *Evid Based Complement Alternat Med*. 2014;2014: 170396.
5. Nissen N, Pedersen CG, Lunde A, Johannessen H. The use of CAM after the completion of hospital treatment for colorectal cancer : findings from a questionnaire study in Denmark, *BMC Complementary Alternat Med*. 2014;14: 388.
6. Buckner CA, Lafrenie RM, Denommee JA, Caswell JM, Want DA. Complementary and alternative medicine use in patients before and after a cancer diagnosis. *Curr Oncol*. 2018; 25(4): e275-e281.
7. Evans RC, Rosner AL. Alternatives in cancer pain treatment: the application of chiropractic care. *Semin Oncol Nurs*. 2005;21:184-189.
8. Schneider J, Gilford S. Integration of complementary disciplines into the oncology clinic: part IV. The chiropractor's role in pain management for oncology patients. *Curr Probl Cancer*. 2000;24: 231-241.
9. Alcantara J, Alcantara JD, Alcantara J. The chiropractic care of patients with cancer: a systematic review of the literature. *Integr Cancer Ther*. 2011, 11: 304-312.
10. Menefee LA, Monti DA. Nonpharmacologic and complementary approaches to cancer pain management. *J Am Osteopath Assoc*. 2005;105: S15-S20.
11. Goldstein MS, Brown ER, Ballard-Barbash R, Morgenstern H, Bastani R, Lee J, et al. The use of complementary and alternative medicine among Californian adults with and without cancer. *Evid Based Complement Alternat Med*. 2005;2: 557-565.
12. Falkensteiner M, Mantovan F, Muller I, Them C. The use of massage therapy for reducing pain, anxiety, and depression on oncological palliative care patients: A narrative review of literature. *ISRN Nurs*. 2011;2011: 929686.
13. Scott JA, Kearney N, Hummerston S, Molassiotis A. Use of complementary and alternative medicine in patients with cancer: A UK survey. *Eur J Oncol Nurs*. 2005;9: 131-137.
14. Shakeel M, Newton JR, Bruce J, Ah-See KW. Use of complementary and alternative medicine by patients attending a head and neck oncology clinic. *J Laryngol Otol*. 2008;122: 1360-1364.
15. Lawsin C, DuHamel K, Itzkowitz SH, Brown K, Lim H, Thelemaque L, Jandorf L. Demographic, medical, and psychosocial correlates to CAM use among survivors of colorectal cancer. *Support Care Cancer*. 2007;15: 557-564.
16. Chrystal K, Allan S, Forgeson G, Isaacs R. The use of complementary/alternative medicine by cancer patients in a New Zealand regional cancer treatment centre. *N Z Med J*. 2003;116: U296.
17. Amin M, Glynn F, Rowley S, O'Leary G, O'Dwyer T, Timon C, Kinsella J. Complementary medicine use in patient with head and neck cancer in Ireland, *Eur Arch Otorhinolaryngol*. 2010;267: 1291-1297.
18. Roth M, Lin J, Kim M, Moody K. Pediatric oncologists' views toward the use of complementary and alternative medicine in children with cancer. *J Pediatr Hematol Oncol*. 2009;31:177-182.
19. Post-White J, Fitzgerald M, Hageness S, Sencer SF. Complementary and alternative medicine use in children with cancer and general and specialty pediatrics. *J Pediatr Oncol Nurs*. 2009;26: 7-15.

20. Fouladbakhsh JM, Stommel M, Given BA, Given CW. Predictors of use of complementary and alternative therapies among patients with cancer. *Oncol Nurs Forum*. 2005;32: 1115-1122.
21. Richardson MA, Sanders T, Palmer JL, Greisinger A, Singletary SE. Complementary/alternative medicine use in a comprehensive cancer center and the implications for oncology. *J Clin Oncol*. 2000;18: 2505-2514.
22. Habermann TM, Thompson CA, LaPlant BR, et al. Complementary and alternative medicine use among long-term lymphoma survivors: a pilot study. *Am J Hematol*. 2009;84: 795-798.
23. Yildirim Y. Patterns of the use of complementary and alternative medicine in women with metastatic cancer. *Cancer Nurs*. 2010; 33: 194-200.
24. McEachrane-Gross FP, Liebschutz JM, Berlowitz D. Use of selected complementary and alternative medicine treatments in veterans with cancer or chronic pain: a cross sectional survey. *BMC Complem Alternat Med*. 2006;6: 34.
25. Curtis P, Gaylord S. Safety issues in the interaction of conventional, complementary and alternative health care. *J Evid Based Integr Med*. 2005;10 (1): 3-31.
26. Demetrius J, Demetrius GJ. Lung cancer metastasis to the scapula and spine: a case report. *Chiropr Osteopat*. 2008;16: 8.
27. French S D, Charity M J, Forsdike K, Gunn J M, Polus B I, Walker B F, Chondros P and Brit H C. Chiropractic Observation and Analysis Study (COAST): providing an understanding of current chiropractic practice. *Med J Aust*. 2013;199(10): 687-691.
28. Coulter ID, Hurwitz EL, Adams AH, Genovese BJ, Hays R, Shekelle PG. Patients using chiropractors in North America: who are they, and why are they in chiropractic care? *Spine*. 2002;27(3): 291-296.
29. Kier A, Timchur MD, McCarthy PW. A case report of an uncommon cause of cauda equina symptoms. *J Manipulative Physiol Ther*. 2007;30: 459-465.
30. Bussi eres A, Cassidy JD, Dzus A. Spinal cord astrocytoma presenting as torticollis and scoliosis. *J Manipulative Physiol Ther*. 1994;17:113-118.
31. Conley RN, Longnecker R. Metastatic carcinoma. *ACA J Chiropr*. 1983;17: 71-74.
32. Gatterman B. Bronchogenic carcinoma with metastasis of the cervical spine. *ACA J Chiropr*. 1987;24: 61-62.
33. Grod JP, Crowther ER. Metastatic bone disease secondary to breast cancer: an all too common cause of low back pain. *J Can Chiropr Assoc*. 1994;38: 139-145.
34. Malone D. Diffuse metastatic disease of the lumbo-sacral spine mimicking a lumbar disc herniation. *Clin Chiropr*. 2004;7: 10-15.
35. Kenny M. Bronchogenic carcinoma presenting as neuromusculoskeletal pain. *J Manipulative Physiol Ther*. 1991;14: 440-442.
36. Jensen A, Nolet PS, Murtaza AD. Oral squamous cell carcinoma: an atypical presentation mimicking temporomandibular joint disorder. *J Can Chiropr Assoc*. 2004;48: 266-272.
37. Osterhouse MD, Kettner NW, Birrer PT, Mankamyer K. Ganglioneuroma masquerading as spinal pain. *J Manipulative Physiol Ther*. 2002; 25: 184-187.
38. Yochum TR, Lile RL, Schultz GD, Mick TJ, Brown CW. Acquired spinal stenosis secondary to an expanding thoracic vertebral hemangioma. *Spine*. 1993;18: 299-305.
39. Stein PJ. A case of cerebellopontine angle meningioma presenting with neck and upper extremity pain. *J Manipulative Physiol Ther*. 2009;32: 776-780.
40. Farrar KL, Gardiner L. Unresolving hip tendonitis leads to discovery of malignant tumor. *J Manipulative Physiol Ther*. 2003;26: 207.
41. Shafir Y, Kaufman B. Quadriplegia after chiropractic manipulation in an infant with congenital torticollis caused by a spinal cord astrocytoma. *J Pediatr*. 1992;120: 266-269.
42. Lennington RB, Laster DW, Moody DM, Ball MR. Traumatic pseudoaneurysm of ascending cervical artery in neurofibromatosis: complications of chiropractic manipulation. *AJNR*. 1980; 269-270.
43. Brown R. Spinal health: the backbone of chiropractic's identity. *J Chiropr Humanit*. 2016;23(1): 22-28.
44. Lafferty WE, Bellas A, Corage Baden A, Tyree PT, Standish LJ, Patterson R. The use of complementary and alternative medical providers by insured cancer patients in Washington State. *Cancer*. 2004;100(7): 1522-1530.
45. Hamish R. Smith. Depression in cancer patients: pathogenesis, implications and treatment. *Oncol Lett*. 2015; 9(4): 1509-1514.
46. Pinheiro MB, Ferreira ML, Refshauge K, Ordo ana JR, Machado GC, Prado LR, et al. 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.
47. Rosner AL. Chiropractic identity: a neurological, professional, and political assessment. *J Chiropr Humanit*. 2016; 23(1): 35-45.
48. Moser RH, Freeman GL. An empirical analysis of the public's attitudes toward advertising chiropractic services: a comparative cross-sectional study. *J Med Marketing*. 2017; 16(1);10-20.
49. Fink A, Kosecoff J, Chassin M et al. Consensus methods: characteristics and guidelines for use. *Am J Public Health*. 1984; 74: 979-983.
50. Hasson F, Keeney S, McKenna H. Research guidelines for the Delphi survey technique. *J Adv Nurs*. 2000;32: 1008-1015.
51. Dalkey NC. The Delphi method: an experimental study of group opinion. In: Dalkey NC, Rourke DL, Lewis R, Snyder D., eds. *Studies in the quality of life: Delphi and*

- decision-making. Lexington, MA: Lexington Books, 1972:13–54.
52. Schneider P, Evaniew N, Rendon JS, McKay P, Randall R, Turcotte R, Velez R, Bhandari M, Gher M. Moving forward through consensus: protocol for a modified Delphi approach to determine the top research priorities in the field of orthopaedic oncology. *BMJ Open*. 2016; 6(5) e011780.
53. Woolf S, Schünemann H, Eccles M, Grimshaw J, Shekelle P. Developing clinical practice guidelines: types of evidence and outcomes; values and economics, synthesis, grading, and presentation and deriving recommendations. *Implement Sci*. 2012; 7: 61.

Clinical management of benign joint hypermobility syndrome: a case series

Patricia Adele Boudreau, BSc, DC¹

Igor Steiman, BSc, DC, MSc, FCCS(C)²

Silvano Mior, DC, FCCS(C), PhD³

Background: *Benign Joint Hypermobility Syndrome (BJHS) is a relatively prevalent condition of the spectrum of heritable connective tissue disorders, with musculoskeletal, visceral and psychological manifestations. The conservative management of the musculoskeletal symptomatology must be modified for optimal effectiveness and minimal sequelae.*

Purpose: *To provide an overview of the presentation, assessment, chiropractic management, and outcomes of patients with BJHS.*

Study Design: *Case series*

Discussion: *Recognizing joint hypermobility as a significant contributing factor in patients presenting with musculoskeletal complaints is often challenging. The lack of awareness of BJHS may delay the diagnosis as well as effective management. Manual therapy should be used judiciously; active exercise is an essential element of care. We provide an overview of the presentations,*

Contexte : *Le syndrome d'hypermobilité articulaire bénigne (SHAB) est une affection relativement répandue faisant partie des maladies héréditaires du tissu conjonctif, qui se caractérise par des manifestations musculosquelettiques, viscérales et psychologiques. Il faut changer le traitement conservateur des troubles musculosquelettiques pour optimiser son efficacité et réduire le plus possible les séquelles.*

Objectif : *Donner un aperçu des manifestations du SHAB, de l'évaluation des symptômes, des traitements chiropratiques et des résultats obtenus chez des patients atteints du SHAB.*

Méthodologie : *Série de cas*

Discussion : *Il est souvent difficile de déterminer si l'hypermobilité articulaire est un important facteur contributif chez les patients atteints de troubles musculosquelettiques. Le SHAB étant une maladie méconnue, le diagnostic et une prise en charge efficace risquent d'être retardés. La thérapie manuelle doit être utilisée judicieusement; l'exercice actif est une composante essentielle du traitement. Nous présentons un aperçu des manifestations, de l'évaluation des symptômes, des traitements chiropratiques et des*

¹ Private practice

² Department of Clinical Education, Canadian Memorial Chiropractic College

³ Department of Research and Innovation, Canadian Memorial Chiropractic College

Corresponding author: Adele Boudreau, 189 Barrieau Rd. Moncton NB, E1G 1J9

E-mail: aboudreau@pthealth.ca

© JCCA 2020

The authors have no disclaimers, competing interests, or sources of support or funding to report in the preparation of this manuscript. The involved patients provided consent for case publication.

assessment, chiropractic management, and outcomes of three patients with BJHS. Future clinical trials are necessary to determine effective clinical management strategies for patients with BJHS.

(JCCA. 2020;64(1):43-54)

KEY WORDS: joint hypermobility, connective tissue disorder, manipulation, chiropractic

résultats obtenus chez trois patients atteints du SHAB. Il faudrait effectuer des essais cliniques pour trouver des stratégies de prise en charge efficaces chez les patients atteints du SHAB.

(JCCA. 2020;64(1):43-54)

MOTS CLÉS : hypermobilité articulaire, trouble du tissu conjonctif, manipulation, chiropratique

Introduction

Hypermobile joints can be a consequence of a number of heritable connective tissue disorders. One such disorder is benign joint hypermobility syndrome (BJHS). BJHS is characterized by generalized ligamentous laxity and the presence of musculoskeletal pain without signs of systemic rheumatologic disease.¹⁻⁵ Many experts suggest that BJHS be considered part of a hypermobility spectrum as a milder form of the hypermobility type Hyper-

mobile Ehlers-Danlos Syndrome (hEDS), consisting of hypermobility and arthralgia in several joints.⁶ The main distinguishing factors between these two conditions are the scores on the Brighton Criteria as well as laboratory tests.^{5,7} The diagnostic Brighton Criteria characterize hypermobility findings into “major” and “minor” categories (Table 1).⁷ Within these criteria is the Beighton Score, a scoring system utilized in the diagnosis of a hypermobility syndrome, to quantify the extensiveness

Table 1.

Revised diagnostic criteria for benign joint hypermobility syndrome (BJHS).⁷

BJHS is diagnosed in the presence of two major criteria, or one major and two minor criteria, or four minor criteria.

Major Criteria
<ol style="list-style-type: none"> 1. A Beighton Score of 4/9 or greater (currently or historically) 2. Arthralgia for 3 months in 4 or more joints
Minor Criteria
<ol style="list-style-type: none"> 1. A Beighton score of 1, 2 or 3/9 (0, 1, 2, or 3 if aged 50+) 2. Arthralgia (≥ 3 months) in 1-3 joints, or back pain ≥ 3 months, spondylosis, spondylolysis/spondylolisthesis 3. Dislocation/subluxation in one or more joints or in one joint on more than one occasion 4. Soft tissue rheumatism ≥ 3 lesions (e.g., epicondylitis, tenosynovitis, bursitis) 5. Marfanoid habitus (tall, slim, span/height ratio >1.03, upper:lower segment ratio <0.89, arachnodactyly [+Steinberg/wrist signs]) 6. Abnormal skin: striae, hyperextensibility, thin skin, papyraceous, or scarring 7. Eye signs: drooping eyelids or myopia or antimongoloid slant 8. Varicose veins, hernia/rectal prolapse

of hypermobility in several predetermined articulations (Table 2).⁵

BJHS is a relatively common phenomenon with a prevalence ranging from 5% to 18% in Caucasian populations^{8,9} and up to 43% in non-Caucasian populations¹⁰. Younger individuals express a greater degree of joint laxity, which decreases with age.^{10,11} There is an approximately 2:1 female to male relative prevalence of BJHS.¹² Therefore, a small percentage of the population presenting to a chiropractor's office may present with findings suggestive of BJHS. This is important, considering that the majority of chiropractors use spinal manipulation in management of joint pain and function.

The restoration of joint mobility and function are typical goals of chiropractors and other manual therapists. While spinal manipulative therapy is a common therapeutic approach, it may not be appropriate for all patients presenting with hypermobile joints which, in theory, should not require treatments intended to impart increased mobility to articulations. The lack of obvious extra-articular signs can present a diagnostic challenge to a manual therapist, complicating the management of what otherwise may appear to be a straightforward case of mechanical pain. Despite a significant amount of research on BJHS, it remains insufficiently identified, inadequately understood and poorly managed by health practitioners.¹³ Therefore, the purpose of our paper is to provide an overview of the presentation, assessment, management and outcomes of three cases of patients presenting with BJHS who sought chiropractic care. The case presentations are discussed in light of the current literature about BJHS.

Case Presentations

Case 1

A 26-year-old Caucasian female chiropractic student was evaluated for chronic, intermittent low back and left lower extremity pains. She attributed the onset to a fall off a swing during childhood and subsequent aggravation by a motor vehicle collision five years prior to presentation. The progressive constant, dull, aching pain was localized to the left sacroiliac region and radiated distally to the posterolateral aspect of the left thigh; she denied radiation past the knee. The intensity of the pain ranged from 3–10/10 in intensity on a verbal pain rating score. It was aggravated by prolonged sitting, cycling, crossing her

Table 2.
*Nine-point Beighton score for joint hypermobility.*⁵

Description	Bilateral Testing	Scoring (maximum points)
Passive dorsiflexion of the fifth metacarpophalangeal joint to ≥ 90 degrees	Yes	2
Passive hyperextension of the elbow ≥ 10 degrees	Yes	2
Passive hyperextension of the knee ≥ 10 degrees	Yes	2
Passive apposition of the thumb to the flexor side of the forearm, while shoulder is flexed 90 degrees, elbow is extended, and hand is pronated	Yes	2
Forward flexion of the trunk, with the knees straight, so that the hand palms rest easily on the floor	No	1
Total		9

legs, fatigue, and positioning for side-posture lumbar manipulations in chiropractic technique class. She obtained some relief by exercising, walking and resting. Three months of chiropractic care had given inconsistent results; she would typically feel sore for hours after manipulation for her low back pain, followed by a short period of relative improvement before the pain would return.

She reported a number of prior musculoskeletal complaints, particularly in her knees, feet and shoulders. Otherwise, she felt she was in good health. She had been a high calibre athlete but since attending chiropractic college her conditioning had decreased significantly.

On examination, no obvious postural deviations were noted. Her lumbar range of motion was mildly painful in forward flexion, reproducing the left thigh pain. All other back ranges of motion were pain-free and appeared significantly greater than normal. External rotation of the left hip was limited to 50% of normal by pain. Straight leg raising was pain-free at 110 - 120 degrees, bilaterally. No neurological deficits were noted. Palpation revealed ten-

derness and decreased mobility at the right C4-5, T5-6, T12-L1, L4-5 and left sacroiliac articulations. Tenderness was palpated in the erector spinae and quadratus lumborum musculature bilaterally, and the left gluteal and tensor fasciae latae musculature. Joint flexibility using the Beighton Score was scored by the chiropractor at 9/9, including findings of mild genu recurvatum, marked hyperextension of both elbows and the ability to passively appose each thumb to the anterior forearm. Lumbar spine radiographs, including AP, lateral and lumbosacral spot views, revealed mild postural changes with a mild left lateral list of the lumbar spine, but were otherwise unremarkable.

The patient was diagnosed with BJHS and left sacroiliac joint dysfunction. The plan of management included ergonomic modification when sitting in class, restriction from involvement as a training partner in technique class, soft tissue therapy to relieve the myofascial component of her complaint, an exercise program directed at improving strength, flexibility and endurance, and limited spinal manipulation. The focus was to limit the passive treatment component and encourage the active component of care. The importance of compliance with the plan of management was impressed upon the patient.

Approximately two months later, the patient reported she was much improved. Seven years later, she reported that she was experiencing occasional low back pain as a consequence of clinical practice, but these episodes were relieved by specific manipulation. She also reported that her symptoms would be aggravated by a lack of physical activity.

Case 2

A 23-year-old Caucasian female presented with left-sided spinal pain, extending from the base of the skull to approximately T8, that had been present for roughly two months. She denied any precipitating event but reported an extensive history of similar complaints. She rated the dull, aching pain as 5/10 in intensity, worse in the evenings, and occasionally present in the mornings. If the pain began in the morning, it would typically persist for the rest of the day. She reported aggravation by sitting and lying supine, and described no relieving factors.

The patient also reported “clicking” in the hips, eye pain with prolonged reading, left jaw pain, and dry skin on the backs of her legs. She reported a past history of a fall

when skiing four years prior. No other significant medical history was elicited. She described her life as stressful due to her schooling, which involved a great deal of desk and computer work. She was attempting to improve her diet and had started an aerobic exercise program (three days weekly). She had received no treatment of any kind for her current or previous episodes.

Physical examination revealed a young woman in no significant distress. She was neurologically intact. No cervical bruits were present. Gross range of motion of the cervical spine was decreased in extension-rotation bilaterally, provoking the neck pain of chief complaint. Thoracic range of motion was restricted in extension-rotation and left rotation by pain at the T6 - T9 region. Spinous palpation was extremely tender at T5-6. Tenderness was present in the trapezius and levator scapulae musculature, primarily on the left. Deep inspiration provoked mid-back pain.

The patient scored 8/9 on the Beighton Score. During mobility testing, both elbow joints gave a popping sound upon gentle hyperextension. The patient reported that this was normal for her. No remarkable skin extensibility was observed. No radiographs were taken.

The patient was diagnosed with BJHS, and thoracic facet irritation with myofascial strain secondary to postural strain. Treatment included manipulation directed to the mid-thoracic spine and soft tissue therapy directed at the upper thoracic and cervical musculature. The patient was given a strengthening routine using weight machines and was encouraged to continue aerobic exercise.

She was treated four times over the course of nine days and reported good resolution of her complaint. She had started the prescribed exercise program and reported some mild muscular stiffness. She declined to attend a two-week follow-up appointment because she felt well.

Case 3

A 23-year-old Caucasian female was evaluated for complaint of neck and back pain that started insidiously approximately three years prior. She described the pain as diffuse throughout the upper and lower back. She also reported frequent “cracking” of her joints. Her neck felt stiff and she experienced sharp pain with movements such as rotation and extension. Aggravating factors included carrying bags and lifting heavy objects. She rated the pain in the upper cervical spine at 5-6/10, lower cervical spine

at 5-7/10 and thoracic spine at 8-8.5/10 in intensity. She had no previous chiropractic treatment for this condition. She used muscle relaxants, massage and frequent “self-adjusting” for relief. The patient scored a 21/45 (moderate perceived disability) on the Neck Disability Index.

Past medical history included panic attacks, anxiety attacks and depression. She typically slept a few hours without sleep aid medication and approximately five hours when taking a sleep aid. She rated her current stress level, which disrupted her sleep, as “severe”. Her current medications were clonazepam, Adderall XR, Ativan and Wellbutrin.

Physical examination revealed a patient who was 5’11” tall and weighed 120 lbs (i.e., a tall ectomorph), with arachnodactyly. The patient’s skin appeared to be slightly more extensible than other patients her age, but did not have a velvety texture. She scored 3/9 on the Beighton Score. However, she actively demonstrated an ability to sublaxate her glenohumeral joints bilaterally and bring her heel to her hip posteriorly while in extreme hip internal rotation. Her finger extension and elbow extension were within normal limits; genu recurvatum was not evident. No skin lesions, muscular atrophy or scoliosis were observed. Postural examination revealed the right shoulder was slightly lower than the left.

Cervical spine flexion was mildly increased and caused a pulling sensation in the thoracic spine. Extension was mildly limited initially, recreating the patient’s pain, but she was able to proceed to full extension, causing pain in the interscapular region. Bilateral lateral flexion was moderately increased and did not elicit pain. Similarly, right rotation was mildly increased and did not cause pain. Left rotation was within normal limits and recreated the thoracic spine pain of chief complaint. Thoracic active range of motion was within normal limits but extension caused interscapular pain and bilateral rotation caused a pulling sensation. Flexion and bilateral lateral flexion were unremarkable.

Orthopaedic testing¹⁴ revealed left cervical Kemp’s test caused a pressure sensation in the cervicothoracic junction and lumbar Kemp’s test bilaterally causing low back pain. Left-sided Jackson’s test caused the patient’s pain of chief complaint in the cervicothoracic junction. Left cervical doorbell test caused ipsilateral interscapular referral but only caused local pain when performed on the right side. The following orthopaedic tests were unremarkable:

right-sided cervical spine Kemp’s, cervical spine neutral compression, Spurling’s, and right-sided Jackson’s. Auscultation of the heart did not reveal any abnormal rhythm or sounds.

The patient was diagnosed with a cervicothoracic strain, as well as BJHS according to the revised diagnostic criteria for BJHS (Table 1) given that she had one major criterion (arthralgia for three months in four or more joints) and two minor criteria (Marfanoid habitus; skin hyperextensibility). The plan of management included soft tissue therapy to affected muscles, spinal manipulative therapy to hypomobile segments, and strengthening exercises. The proposed frequency of care was two to three times per week for six weeks; however, due to other health issues the patient only attended 12 of the 16 recommended visits. The patient’s presenting complaint had improved but psychological issues had increased. She also suffered from numerous viral illnesses, which limited her ability to attend treatment. Both of these factors limited the success of her treatment.

Discussion

The above case presentations can be encountered by chiropractors and other manual therapists in clinical practice. Each of these female Caucasian patients, all in their twenties, had histories of a variety of musculoskeletal complaints over a number of years. They had orthopaedic signs of mechanical joint pain with no apparent neurological deficits nor overt features of an arthritic or other systemic pathology. In Case 1, the history of lack of long-term benefit from chiropractic manipulation cued the clinician to re-evaluate the patient and treatment plan, resulting in a more specific diagnosis and an effective treatment plan that yielded excellent results. In both Cases 2 and 3, the lack of an apparent etiology and the long history of similar complaints led the clinician to evaluate for hypermobility in the physical examination. However, all patients demonstrated joint laxity according to the revised Brighton Criteria.⁷

The Brighton Criteria categorize hypermobility findings into “major” and “minor” categories (Table 1). The Beighton Score is a scoring system commonly utilized in the diagnosis of a hypermobility syndrome to quantify the extensiveness of hypermobility in several predetermined articulations (Table 2).⁵ It is a measure of articular laxity¹¹ that incorporates a composite score based on



Figure 1. *Elbow hyperextension*



Figure 2. *Finger hyperextension*



Figure 3. *Thumb hyperextension*

passive elbow hyperextension beyond 10 degrees (Figure 1), passive finger hyperextension (Figure 2), passive thumb apposition to the anterior surface of the forearm (Figure 3), passive knee hyperextension (Figure 4), and the ability to place both palms on the floor while standing (Figure 5) to rate generalized joint laxity. Each of these criteria receives a score of one with a total possible score of nine. A patient is considered to be hypermobile if the score is four or greater. The cut-off of 4/9 is arbitrary but is commonly used in the literature.¹¹ Some authors have advocated measuring only the non-dominant side (giving a maximum score of five) to avoid joints that may be lax due to an exercise training effect.¹⁵

Cases 1 and 2 scored 9/9 and 8/9 on the Beighton Score, respectively. Case 3 scored a 3/9 on the Beighton Score, but the patient demonstrated joint laxity in her glenohumeral and femoro-acetabular joints, which are not included as part of the Beighton Score. Case 3 demonstrated one major criterion (arthralgia for three months in four or more joints) and two minor criteria (Marfanoid habitus; skin hyperextensibility), which therefore meets the Brighton Revised Criteria⁷ for BJHS (Table 1).

Both the Beighton Score and Brighton Criteria for BJHS have been examined and found to demonstrate good-to-excellent inter-examiner reproducibility.¹⁶ Another measure of joint hypermobility, not applied in this case series, is a



Figure 4. *Knee hyperextension*



Figure 5. *Trunk forward flexion*

Table 3.
Clinical signs of Marfan syndrome and Ehlers-Danlos syndrome.*

Syndrome	Clinical signs	
Marfan Syndrome	Arachnodactyly Aortic root dilatation Positive family history	Ectopia lentis Dolichosternomelic habitus Scoliosis, anterior chest deformity Mitral valve prolapse
Classical Ehlers-Danlos Syndrome (cEDS)	Joint hypermobility Bruising/tissue friability	Skin hyperextensibility (velvety texture) “Cigarette paper” scars
Hypermobile EDS (hEDS)	Joint hypermobility	Mild skin hyperextensibility
Vascular EDS (vEDS)	Translucent skin Joint laxity in the hand Bowel and uterine rupture	Prominent bruising Relatively normal large joint mobility Mildly extensible skin
Kyphoscoliotic EDS (kEDS- PLOD1/kEDS-FKBP14)	Ocular fragility	Soft velvety hyperextensible skin
Arthrochalasia EDS (aEDS)	Marked joint laxity	Soft skin Congenital hip dislocation
Periodontal EDS (pEDS)	Easily bruised fragile skin Abundant scarring	Progressive periodontal disease (loss of teeth in second or third decade)

Adapted from: Malfait F, Francomano C, Byers P, Belmont J, et al. The 2017 International classification of the Ehlers-Danlos syndromes. *Amer J Med Genetics* 2017; 175c (1): 8-26.

five-item questionnaire by Hakim and Grahame (Table 4). This questionnaire can be useful for the clinician to incorporate in their initial history to screen for BJHS. Answering “yes” to two or more of these questions suggests hypermobility with sensitivity of 85% and specificity of 90%.¹⁷

As in these cases, when generalized hypermobility is combined with myalgia of over three months’ duration, the criteria for BJHS have been met (Table 1).¹⁸ If BJHS is considered, then alternative causes of generalized joint laxity should first be ruled out: heritable connective tissue disorders such as Marfan syndrome, Ehlers-Danlos syndrome, Stickler’s syndrome, Larsen syndrome and osteogenesis imperfecta.¹⁹ Generalized laxity is a prominent finding in such patients but, unlike those with BJHS, they present with significant cardiovascular, skin, bone and eye abnormalities.²⁰ Common features of Marfan and Ehlers-Danlos syndromes are listed in Table 3. The skin may be stretchy in BJHS similarly to Ehlers-Danlos syndrome, and can manifest as eyelid laxity or drooping eyelids. However, the skin in patients with BJHS lacks

the velvety texture and reduced thickness that is seen in Ehlers-Danlos syndrome.²⁰

A recent study²¹ reviewed the controversy regarding the association of joint hypermobility and osteoarthritis, and added new data pointing away from such an associ-

Table 4. *Five-point hypermobility questionnaire.*¹⁸

1. Can you now (or could you ever) place your hands flat on the floor without bending your knees?
2. Can you now (or could you ever) bend your thumb to touch your forearm?
3. As a child, did you amuse your friends by contorting your body into strange shapes or could you do the splits?
4. As a child or teenager, did your kneecap or shoulder dislocate on more than one occasion?
5. Do you consider yourself “double-jointed”?

Answering yes to 2 or more of these questions suggests hypermobility with sensitivity of 85% and specificity of 90%.

ation. Significant extra-articular manifestations of BJHS²² include autonomic disturbances²³, skin fragility²⁴, easy bruising^{25,26}, ocular ptosis, varicose veins²⁰, Raynaud's phenomenon²⁷, urogenital prolapses^{28,29}, developmental motor co-ordination delay (DCD)³⁰, alterations in neuromuscular reflex action³¹, fibromyalgia³², carpal and tarsal tunnel syndromes and neuropathies^{33,34}, lower bone density^{20,35}, depression³⁶, anxiety and panic attacks³⁷, lumbar disc herniations³⁸, and gastrointestinal symptoms³⁹. Case 3 included common extra-articular features of BJHS in the clinical history, reporting anxiety, panic attacks and depression. There appears to be a relationship between joint hypermobility and a higher risk of developing an anxiety disorder.⁴⁰ Recent investigations found structural differences between individuals with and without joint hypermobility: joint hypermobility has been associated with a greater bilateral amygdala volume - the key emotion-processing region of the brain.⁴¹ The association likely reflects the genetic basis for BJHS.⁴²

BJHS has been demonstrated to be a genetically inherited disorder containing a strong genetic component with an autosomal dominant pattern, believed to affect the encoding of the connective tissue collagen's protein.²⁴ Nevertheless, there are currently no laboratory tests to diagnose BJHS, as in other systemic disorders⁴³, only the "Revised diagnostic criteria for BJHS" (Table 1). Many experts now consider the commonalities in symptoms between BJHS and the hypermobility type Hypermobile Ehlers-Danlos Syndrome (hEDS) to be indistinguishable and suggest that BJHS is part of a hypermobility spectrum, as the mildest form of the hypermobility type Ehlers-Danlos.⁶

The primary underlying cause of joint hypermobility is the ligament laxity, which mainly determines the maximum range of motion.⁴⁴ Early studies utilizing electron microscopic evaluation of ligaments of patients with BJHS demonstrated reduced thickness of collagen fibrils.⁴⁵ It has been also proposed that individuals with BJHS have an abnormal ratio of type III to type I collagen.² Type I collagen, containing a high tensile property, is the most common collagen in the body. Type III collagen, located mainly in organs such as the gut, skin and blood vessels, is much more extensible.⁴⁶ Furthermore, patients with BJHS demonstrate significantly lower activity of prolydase, an enzyme influential in the collagen structure of ligaments, suggesting an altered collagen metabolism causing joint hypermobility.⁴⁷

Research has also evaluated the symptomatic overlap of fibromyalgia (FM) and BJHS^{32,48} in both adults and children⁴⁹. While the underlying mechanism for pain hypersensitivity in FM has been extensively evaluated, there is little knowledge regarding the enhanced sensitization to pain in BJHS.⁵⁰ Individuals with FM have an overall lower threshold to pain, attributed to a form of central sensitization or small-fibre polyneuropathy in response to repeated noxious stimulation.^{51,52} Conversely, in BJHS, it is hypothesized that pain is a result of repeated microtrauma from abnormal joint hypermobility that contributes to chronic arthralgia.³⁶ It is also theorized that related structural differences in emotion-processing systems may cause individuals with hypermobility to have a heightened susceptibility to (threat of) pain and/or a perturbation of autonomic control.⁴¹ Interestingly, variations in emotion-processing systems also occur in other pain disorders including FM, irritable bowel syndrome and complex regional pain syndrome.⁵³ Moreover, women with FM are 44% more likely to be hypermobile.⁵⁴ In keeping with the female predominance reported in the literature, all three of our cases were female.

In addition to sex, ethnicity and age are significant factors in the occurrence of BJHS, with increased prevalence among individuals of Asian and African descent.^{55,56} In the adolescent and child populations, BJHS tends to occur with juvenile fibromyalgia (JFM).⁵⁷⁻⁵⁹ Exploring the patient's childhood history of joint hypermobility and repeated injuries may be clinically beneficial in identifying adults with BJHS, as seen in the Hakim and Grahame¹⁷ five-item questionnaire (Table 4). Similarly, if a female patient has a clinical history of FM, evaluating joint hypermobility may be beneficial with respect to clinical management.

The sports injury literature demonstrates no clear relationship between generalized joint laxity and injury.^{60,61} It remains unclear if individuals with BJHS are at increased risk of injury compared to their non-hypermobility counterparts. Studies have demonstrated a higher risk of injury in military recruits and ballet dancers who were identified as lax or very lax.^{62,63} In contrast, Krivickas and Feinberg⁶⁴ found that hypermobile male athletes (Beighton 4-6/9) had a 66% lesser chance of injury than their less mobile colleagues. However, no difference in overall injury rates was observed in NCAA lacrosse players, though hypermobile athletes showed an increased rate of

ankle injuries.⁶⁵ More recent reports have found increased injury rates in hypermobile (vs. “tight”) rugby players⁶⁶, netball players (ankle, knee and finger injuries)⁶⁷, and soccer players⁶⁸. Thus, the consensus is building about risk from hypermobility in the athletic population.

The athletic population, in any case, may not be an ideal comparison for the general population for two reasons. One is that strengthening of musculature around the joint may aid the dynamic stability of joints in the trained individual.^{64,69} The second is that exercise may improve joint proprioception⁷⁰, which appears decreased at specific joint angles in hypermobile individuals when compared to controls^{70,71}. Vigorous exercise in relatively deconditioned individuals may lead to injury⁷² but may have little effect on injury rates in highly trained individuals, as athletes have enhanced proprioceptive abilities when compared to non-athletes⁷³.

Consideration of the role of exercise and the potential risks of high impact activity in deconditioned individuals has implications for exercise prescription in patients with BJHS. A graded increase in activity was beneficial in the presented cases and seems appropriate for deconditioned patients with BJHS. Recently, a study by Celenay and Kaya⁷⁴ demonstrated that a spinal stabilization program can decrease pain complaints, and improve postural stability and muscle endurance in women with BJHS. An interesting perspective on the role of exercise in the management of hypermobile patients is our first case, in which a highly trained athlete became progressively deconditioned and had an increase in symptomatology. Vigorous activity may be relatively contraindicated in hypermobile individuals^{58,62} but this restriction might be lifted once a sufficient training effect has been achieved. Overall, maintenance of physical fitness is imperative for managing symptoms of BJHS, especially activities that are focused on neuromusculoskeletal control; e.g., swimming, Tai Chi, pilates, yoga and dance.⁷⁵

The management of patients with BJHS can be challenging for the patient as well as the practitioner. As described by Simmonds and Keer⁷⁵, “patience, coupled with good communication and sensitive handling skills are required as physical problems are often longstanding and include secondary complications and psycho-social issues.” There is no conclusive evidence in the literature regarding best practices for patients with BJHS. However, one report highlights the importance of patient edu-

cation, therapeutic exercise, and modification of work and lifestyle in the management of BJHS⁷⁶, reflected in our cases. Patients may also be advised specifically on rest and pacing activities, and have benefited from treatment with modalities including ultrasound and transcutaneous nerve stimulation, taping and splinting, or wearing firm fitting clothing to improve perceived joint stability.⁷⁵ Although a recent review suggests that such passive treatment modalities may be ineffective in the management of neck pain and associated disorders⁷⁶, it is unclear if such findings are also pertinent to hypermobile patients based on the existing science.

Patients should be provided realistic expectations since their recovery and healing is often slower than in their non-hypermobile counterparts³⁶ by the time required to improve joint proprioception and strength. In the three cases presented above, the goal of management shifted to the protection of the joints by emphasizing an active exercise program to increase endurance and strength. After seven years, the patient in Case 1 maintained her excellent results as she continued her exercise regime, being able to participate in relatively high impact activities, such as hiking and backpacking.

It may seem paradoxical to apply manipulation, a treatment intended to impart mobility to articulations⁷⁷, in patients with BJHS. However, joint dysfunctions were detected and treated with manipulation in the cases described above. The judicious application of high velocity, low amplitude (HVLA) spinal manipulation appears to have benefited these three patients. Currently, only one other study describes a similar successful treatment of a patient with BJHS using a multimodal approach including HVLA spinal manipulations.⁷⁶ However, the potential for concomitant decreased pain thresholds in individuals with ligament laxity^{41,76} supports our clinical experience that hypermobile individuals seem to report soreness after physical therapies, such as massage or manipulation, more often than non-hypermobile individuals. Modified techniques, including modification of the application of pressure and force, should be considered when treating hypermobile patients after informing them of this possibility.

The management of BJHS is complex and pain management can be difficult in most cases. Prolotherapy is an alternative therapy, not explored in the three cases presented, which can be considered if conservative therapy

has been exhausted. Prolotherapy consists of the injection of growth factors or growth factor stimulators that cause a brief inflammatory response, thus causing increased cellular activity that generates new collagen and extra-cellular matrix.⁷⁸ This process increases connective tissue strength and has demonstrated potential to aid symptoms from BJHS.⁷⁹ Nonetheless, current research has not fully demonstrated the effectiveness of prolotherapy in BJHS.⁸⁰

A case series cannot draw conclusions about efficacy nor effectiveness of the treatment interventions as presented herein. However, a case series can be hypothesis generating, and future research may illuminate best practice in the management of BJHS patients.

Summary

Recognizing joint hypermobility as a significant contributing factor in patients presenting with musculoskeletal complaints is often challenging for chiropractors who rely on clinical judgement, best evidence and knowledge of physiology to provide effective care. The lack of awareness of BJHS may delay the diagnosis as well as delay effective care, thus exacerbating symptoms.¹³ Manual therapy should be used cautiously, but may afford unique benefits in managing pain due to spinal motion segment dysfunction in hypermobile patients. As demonstrated by the cases presented, active exercise is an essential element of care, especially in maintaining joint proprioception. Considering differential diagnoses and quickly reconsidering management in the absence of expected improvement in these cases allowed for a beneficial shift from passive to active care. Simple clinical tests, such as the Brighton Criteria and Beighton Score (Tables 1 and 2) as well as the Hakim and Grahame questionnaire (Table 4), are valid tools that enable the clinician to identify patients with BJHS who will benefit from such a clinical approach. Future research should determine effective clinical management strategies for patients with BJHS, as well as elucidating provocative activities and occupations.

References

1. Kirk JA, Ansell BM, Bywaters EG. The hypermobility syndrome. Musculoskeletal complaints associated with generalized joint hypermobility. *Ann Rheum Dis*. 1967;26(5):419-425.
2. Child AH. Joint hypermobility syndrome: inherited disorder of collagen synthesis. *J Rheumatol*. 1986;13(2): 239.
3. Hakim AJ, Cherkas LF, Grahame R, Spector TD, MacGregor AJ. The genetic epidemiology of joint hypermobility: a population study of female twins. *Arthr Rheumat*. 2004;50(8): 2640-2644.
4. Prockop DJ, Kivirikko KI. Collagens: molecular biology, diseases, and potentials for therapy. *Annual Rev Biochem*. 1995;64(1): 403-434.
5. Beighton PH, Solomon L, Soskolne CL. Articular mobility in an African population. *Ann Rheum Dis*. 1973;32(5): 413.
6. Tinkle BT, Bird HA, Grahame R, Lavalley M, Levy HP, Silience D. The lack of clinical distinction between the hypermobility type of Ehlers–Danlos syndrome and the joint hypermobility syndrome (aka hypermobility syndrome). *Am J Med Gen (part A)*. 2009;149(11): 2368-2370.
7. Grahame R, Bird HA, Child A. The revised (Brighton 1998) criteria for the diagnosis of benign joint hypermobility syndrome (BJHS). *J Rheumatol*. 2000;27(7): 1777.
8. Biro F, Gewanter HL, Baum J. The hypermobility syndrome. *Pediatrics*. 1983;72(5):701-706.
9. Larsson LG, Baum J, Mudholkar GS, Srivastava DK. Hypermobility: prevalence and features in a Swedish population. *Rheumatol*. 1993;32(2):116-119.
10. Birrell FN, Adebajo AO, Hazleman BL, Silman AJ. High prevalence of joint laxity in West Africans. *Rheumatol*. 1994;33(1):56-59.
11. Klemp P. Hypermobility. *Ann Rheum Dis*. 1997;56(10): 573-575
12. Larsson LG, Baum J, Mudholkar GS. Hypermobility: features and differential incidence between the sexes. *Arthr Rheumat*. 1987;30(12):1426-1430.
13. Maillard SM, Payne J. Physiotherapy and occupational therapy in the hypermobile child. In: Hakim AJ, Keer R, Grahame R, eds. *Hypermobility, fibromyalgia and chronic pain*. London: Churchill Livingstone; 2010. p. 179-196.
14. Magee, David J. *Orthopedic Physical Assessment*. St. Louis, Mo: Saunders Elsevier, 2008.
15. Acasuso-Diaz M, Ciscal A, Collantes-Estevéz E. Quantification of joint laxity. ‘The non-dominant (Spanish) modification’. *Rheumatol*. 1995;34(8):795-796.
16. Juul-Kristensen B, Schmedling K, Rombaut L, Lund H, Engelbert RH. Measurement properties of clinical assessment methods for classifying generalized joint hypermobility—a systematic review. *Am J Med Gen (part C)*. 2017; 175: 116-147.
17. Hakim AJ, Grahame R. A simple questionnaire to detect hypermobility: an adjunct to the assessment of patients with diffuse musculoskeletal pain. *Int J Clin Pract*. 2003;57(3):163-166.
18. Bird DA. Joint hypermobility. Reports from special interest groups of the annual general meeting of the British Society for Rheumatology. *Br J Rheumatol*. 1992;31(3):205-208.
19. Lewkonja RM. The biology and clinical consequences of articular hypermobility. *J Rheumatol*. 1993;20(2):220-222.

20. Mishra MB, Ryan P, Atkinson P, Taylor H, Bell J, Calver D, Fogelman L, Child A, Jackson G, Chambers JB, Grahame R. Extra-articular features of benign joint hypermobility syndrome. *Rheumatol*. 1996;35(9):861-866.
21. Gullo TR, Golightly YM, Flowers P, Jordan JM, Renner JB, Schwartz TA, Kraus VB, Hannan MT, Cleveland RJ, Nelson AE. Joint hypermobility is not positively associated with prevalent multiple joint osteoarthritis: a cross-sectional study of older adults. *BMC Musculoskel Dis* 2019; 20: 165.
22. Baeza-Velasco C, Gely-Nargeot M-C, Pailhez G, Vilarrasa A. Joint hypermobility and sport: a review of advantages and disadvantages. *Curr Sports Med Reports*. 2013; 12: 291-295.
23. Gazit Y, Nahir AM, Grahame R, Jacob G. Dysautonomia in the joint hypermobility syndrome. *Am J Med*. 2003;115(1):33-40.
24. Grahame R. Hypermobility and the heritable disorders of connective tissue. *Best Pract Res Clin Rheumatol*. 2000; 14(2): 345-361.
25. Bridges AJ, Smith E, Reid J. Joint hypermobility in adults referred to rheumatology clinics. *Ann Rheum Dis*. 1992;51(6): 793-796.
26. Kaplinsky C, Kenet G, Seligsohn U, Rechavi G. Association between hyperflexibility of the thumb and an unexplained bleeding tendency: is it a rule of thumb? *Br J Haematol*. 1998;101(2): 260-263.
27. El-Garf AK, Mahmoud GA, Mahgoub EH. Hypermobility among Egyptian children: prevalence and features. *J Rheumatol*. 1998;25(5):1003-1005.
28. Al-Rawi Z, Al-Rawi Z. Joint hypermobility in women with genital prolapse. *Lancet* 1982;319(8287):1439-1441.
29. El-Shahaly HA, El-Sherif AK. Is the benign joint hypermobility syndrome benign? *Clin Rheumatol*. 1991;10(3): 302-307.
30. Kirby A, Sugden DA. Children with developmental coordination disorders. *J Royal Soc Med*. 2007;100(4):182-186.
31. Iatridou K, Mandalidis D, Chronopoulos E, Vagenas G, Athanasopoulos S. Static and dynamic body balance following provocation of the visual and vestibular systems in females with and without joint hypermobility syndrome. *J Bodywork Movement Ther*. 2014;18(2): 159-164.
32. Acasuso-Diaz M, Collantes-Estevez E. Joint hypermobility in patients with fibromyalgia syndrome. *Arthr Care Res*. 1998;11(1): 39-42.
33. Francis H, March L, Terenty T, Webb J. Benign joint hypermobility with neuropathy: documentation and mechanism of tarsal tunnel syndrome. *J Rheumatol*. 1987;14(3):577-581.
34. March LM, Francis H, Webb J. Benign joint hypermobility with neuropathies: documentation and mechanism of median, sciatic, and common peroneal nerve compression. *Clin Rheumatol*. 1988;7(1): 35-40.
35. Gulbahar S, Şahin E, Baydar M, Bircan Ç, Kızıl R, Manisalı M, Akalin E, Peker Ö. Hypermobility syndrome increases the risk for low bone mass. *Clin Rheumatol*. 2006;25(4): 511-514.
36. Grahame R. Pain, distress and joint hyperlaxity. *Joint Bone Spine*. 1999;67(3):157-163.
37. Bulbena A, Duró JC, Porta M, Martín-Santos R, Mateo A, Molina L, Vallescar R, Vallejo J. Anxiety disorders in the joint hypermobility syndrome. *Psychiatry Res*. 1993 ;46(1): 59-68.
38. Aktas I, Ofluoglu D, Akgun K. Relationship between lumbar disc herniation and benign joint hypermobility syndrome. *Turk J Phys Med Rehab*. 2011; 57: 85-88.
39. Zarate N, Farmer AD, Grahame R, Mohammed SD, Knowles CH, Scott SM, Aziz Q. Unexplained gastrointestinal symptoms and joint hypermobility: is connective tissue the missing link? *Neurogastroenterol Motil*. 2010;22(3): 252-278.
40. Bulbena A, Gago J, Pailhez G, Sperry L, Fullana MA, Vilarroya O. Joint hypermobility syndrome is a risk factor trait for anxiety disorders: a 15-year follow-up cohort study. *Gen Hosp Psychiatry*. 2011;33(4): 363-370.
41. Eccles JA, Beacher FD, Gray MA, Jones CL, Minati L, Harrison NA, Critchley HD. Brain structure and joint hypermobility: relevance to the expression of psychiatric symptoms. *Br J Psychiatry*. 2012;200(6): 508-509.
42. Campayo JG, Asso E, Alda M, Andres EM, Sobradie N. Association between joint hypermobility syndrome and panic disorder: a case-control study. *Psychosomat*. 2010;51(1): 55-61.
43. Mayer K, Kennerknecht I, Steinmann B. Clinical utility gene card for: Ehlers-Danlos syndrome types I-VII and variants-update 2012. *Eur J Hum Genet*. 2013;21(1).
44. Grahame R. Joint hypermobility and genetic collagen disorders: are they related? *Arch Disease Childhood*. 1999;80(2):188-191.
45. Handler CE, Child AN, Light ND, Dorrance DE. Mitral valve prolapse, aortic compliance, and skin collagen in joint hypermobility syndrome. *Br Heart J*. 1985;54(5): 501-508.
46. Beighton P, Grahame R, Bird HA. Biochemistry of hypermobility. In: Beighton P, Grahame R, Bird HA, eds. *Hypermobility of joints*. 2nd ed. Berlin: Springer; 1989 (chapter 3). p. 25-52.
47. Em S, Ucar D, Oktayoglu P, Bozkurt M, Caglayan M, Yıldız I, Evliyaoglu O, Nas K. Serum prolidase activity in benign joint hypermobility syndrome. *BMC Musculoskel Dis*. 2014;15(1):75.
48. Cassisi G, Sarzi-Puttini P, Casale R, Cazzola M, Boccassini L, Atzeni F, Stisi S. Pain in fibromyalgia and related conditions. *Reumatismo*. 2014;66(1): 72-86.
49. Russek LN, Errico DM. Prevalence, injury rate and, symptom frequency in generalized joint laxity and joint hypermobility syndrome in a "healthy" college population. *Clin Rheumatol*. 2016;35(4):1029-1039.

50. Meeus M, Nijs J. Central sensitization: a biopsychosocial explanation for chronic widespread pain in patients with fibromyalgia and chronic fatigue syndrome. *Clinical Rheumatol.* 2007;26(4): 465-473.
51. Staud R, Craggs JG, Robinson ME, Perlstein WM, Price DD. Brain activity related to temporal summation of C-fiber evoked pain. *Pain.* 2007;129(1): 130-142.
52. Oaklander AL, Herzog ZD, Downs HM, Klein MM. Objective evidence that small-fiber polyneuropathy underlies some illnesses currently labeled as fibromyalgia. *Pain.* 2013;154(11): 2310-2316.
53. Tracey I, Bushnell MC. How neuroimaging studies have challenged us to rethink: is chronic pain a disease? *J Pain.* 2009;10(11): 1113-1120.
54. Ofluoglu D, Gunduz OH, Kul-Panza E, Guven Z. Hypermobility in women with fibromyalgia syndrome. *Clin Rheumatol.* 2006;25(3): 291-293.
55. Russek LN. Hypermobility syndrome. *Phys Ther.* 1999;79(6):591-599.
56. Cherpel A, Marks R. The benign joint hypermobility syndrome. *NZ J Physiother.* 1999;27(3): 9-23.
57. Gedalia A, Press J, Klein M, Buskila D. Joint hypermobility and fibromyalgia in schoolchildren. *Ann Rheum Dis.* 1993;52(7): 494-496.
58. Siegel DM, Janeway D, Baum J. Fibromyalgia syndrome in children and adolescents: clinical features at presentation and status at follow-up. *Pediatrics.* 1998;101(3):377-382.
59. Ting TV, Hashkes PJ, Schikler K, Desai AM, Spalding S, Kashikar-Zuck S. The role of benign joint hypermobility in the pain experience in Juvenile Fibromyalgia: an observational study. *Pediatr Rheumatol Online J.* 2012;10(1):16.
60. Knapik J, Jones BH, Bauman CL, Harris JM. Strength, flexibility and athletic injuries. *Sports Med.* 1992;14(5): 277-288.
61. Taimela S, Kujala UM, Osterman K. Intrinsic risk factors and athletic injuries. *Sports Med.* 1990;9(4):205-215.
62. Diaz MA, Acasuso E, Estevez EC, Guijo PS. Joint hyperlaxity and musculoligamentous lesions: study of a population of homogeneous age, sex and physical exertion. *Rheumatol.* 1993;32(2): 120-122.
63. Klemp P, Chalton D. Articular mobility in ballet dancers. A follow-up study after four years. *Am J Sports Med.* 1989;17(1):72-75.
64. Krivickas LS, Feinberg JH. Lower extremity injuries in college athletes: relation between ligamentous laxity and lower extremity muscle tightness. *Arch Phys Med Rehabil.* 1996;77(11):1139-1143.
65. Decoster LC, Bernier JN, Lindsay RH, Vailas JC. Generalized joint hypermobility and its relationship to injury patterns among NCAA lacrosse players. *J Athl Train.* 1999;34(2):99.
66. Stewart DR, Burden SB. Does generalised ligamentous laxity increase seasonal incidence of injuries in male first division club rugby players? *Br J Sports Med.* 2004; 38: 457-460.
67. Smith R, Damodaran AK, Swaminathan S, Campbell R, Barnsley L. Hypermobility and sports injuries in junior netball players. *Br J Sports Med.* 2005; 39: 628-631.
68. Konopinski MD, Jones GJ, Johnson MI. The effect of hypermobility on the incidence of injuries in elite-level professional soccer players: a cohort study. *Am J Sports Med.* 2012; 40(4): 763-679.
69. Bird HA, Hudson A, Eastmond CJ, Wright V. Joint laxity and osteoarthritis: a radiological survey of female physical education specialists. *Br J Sports Med.* 1980;14(4):179-180.
70. Hall MG, Ferrell WR, Sturrock RD, Hamblen DL, Baxendale RH. The effect of the hypermobility syndrome on knee joint proprioception. *Rheumatol.* 1995;34(2):121-125.
71. Mallik AK, Ferrell WR, McDonald AG, Sturrock RD. Impaired proprioceptive acuity at the proximal interphalangeal joint in patients with hypermobility syndrome. *Rheumatol.* 1994;33(7):631-637.
72. Hull RG. Articular hypermobility presenting after 'aerobic' exercise. *Clin Exper Rheumatol.* 1985;3(4):359.
73. Barrack RL, Skinner HB, Brunet ME, Cook SD. Joint kinesthesia in the highly trained knee. *J Sports Med Phys Fitness.* 1984;24(1):18
74. Celenay ST, Kaya DO. Effects of spinal stabilization exercises in women with benign joint hypermobility syndrome: a randomized controlled trial. *Rheumatol Int.* 2017;37(9):1461-1468.
75. Simmonds JV, Keer RJ. Hypermobility and the hypermobility syndrome. *Man Ther.* 2007;12(4): 298-309.
76. Russek LN. Examination and treatment of a patient with hypermobility syndrome. *Phys Ther.* 2000;80(4): 386-398.
77. Wong JJ, Shearer HM, Mior S, Jacobs C, et al. Are manual therapies, passive physical modalities, or acupuncture effective for the management of patients with whiplash-associated disorders or neck pain and associated disorders? An update of the Bone and Joint Decade Task Force on neck pain and its associated disorders by the OPTIMA collaboration. *Spine J.* 2016; 16: 1598-1630.
78. Strunk RG, Pfefer MT, Dube D. Multimodal chiropractic care of pain and disability for a patient diagnosed with benign joint hypermobility syndrome: a case report. *J Chiropr Med.* 2014;13(1): 35-42.
79. Reeves KD, Hassanein K. Randomized prospective double-blind placebo-controlled study of dextrose prolotherapy for knee osteoarthritis with or without ACL laxity. *Alt Ther Health Med.* 2000;6(2):68-80.
80. Hauser RA, Phillips HJ. Treatment of joint hypermobility syndrome, including Ehlers-Danlos syndrome, with Hackett-Hemwall prolotherapy. *J Prolother.* 2011;3: 612-629.

Health-related quality of life and balance confidence among participants in a senior community-based exercise (SWIFT) program compared to age matched controls: a cross-sectional study

Erin Cogle, BHSc, DC^{1,2}

Margaret Black, RN, BScN, MEd, EdD³

Sheilah Hogg-Johnson, MMath, PhD^{4,5}

Philip Decina, DC, FCCS(C)¹

Anthony Tibbles, BSc, DC, FCCS(C)¹

Silvano A. Mior, DC, PhD, FCCS(C)^{2,4,6}

Background: *Staying Well, Independent and Fit Together (SWIFT), a seniors' exercise program, aims to promote health, strength, mobility and community engagement. We compared quality of life and balance confidence in SWIFT participants and non-participants, aged 60 years and older.*

Methods: *Cross-sectional study comparing participants and non-participants in SWIFT program*

Contexte : *Staying Well, Independent and Fit Together (SWIFT) est un programme d'exercice physique pour personnes âgées visant à promouvoir la santé, la force, la mobilité et la participation aux activités de la collectivité. Nous avons comparé la qualité de vie et le degré de confiance de la personne dans son équilibre entre des sujets participant au programme SWIFT et des sujets n'y participant pas, tous ayant 60 ans et plus.*

Méthodologie : *Étude transversale visant à comparer l'état de participants au programme SWIFT à celui de*

¹ Division of Clinical Education, Canadian Memorial Chiropractic College, Toronto, ON

² Faculty of Health Sciences, University of Ontario Institute of Technology, Oshawa, ON

³ Parish Nurse Tri-Congregational Churches, North York, ON

⁴ Department Research, Canadian Memorial Chiropractic College, Toronto, ON

⁵ Dalla Lana School of Public Health, University of Toronto

⁶ Centre for Disability Prevention and Rehabilitation at Ontario Tech University and Canadian Memorial Chiropractic College, Toronto, ON

Corresponding author:

Silvano Mior, UOIT-CMCC Centre for the Study of Disability Prevention and Rehabilitation, Toronto, Ontario, Canada. M2H 3J1

Tel: 416-482-2340 x 132.

E-mail: smior@cmcc.ca

© JCCA 2020

The authors have no disclaimers or competing interests to report in the preparation of this manuscript. Funding provided by the Government of Ontario's Ministry of Seniors Affairs.

using Older People's Quality of Life Questionnaire (OPQOL) and Activities-specific Balance Confidence Scale (ABCS).

Results: Seventy participants completed surveys, 41 in experimental and 29 in control group. We found a statistically significant between group difference favoring the control group in overall OPQOL score but not in OPQOL subscale nor overall ABCS scores. Participants in both groups participating in weekly exercises had non-significantly higher quality of life subscale scores.

Conclusion: Results suggest seniors in both study groups who participate in exercise have non-significantly higher quality of life scores compared to those who do not participate in exercise. Participation in the SWIFT exercise program or activity in general, contributes to quality of life in seniors.

(JCCA. 2020;64(1):55-64)

KEY WORDS: seniors, exercise program, quality of life, balance, chiropractic

Introduction

Group exercise classes improve not only physical outcomes in seniors but emotional and quality of life outcomes.^{1,2} Improving such outcomes leads to additional benefits such as better balance, increases in weekly social activities and the ability of seniors to achieve their activities of daily living without assistance.^{1,2} These benefits increase the likelihood that seniors can live independently in their own home for longer periods. Seniors' group fitness programs can provide such benefits.

In a study assessing a seniors' fitness program focusing on flexibility, strength, balance and endurance, the authors reported a significant increase in function, physical and emotional health in the exercise group participants' program compared to non-participant matched controls.³ In another study, senior subjects participating in a Pilates

non-participants à l'aide de l'Older People's Quality of Life Questionnaire (OPQOL) et de l'échelle de mesure du degré de confiance de la personne dans son équilibre associé aux gestes de la vie quotidienne (échelle ABC-S).

Résultats : Soixante-dix participants ont rempli les questionnaires, 41 dans le groupe expérimental et 29 dans le groupe témoin. On a observé une différence importante sur le plan statistique entre les groupes en faveur du groupe de contrôle pour ce qui est du score global au OPQOL, mais non pour le score de la sous-échelle du OPQOL ni pour le score global de l'échelle ABC-S. Chez les sujets des deux groupes faisant les exercices hebdomadaires, on n'a pas observé de scores supérieurs significatifs de la sous-échelle de la qualité de vie.

Conclusion : Les résultats semblent montrer que chez les sujets âgés des deux groupes suivant le programme d'exercices, les scores de qualité de vie ne sont pas plus élevés, d'une manière significative, que ceux des sujets ne suivant pas le programme. Le programme SWIFT, et de façon générale l'activité physique, contribue à la qualité de vie des personnes âgées.

(JCCA. 2020;64(1):55-64)

MOTS CLÉS : personnes âgées, programme d'exercice physique, qualité de vie, équilibre, chiropratique

exercise program, comprised of 30-minute sessions twice a week for six months, reported an increase in quality of life as measured by the SF-36 compared to a control group.⁴ Similarly, exercise programs have also yielded significant improvements in physical, mental and social well-being.⁵⁻⁸ Such improvements were assessed after a short time period (two to eight weeks) of structured group exercise programs. Another study showed that individuals who had higher leisure time physical activity throughout adulthood had lower risks for all-cause, cardiovascular- and cancer-related mortality.⁹ These findings suggest there are positive benefits for seniors who participate in structured group exercise programs.

Staying Well, Independent and Fit Together (SWIFT) is such a structured seniors' exercise program. The program aims to promote, improve and maintain physical strength,

balance confidence and mobility, promote health and safety, and encourage community engagement through social interaction in the group. Preliminary empirical evidence suggests that the SWIFT program has a positive effect on seniors' quality of life.¹⁰ Despite a small sample size, participants reported improvements in strength, movement, balance and endurance. Unfortunately, more robust data collection processes to capture basic administrative and measurable outcomes were not available. Collection of program data is important to assess the usefulness of this educational experience for interns and ensure adequate resources are available to sustain growth and determine the comparative effectiveness and benefit to seniors.

Our main objective was to assess if seniors participating in a community-based exercise program have higher quality of life, balance confidence, and social engagement scores compared to a non-participant group of seniors. We hypothesized that participants in the SWIFT program would experience more positive physical, mental and social well-being outcomes compared to non-participants. Our secondary objective was to develop a data collection tool to capture key indicators of program outcomes.

Methods

To assess the impact of the SWIFT program, we compared the main outcomes collected from the program participants (exercise group) to an age-matched senior cohort from same three-church (TCC) community who were not participating in the SWIFT program (control group). Ethics approval was obtained through the Research Ethics Board of the Canadian Memorial Chiropractic College (CMCC) (REB # 1807B01).

Study design

We conducted a cross-sectional study between September and December 2018 from a convenience sample of TCC community members over the age of 60 years.

Participants

All participants in the SWIFT program (exercise group) who were 60 years and older were eligible to participate. We recruited participants via announcements made at the beginning of exercise classes, as well as via TCC announcements. Participants in the SWIFT program attend free of charge and sessions are offered three times a week. Each session consists of approximately 15 minutes

of cardiovascular exercise warm-up, 30 minutes of upper and lower body muscle strengthening using resistance bands, and a 15-minute cool down including stretching and balance exercises.

We recruited non-participants (control group), aged 60 and older via announcements made during formal TCC events, electronic and print bulletins, and posters placed at TCC. Leadership of the TCC endorsed the study and committed to assist in making announcements during their services. Participants were not compensated for completing the surveys, although we offered them an opportunity to win one of four \$25 Shoppers Drug Mart gift cards via random draws of participants from both groups who agreed to provide their contact information.

Outcome measures

Our primary outcomes were the Older People's Quality of Life Questionnaire (OPQOL) and Activities-specific Balance Confidence Scale (ABCS). The OPQOL was developed from a constructivist approach that integrates theory with lay views of the meaning of quality of the lived lives of a representative sample of older people.^{11,12} The OPQOL is a 35-item questionnaire, with items scored on a five-point scale ranging between strongly agree and strongly disagree. Higher scores relate to higher quality of life. It assesses the following dimensions of quality of life (QOL): life overall (four items), health (four items); social relationships (five items); independence, control over life and freedom (four items); home and neighbourhood (four items); psychological and emotional well-being (four items); financial circumstances (four items); leisure and activities (four items); and religion (two items).⁸ The OPQOL has sound psychometric properties (e.g. internal consistency: α 0.78-0.90; test-retest intra-class correlations at four weeks ranged 0.40 to 0.78, with lower correlations related in changes in life; construct validity significantly correlated with self-rated active ageing), and it also performs well among ethnically diverse senior populations.¹¹⁻¹³

The ABCS is a 16-item, self-report measure that assesses the participant's confidence in performing different ambulatory skills without falling. It is based on Bandura's theory of self-efficacy and assesses the subject's confidence in maintaining balance while performing various tasks.¹⁴ The assessment typically takes five to ten minutes to administer.¹⁵ Respondents rate their confidence in these

activities on a scale from 0% to 100%, where a score less than 67% suggests an increased risk of falling. The ABCS has adequate to excellent concurrent (balance confidence inversely related to worry about falling) and construct (scores related to demographic, functional measures and clinical variables) validity, excellent test-retest reliability (intra-class correlation 0.79) and internal consistency (Cronbach's $\alpha = 0.95$) in older adults.^{14,15}

In addition, we collected participant demographic data. These data included: age, gender, living situation, if they volunteered in their community, and if they participated in any physical activity outside of the SWIFT program. These variables were included because previous studies suggested they were associated with physical inactivity and poorer outcomes.^{9,17-19} Finally, we assessed the functionality of the data collection tool by evaluating its ease of use and user preference. Qualitative feedback was obtained from a convenience sample of participants.

Procedure

We obtained informed consent from both the experimental and control groups. After obtaining informed consent, both groups completed the same questionnaire that was made available in two formats: 1) paper based and 2) electronically via an email link to SurveyMonkey (SurveyMonkey Inc.; San Mateo, California, USA; www.surveymonkey.com). These different formats optimized data collection as some seniors were more comfortable with one format than the other and some had limited access to the internet. All participants used ID codes provided by the SWIFT program coordinator. Paper surveys included a signed consent form that was removed and secured by the Parish Nurse before completed surveys were submitted for analysis. Participants who used the online format provided consent by submitting the survey. We collected data over a four-week interval following the first in-class announcement.

Paper surveys were distributed to participants in the exercise group at the end of classes, and to those in the control group following a regular Sunday worship service. They completed the surveys at home and returned them before the end date of the study. Participants in both groups who chose to do the surveys online were given email access at the same time.

We de-identified the paper-based surveys and entered responses in an Excel spreadsheet. The data were collated

on a password-protected computer and stored in a secure office at the CMCC. Data downloaded from the electronic online survey were securely stored on a CMCC server using VPN access with appropriate password protection and encryption.

Finally, at the conclusion of the study we invited all participants, including all members of the TCC community, to a free information seminar during which refreshments were served. The community was informed via posters distributed about the TCC building, announcements placed on TCC website, and the provision of take-away handout seminar notices which all acknowledged support from the Ministry of Seniors Affairs. A CMCC intern and TCC staff presented at each of the events. The seminar provided an overview of the SWIFT program and study results, whilst also promoting the Ontario Ministry of Seniors Affairs' Staying Healthy and Active Campaign.

Statistical analysis

To assess the potential impact of the SWIFT program, we compared the main outcomes collected from participants in the experimental group with those not involved in the SWIFT program (control group). The data were descriptively analyzed (frequencies, means, medians, standard deviations (SD), 95% confidence intervals (95%CI)). Tests of difference were used to assess outcomes between groups, including Pearson chi-square (categorical variables of gender, dwelling type, living situation and volunteer status); t-tests for independent samples for age, and outcome scores (ABCS and OPQOL). In addition, the outcome scores were compared between seniors reporting participation in weekly activity and those not, regardless of study group using t-tests. Missing data were recorded as missing and not imputed. The level of significance was set at the 0.05 level. Free text comments were reviewed and collapsed into common categories and descriptively analyzed. The statistical analysis for this study was generated using SAS© v9.4 (SAS Institute Inc., Cary, NC, USA).

Results

Subject Characteristics

Seventy subjects completed the surveys, 29 were participants in the control group and 41 in the experimental group. There were 52 females and 18 males with no dif-

Table 1.
*Demographics of the Experimental and Control groups
 (gender, age, living situation, volunteering/social interactions throughout the month).*

Characteristic	Experimental group N=41	Control group N=29	Statistical significance of variables between experimental and control groups
Gender			0.45 (p-value from χ^2)
Female n (%)	33 (81)	19 (66)	
Age mean (SD)	71.7 (6.68)	76.1 (8.61)	0.019 (p-value from t-test)
Mean (range) time in SWIFT (months)	38.4 (1-78)	NA	
Living situation			0.86 (p-value from χ^2)
Living alone n (%)	14 (34)	9 (31)	
Living with someone n (%)	20 (49)	16 (55)	
Did not specify n (%)	7 (17)	4 (14)	
Dwelling Type			0.34 (p-value from χ^2)
Apartment/Condo n (%)	21 (51)	10 (35)	
House/Townhouse n (%)	17 (42)	15 (52)	
Did not specify n (%)	3 (7)	4 (14)	
Volunteer Yes n (%)	21 (51)	22 (76)	0.04 (p-value from χ^2)

ference in sex distribution between groups. The overall mean age was 73.68 (SD 7.87), ranging between 59 to 91 years. The mean age between groups was significantly different, being about 4.4 years older in the control group on average compared to the experimental group. Subjects in the experimental group reported average participation in the SWIFT program of 38.4 months, ranging from one to 78 months. Volunteering and social activities that the participants partake in were also recorded. Some of the volunteering activities the participants partake in include: 25 individuals volunteering at a church, six assisting other seniors in the community, two assisting at libraries and nine that assist at other associations throughout the community. Subject details for all participants are presented in Table 1.

We found no significant difference in the overall ABCS scores between the experimental and the control groups. We found a significant difference between the two groups for their overall OPQOL scores; the control group had a higher OPQOL average score (Table 2). Mean OPQOL subscale scores were not significantly higher in the control group compared to the experimental group (Table 2).

There was no statistically significant difference in the average ABCS score for female participants (86.79, SD 13.19) compared to the male participants (88.39, SD 9.26). There was also no significant difference between the average OPQOL score for female participants (4.02, SD 0.44) compared to the male participants (4.07, SD 0.37). There were also no differences in the mean ABCS

Table 2.
Overall scores of ABCS and OPQOL, and subscales of OPQOL for participants (Experimental group) and non-participants (Control group) of the SWIFT program.

Scale	Experimental group mean (SD) N=41	Control group Mean (SD) N=29	p-value from t-test
Overall ABC Scores	87.96 (12.82)	86.74 (11.20)	0.68
Overall OPQOL Scores	3.91 (0.39)	4.20 (0.42)	0.0075
OPQOL Subscale Scores			
Life Overall	4.18 (0.50)	4.43 (0.49)	0.93
Health	3.73 (0.58)	4.00 (0.54)	0.69
Social	3.40 (0.47)	3.86 (0.55)	0.32
Independence	4.02 (0.48)	4.22 (0.56)	0.34
Home	4.23 (0.47)	4.43 (0.48)	0.88
Psychological	4.15 (0.43)	4.35 (0.51)	0.32
Finance	3.97 (0.66)	4.12 (0.73)	0.60
Leisure	3.83 (0.51)	4.04 (0.55)	0.68
Religion	4.32 (0.54)	4.35 (0.70)	0.22

scale or mean OPQOL scores related to participants' living situations.

We found that in the total sample, 20 individuals did not participate in any other exercise and 50 did participate in other exercises, including the SWIFT program. Eighteen of those who participated in regular additional exercise walked, eleven worked out at home or at a gym, four did yoga, seven participated in Tai Chi, six danced for exercise and four golfed. Mean ABCS scores, overall OPQOL score and all OPQOL subscale scores were consistently higher among those reporting participation in exercise than in those not, with only the difference for OPQOL Life Overall and Home subscales achieving significance (Table 3).

Social-educational events:

Upon completion of data collection and analysis, we held

two social-educational events for invited participants. Twenty-nine participants attended the first presentation, including 21 SWIFT participants and eight from the community. Twenty-two participants attended the second presentation, with 15 SWIFT participants and seven from the community. After these sessions, seven individuals were interested in joining the SWIFT program. We also received several testimonials from the SWIFT program participants after these sessions which included one female participant who explained that she had undergone knee surgery earlier in the previous year and how much the exercise program helped to get on her feet faster than she would have ever expected. Another female participant remarked how much she would sweat during the exercises and how she knew how it was helping her keep active.

Table 3.
Scores of the OPQOL subscales comparing participants who participated in weekly exercise.

Scale	Participation in Weekly exercise (no) mean (SD) N=20	Participation in Weekly exercise (yes) mean (SD) N=50	p-value from t-test
Overall ABC Scores	83.56 (14.63)	88.57 (10.88)	0.12
Overall OPQOL Scores	3.91 (0.47)	4.10 (0.40)	0.11
OPQOL Subscale Scores			
Life Overall	4.09 (0.55)	4.40 (0.47)	0.02
Health	3.70 (0.55)	3.91 (0.57)	0.16
Social	3.50 (0.40)	3.65 (0.60)	0.31
Independence	4.04 (0.53)	4.16 (0.48)	0.36
Home	4.15 (0.57)	4.41 (0.42)	0.04
Psychological	4.11 (0.57)	4.30 (0.20)	0.14
Finance	4.00 (0.60)	4.05 (0.73)	0.70
Leisure	3.78 (0.55)	3.98 (0.52)	0.15
Religion	4.32 (0.64)	4.39 (0.54)	0.73

Software programming for future data collection

We programmed the online survey, including the demographic, balance confidence and quality of life outcomes in an offline-capable format. All features of the survey (including demographic information) remained constant between the online/offline formats. After assessing the landscape for offline-capable survey platforms, we decided that the most feasible option was to utilize Microsoft Excel software's ability to save locally to work around the networking issues.

We uploaded the program onto two Lenovo TAB3 10 Business tablets, with 32GB of internal memory. The use of the tablet allows for offline and online capabilities, within a functional yet resistant body. In addition, the "Business Features" of the standard Android 6.0 installed platform includes hardware encryption and remote ac-

cess, so that if the onsite internet becomes accessible, the online version of the survey is accessible.

Discussion

We found a significant difference in the overall quality of life score favouring the control group but not in balance confidence between the experimental and control groups. When looking into the living situation for individuals, we found no significant difference in quality of life scores or balance confidence for those who lived with another person in their household and those that did not. We also found no significant differences in overall ABCS and OPQOL scores by age and gender. Although the overall OPQOL and ABC scores between the two groups differed, we did find that exercise in any capacity was related to the outcomes scores.

We asked all participants (in both experimental and

control groups) whether they participated in any regular exercise (apart from the SWIFT program) throughout the week. We found consistently higher mean scores for the ABCS and all components of the OPQOL including overall happiness, ability to enjoy their life more, and feeling safer within their homes and communities among those reporting exercise participation, albeit only significant differences for the life and home sub-sections of the OPQOL.

Lastly, we noted how many participants chose to complete the questionnaires online or in paper format for future reference. Thirty-seven out of the 70 participants chose to complete the online version, and most reported how simple it was to use the SurveyMonkey link that was provided through email. The majority of survey respondents in both groups (online and paper formats) responded positively to questions about the ease of data collection when verbally questioned by the Parish Nurse. Those who reported being experienced computer users preferred the online version. In contrast, those with limited or no computer skills preferred the paper version.

Our finding of no significant difference in scores between the experimental and control groups for the ABCS and OPQOL subscale scores, but a significant difference in overall OPQOL score favouring the control group, is not consistent with previous studies assessing seniors in similar exercise groups.^{1-3,9,14,17,18} One such study reported that seniors' fitness programs significantly improve function, physical and emotional health when compared to non-participant matched controls.³ Others have shown that group exercise programs have a beneficial effect on risk factors for falls, such as balance, gait and strength.¹⁴ In a meta-analysis of twenty-one studies assessing exercise interventions for preventing falls among frail older people living in care facilities, including 5540 participants, exercise interventions were found to effectively reduce the rate of falls in older people. Further, the evidence supports the use of balance training for fall prevention and highlights the importance of combining exercise and fall interventions in reducing rates of falling.¹ In a systematic review that included 18 studies administering exercise programs to participants over the age of 65, exercise therapy was effective in decreasing depressive symptoms, and led to improvements in both the quality of life and self-esteem scores.² In a study looking at older adults living alone and living with someone, the participants that were living

alone or not being able to remain alone at home had lower quality of life scores.¹⁷ Another study showed that older women were more likely to experience joint pain if they lived rurally, were more overweight, have poorer physical and emotional health related to quality of life and use more medications.¹⁸ It has also been shown that increased physical activity engaged throughout adulthood was correlated with a decrease in all-cause, cardiovascular and cancer related deaths.⁹ Thus, exercise and physical activity appear to be associated with improved quality of life, well-being and reduced rates of falling.

The lack of significant differences in the exercise group reported in our study, compared to the aforementioned studies, is likely due to the healthy and active attributes of our control group participants, who were already participating in other exercise or physical activities. Our control group had a high participation rate in exercise that did not include the SWIFT exercise program, therefore they were keeping active in other ways. Both groups came from the same church community that shares a strong faith and similar socioeconomic status, which led to both groups being very similar in demographic information. Furthermore, since we were unable to include a baseline measurement for the experimental group, we were unable to assess if there was change in the SWIFT program participants' balance confidence and quality of life over time. Future study with a more robust design could assess if the SWIFT program results in improved quality of life and balance confidence in seniors.

We conducted two educational sessions with both SWIFT participants and community members. The sessions allowed for the dissemination of the knowledge acquired from our results to the community. Despite encouragement by funding agencies and institutional review and ethics boards to disseminate research results to participants and communities, this is rarely done.²⁰ More importantly, the majority of participants in health research are interested in receiving results from studies they were involved and validates their participation.^{20,21} This is particularly pertinent when study findings are related to personal or family members' health.²⁰ It appears that those attending our sessions were interested in the results, as seven additional community members were motivated and encouraged to enroll in the SWIFT program.

Finally, we accomplished our second objective by replicating all features of the survey (including demographic

information) into both online/offline formats accessible on a functional tablet. In so doing, baseline data from all new participants in the SWIFT program can be tracked and their outcomes monitored and compared over time. This will allow for program assessment and potential modifications as required.

Strengths and limitations

Our study had some strengths, for instance we used psychometrically sound outcome measures. We had similar communities within the Tri-Congregational Churches when comparing the experimental and control groups. This made it easier for the study to be completed since the groups were similar in age, gender and living situations.

The study also had several limitations. First, despite various efforts to recruit participants, our samples sizes were small and varied between the groups, which may explain differences in demographic variables, as well as higher but not significant between-group scores suggesting a possible Type II error. Second, we did not know the experimental group's baseline measurements prior to their starting the SWIFT exercise program, so could not assess change over time. Third, the range of time participants were engaged in the SWIFT program was large and the impact of SWIFT may have been lessened on participants with limited time in the program. However, only four participants were new to the program and were unlikely to have substantially affected mean scores. Fourth, the surveys were completed in the privacy of the participant's own home, so there is a possibility of a response bias. A non-response bias, especially in the control group is possible. Since the control group was recruited through announcements, those that felt like they had a poorer quality of life or physical abilities may have chosen not to participate in the study. There was no measure used to test participants' physical capabilities. Finally, since the participants came from a similar community they might have provided answers that were more socially acceptable, such as being happier with their overall life or being healthier than they actually are. Having such a similar group from the Tri-Congregational Churches also could have resulted in a selection bias. This community is likely not completely representative of the older population because there are many opportunities for the participants to be more socially engaged than those that may be more socially isolated.

Conclusion

We found no significant differences in the ABCS and the OPQOL subscales, and a significant difference in the overall OPQOL scores, between those who participated in the SWIFT program and those that did not. However, when comparing the overall results of the two groups, we found that exercising in any capacity was associated with significantly higher reported quality of life scores in regards to enjoying their lives more, and feeling safer in their homes and communities. This suggests that being part of the SWIFT exercise program or remaining active one way or another contributes to enhanced quality of life in seniors. Finally, conducting educational sessions to inform seniors of the benefits of exercise programs appears helpful in encouraging those who may not have otherwise considered participating. Future research should assess if the SWIFT program balance confidence, balance and quality of life outcomes in seniors improves compared to baseline measures and over time.

Acknowledgments

Thank you to the participants for their involvement in the study and to the volunteers at the Tri-Congregational Churches that helped collect the surveys and answer any questions that the participants may have had.

References

1. Lee S, Kim H. Exercise interventions for preventing falls among older people in care facilities: a meta-analysis. *Worldview Evid Based Nurs*. 2017;14(1): 74-80.
2. Park S, Han K, Kang C. Effects of exercise programs on depressive symptoms, quality of life, and self-esteem in older people: a systematic review of randomized controlled trials. *Appl Nurs Res*. 2014;27(4): 219-226.
3. Hamar B, Coberley C, Pope J, Rula E. Impact of a senior fitness program on measures of physical and emotional health and functioning. *Popul Health Manage*. 2013;16(6): 364-372.
4. Liposki D, da Silva Nagata I, Silvano G, Zanella K, Schneider R. Influence of a Pilates exercise program on the quality of life of sedentary elderly people: a randomized clinical trial. *J Bodywork Move Ther*. 2019;23(2): 390-393.
5. Papadopoulos C, Jager JM. The effect of an educational program on strength-training adherence in older adults. *Educ Gerontol*. 2016; 42(5): 342-351.
6. Bray N, Smart R, Jakobi J, Jones G. Exercise prescription to reverse frailty. *Appl Physiol Nutr Metabol*. 2016; 41(10):1112-1116.
7. Wallace R, Lees C, Minou M, Singleton D, Stratton G.

- Effects of a 12-week community exercise programme on older people. *Nursing Older People*. 2014; 26(1): 20-26.
8. Tamari K. Self-reported home exercise and younger age predict improved health-related quality of life among community-dwelling elderly participants in a three-month group exercise class. *J Physiol Anthropol*. 2011; 30(2): 77-85.
 9. Saint-Maurice PF, Coughlan D, Kelly SP, Keadle SK, Cook MB, Carlson SA, et al. Association of leisure-time physical activity across the adult life course with all-cause and cause-specific mortality. *JAMA Net Open*. 2019; 2(3):e190355.
 10. Inwood K, Johnson L, Moore T, Rioux C. Evaluation of a Community Based Physical Activity Program for Older Adults: Pilot Study. Toronto (ON): Canadian Memorial Chiropractic College; 2015.
 11. Bowling A. The psychometric properties of the Older People's Quality of Life Questionnaire, Compared with the CASP-19 and the WHOQOL-OLD. *Curr Gerontol Geriatr Res*. 2009; 298950.
 12. Bowling A, Stenner P. Which measure of quality of life performs best in older age? A comparison of the OPQOL, CASP-19 and WHOQOL-OLD. *J Epidemiol Comm Health*. 2011; 56: 273-280.
 13. International Longevity Centre – UK. Good Neighbours: Measuring Quality of Life in Older Age. London; 2011: 4-6, 12. Available from <https://ilcuk.org.uk/good-neighbours-measuring-quality-of-life-in-old-age/>. Accessed November 28, 2019.
 14. Tally KMC, Wyman JF, Gross CR. Psychometric properties of the Activities-Specific Balance Confidence Scale and the Survey of Activities and Fear of Falling in Older Women. *J Am Geriatr Soc*. 2008; 56: 328-333.
 15. Raad J, Moore J, Hamby J, Rivadelo R, Straube D. A brief review of the Activities-Specific Balance Confidence Scale in older adults. *Arch Phys Med Rehabil*. 2013; 94(7): 1426-1427.
 16. Franco MR, Sherrington C, Tiedemann A, Pereira LS, Perracini MR, Faria CRS, et al. Effectiveness of senior dance on risk factors for falls in older adults (DanSE): a study protocol for a randomised controlled trial. *BMJ Open*. 2016; 6(12): e013995.
 17. Hellstrom Y, Persson G, Hallberg I. Quality of life and symptoms among older people living at home. *J Adv Nurs*. 2004; 48(6):584-593.
 18. de Luca K, Wong A, Eklund A, Fernandez M, Byles JE, Parkinson L, et al. Multisite joint pain in older Australian women is associated with poorer psychosocial health and greater medication use. *Chiropr Man Ther*. 2019; 27:8.
 19. Rivera-Torres S, Fahey TD, Rivera MA. Adherence to exercise programs in older adults: informative report. *Gerontol Geriatr Med*. 2019; 5: 1-10.
 20. Purvis RS, Abraham TH, Long CR, Stewart MK, et al. Qualitative study of participants' perceptions and preferences regarding research dissemination. *AJOB Empir Bioeth*. 2017; 8(2): 69-74.
 21. Long CR, Purvis RS, Flood-Grady E, Kimminau KS, et al. Health researchers' experiences, perceptions and barriers related to sharing study results with participants. *Health Res Pol Syst*. 2019; 17:25.

Locus of control in patients with Huntington disease: a cross-sectional study

Brynne E. Stainsby, BA, DC, FCCS(C)¹

Silvano Mior, DC, FCCS(C), PhD¹

Mark Guttman, MD, FRCPC²

Background: *Health locus of control (LOC) represents an individual's beliefs regarding one's ability to influence health outcomes. In patients with chronic and neurodegenerative diseases, greater internal LOC has been associated with lower levels of disability.*

Objective: *To examine LOC in patients with Huntington disease (HD).*

Methods: *A cross-sectional study of individuals affected by HD, stratified by disease status, was conducted. Participants completed a demographic questionnaire, the Internal Control Index (ICI), and the Hospital Anxiety and Depression Scales.*

Results: *Thirty-four subjects completed the study. All groups demonstrated greater internal LOC (measured by ICI scores), and significant differences between groups were observed. Secondary analysis demonstrated relationships between depressive symptoms and*

Contexte : *Le locus de contrôle de la santé (LCS) représente les croyances d'une personne sur sa capacité d'influer sur son état de santé. Chez les patients atteints de maladies chroniques et neurodégénératives, un locus de contrôle plus interne est associé à des degrés d'invalidité moindres.*

Objectif : *Observer le LCS chez des patients atteints de la maladie de Huntington.*

Méthodes : *On a mené une étude transversale auprès de personnes atteintes de la maladie de Huntington, regroupées en fonction du stade de la maladie. Les participants ont rempli un questionnaire démographique, le questionnaire Internal Control Index (ICI) et le questionnaire HADS (Hospital Anxiety and Depression Scale).*

Résultats : *Trente-quatre sujets ont participé à l'étude jusqu'à la fin. Dans tous les groupes, on a observé un locus de contrôle plus interne (mesuré par les scores ICI), et des différences significatives entre les groupes. Une étude secondaire a montré l'existence de liens entre les symptômes de la dépression et les symptômes de l'anxiété, de même qu'entre le score ICI et le temps*

¹ Canadian Memorial Chiropractic College, Toronto, Ontario, Canada

² Center for Movement Disorders, Toronto, Ontario, Canada

Corresponding author: Brynne E. Stainsby, Department of Clinical Education, Canadian Memorial Chiropractic College, 6100 Leslie Street, Toronto, ON M2H 3J1

Tel: 416.482.2340

E-mail: bstainsby@cmcc.ca

© JCCA 2020

The authors have no disclaimers or competing interests to report in the preparation of this manuscript.

Funding for this study was provided by the Canadian Memorial Chiropractic College

anxiety symptoms, and ICI score and time from clinical diagnosis of HD.

Conclusion: As patients with chronic pain and neurodegenerative diseases such as HD are likely to present for chiropractic care, identifying factors such as anxiety, depression and LOC may affect patients' response to care.

(JCCA. 2020;64(1):65-75)

KEY WORDS: anxiety, chronic pain, depression, Huntington disease, internal-external control

Introduction

Chronic musculoskeletal pain is a complex problem affecting up to 85% of patients in chiropractic clinical practice.¹ Chronic pain is one of the leading burdens of illness in Canada with direct and indirect costs of \$5.8 billion in 2008. Patients with chronic pain conditions frequently experience misdiagnosis, delayed diagnosis, unnecessary tests and referrals, frustration, and poor outcomes; all of which may lead to increased burden on the health care system.¹ Given the financial and societal burden, appropriate diagnosis and management of these complaints is critical, and understanding how patients respond to their diagnosis may assist clinicians in developing appropriate educational or manual interventions. For example, locus of control (LOC), based on Rotter's social learning theory², is a personality trait that influences human behaviour²⁻⁴. This theory posits that behaviour is influenced by an individual's expectation of reinforcement, the perceived value of the reinforcement, and the psychological context of the situation.² LOC is specifically related to the "expectation for reinforcement" component of this theory²⁻⁴, as well as how an individual perceives and interprets an event, and how s/he then chooses to respond to the situation³.

In chronic diseases, health LOC represents an individual's beliefs regarding his/her ability to influence health outcomes.³ Individuals with a greater internal LOC believe health outcomes are self-determined, via their actions or strategies, and demonstrate greater self-esteem, experience less depression, trait anxiety and neurotic symptoms.³ Using measures including the Internal Con-

écoulé à partir du diagnostic clinique de la maladie de Huntington.

Conclusion : Les patients souffrant de douleurs chroniques et de maladies neurodégénératives, comme la maladie de Huntington, sont susceptibles de chercher de l'aide auprès des chiropraticiens. Certains facteurs comme l'anxiété, la dépression et le LCS peuvent influencer sur la réponse des patients aux soins.

(JCCA. 2020;64(1):65-75)

MOTS CLÉS : anxiété, douleur chronique, dépression, maladie de Huntington, contrôle interne-externe

trol Index (ICI)⁴, patients with HIV were divided into subtypes related to their mood, anxiety and ability to adapt to the disease⁵. Those in the 'highly adaptive' subtype were found to have greater internal LOC when compared with the 'average performing' and 'severely dysfunctional' subtypes.⁵ In contrast, those with lesser LOC believe health outcomes are due to fate, luck or the decisions of doctors.²⁻⁴

While traditionally considered to be a personality trait that is stable over time, numerous authors have reported differences in LOC when examined in the context of illness or disease.⁵⁻⁸ Gruber-Baldini *et al.* examined LOC relative to disability scores and quality of life in patients with Parkinson's disease.⁶ In their cross-sectional study, greater internal LOC scores were associated with lower levels of disability.⁶ In consideration of the psychological context variable of the social learning theory³, the authors suggest those with greater internal LOC may have adopted behaviours and strategies to maintain functional abilities⁶. The authors further hypothesize that LOC may affect the course of disability as a consequence of a condition.⁶

Low back pain and neck pain are the most commonly reported conditions for seeking chiropractic care; however, those with Parkinson's disease, multiple sclerosis, stroke and diabetes may also seek care.⁹ These chronic conditions may present with unique neuromusculoskeletal complaints, for which chiropractic care may be helpful. Huntington disease (HD) is an example of a neurodegenerative disease that may cause neuromusculoskeletal

complaints. HD is a fatal, autosomal dominant neurodegenerative disorder characterized by progressive motor dysfunction, decreased cognitive abilities, and psychiatric or behavioural disturbances (such as depression).¹⁰⁻¹² The prevalence of HD is 4-10 per 100,000 people; however, it impacts many more people. One of its most devastating effects is the impact on family life, including those at-risk, family members and caregivers.¹³⁻¹⁷

The genetic mutation that confirms the diagnosis is a cytosine-adenine-guanine (CAG) trinucleotide repeat expansion in the gene that encodes for the huntingtin protein on chromosome 4.^{18,19} Typically, longer repeat lengths represent an earlier age of onset and more rapid progression of symptoms. While the length of the trinucleotide repeat accounts for 50-70% of the variability in these factors, little is known about how other genes or mechanisms influence the development or rate of progression of HD.^{10,11,20-22} The role of non-genetic factors (such as coping strategies or behavioural modifications) in the delay of symptom onset and progression is believed to be important, but not well understood.¹¹

In addition to assisting with prediction of the age of onset, the discovery of the DNA markers associated with the gene responsible for HD led to the development of predictive testing in those at-risk.^{23,24} The risk of developing HD is considered a major stressor with tremendous influence on the life and major decisions of the at-risk population^{14-16,25}, yet only 10-20% have participated in predictive testing^{14,16}. It has been suggested that the avoidance of testing may be related to passive or maladaptive coping strategies.²⁵

Clinically, HD is recognized by the triad of motor, cognitive and psychiatric symptoms.¹⁰⁻¹² Clinical diagnosis of HD is a complex process based on the clinical impression of the treating physician and is not homogenous in clinical settings as there is no current consensus regarding clinical diagnostic criteria. The Unified Huntington Disease Rating Scale (UHDRS) diagnostic criterion is based on the motor assessment component of the UHDRS and requires the unequivocal presence of an extrapyramidal movement disorder, such as choreic movement, in a patient with a family history of HD.^{10,27} In addition to its role in diagnosis, the UHDRS is the current gold standard measure used in research protocols; including clinical trials, as well as being used to stratify patients into groups based on stages of disease.^{10,11,18,21,22,27} The UHDRS also includes a behav-

ioural examination, which investigates symptoms such as depression and suicidal thoughts.²⁷ Behavioural examination is important as up to 63% of HD patients experience depressive symptoms, which have also been associated with decrease in functional abilities and quality of life.²⁸

Beyond the obvious medical implications, an affected individual or their family member may also be required to cope with new financial burdens, social stigma, genetic discrimination, and the risk that children may inherit the disease.^{14-16,29,30} To deal with this myriad of difficulties, patients and their families may adopt passive coping strategies. Passive coping strategies have been considered maladaptive, as the individual relinquishes control of their pain or situation and/or allows other areas of life to be adversely affected.³¹⁻³³ Such coping strategies have been linked to poorer adjustment in chronic health conditions, including chronic pain, rheumatoid arthritis and whiplash disorders.^{6,31-33} Passive coping strategies have also been observed both in patients demonstrating lesser internal locus of control (LOC) and those with depression.^{6,25}

The work by Gruber-Baldini provides new insight into the role of LOC in neurodegenerative diseases.⁶ Specifically in HD patients, research has focused on physical and cognitive symptoms; however, little research has addressed the psychosocial aspects of the disease.³⁴ Based on the scarce research related to LOC and the results of Gruber-Baldini *et al.*⁶, and in consideration of the potential for coping strategies or behavioural modifications to effect the course of disability, we aimed to examine the role of LOC in people affected by HD. Specifically, we examined:

- 1) the LOC in subjects affected by HD as measured by the Internal Control Index (ICI); and
- 2) their levels of depression and anxiety using the Hospital Anxiety and Depression Scale (HADS).

Methods

Study design

We used a cross-sectional survey design. Given the number of ways that an individual may be affected by HD, we allocated subjects to four groups based on their HD status. We examined differences between 'at-risk' (positive family history of HD without genetic testing results), 'pre-symptomatic' (genetic test positive for CAG muta-

tion, without clinical diagnosis by HD physician), 'early symptomatic' (genetic test positive for CAG mutation, plus clinical diagnosis by HD physician), and 'genetic negative' (family history of HD with a negative genetic test result) groups at a given point in time. Despite a paucity of evidence to suggest that LOC may be linked to the stage of disease progression, individuals are commonly allocated to these groups in order to receive support in the HD community or participate in research.^{10,11,18,21,22,27,34}

Sample specification

The target population was a convenience sample of males and females between 25 and 45 years who were affected by HD. The most recent clinical information provided by the patient's HD physician was used to allocate subjects into the pre-symptomatic or early symptomatic group. HD is not specifically associated with particular demographic variables; therefore, subjects representing a wide variety of ethnicities, education levels and socioeconomic status were eligible to participate. Subjects were excluded if they did not fit the sample specifications or were not competent to consent. Subjects were recruited from the Centre for Movement Disorders (Markham, ON) and via on-line communications through the Huntington Society of Canada (HSC) to its members.

Due to limited subject recruitment at six months, the protocol was amended to allow for online data collection. As those who provided data via online assessment did not undergo UHDRS testing, it was not possible to validate their correct allocation to the assigned study groups. However, we performed a post-hoc analysis to evaluate differences in results between the subjects who completed data collection in-person and those who completed it online to assess for differences in responses in these groups.

Finally, to increase participation in the study, we used a modification of a widely accepted framework for survey methods.³⁵ The HSC agreed to distribute only three emails to its membership at one week intervals to inform and remind members of the study and encourage participation. The study received institutional Research Ethics Board approval (REB approval 1007X05).

Description of outcome measures

Locus of control was evaluated using the ICI. The ICI was developed by Duttweiler in 1984 based on variables asso-

ciated with internal LOC, such as cognitive processing, autonomy, resistance to influence attempts, delayed gratification and self-reliance.⁴ It is a 28-item questionnaire and each item is rated on a 5-point scale, where higher scores indicate greater levels of internal LOC.^{4,36} The maximal score (high internal response pattern) is 140, and the minimum score (low internal response pattern) is 28.⁴ In follow-up studies, the ICI has been shown to be a reliable measure of LOC with an internal consistency of 0.85.³⁶ It is significantly correlated to the Beck Depression Inventory, the State-Trait Anxiety Inventory (Form Y), the Coopersmith Self-Esteem Inventory (Form A), and the Eysenck Neuroticism Scale.³⁶ Further, convergent validity of this scale has been demonstrated against Rotter's Internal-External Scale.³⁶ While LOC is traditionally considered to be a personality trait that is stable over time, there have been reported changes in LOC when measured in the context of illness or disease.⁵⁻⁸

We used the Hospital Anxiety and Depression Scale (HADS) to address the secondary aim of the study.³⁸ The HADS has been used to assess the presence and severity of depression (HADS-D) and anxiety (HADS-A), and has been demonstrated to have good internal consistency.^{38,39} It contains 14 questions, with scores ranging from zero to 42, where higher scores indicate greater levels of depression and anxiety.³⁸ The HADS has been validated for use in patients with HD with an area under curve of 0.90.²⁸ In this population, it has acceptable psychometric properties with a sensitivity of 1.00 and specificity of 0.79 using optimal cut-off values.²⁸

Clinical features of HD were assessed using the UHDRS, a research tool produced and revised by the Huntington Study Group (HSG).²⁷ It was developed to provide a uniform assessment of the clinical features and disease progression, and allows for comparison of clinical signs, disease progression and the effects of therapy, within and between trials.²⁷ The UHDRS is composed of motor, cognitive, behavioural and functional assessments, an independence scale and a measure of total functional capacity.²⁷ The UHDRS was revised based on research experience and available evidence and refinements made to the cognitive and behavioural assessment sections.²⁷ The UHDRS is the current gold standard for research protocols, has undergone extensive reliability and validity testing, and has been used as a major outcome measure by the HSG in controlled clinical trials.^{10,11,18,21,22}

Description of experimental maneuver

We included two strategies as noted above. The first strategy involved in-person interaction with the principal investigator (PI); the subject completed paper copies of the demographic questionnaire, ICI and HADS, and underwent complete UHDRS assessment. This strategy enabled the investigators to determine if the UHDRS scores obtained during the study were consistent with the most recent clinical information provided by an HD physician via the patient's self-reported HD status. The second strategy involved subjects completing only the demographic questionnaire (including clinical information for group allocation), ICI and HADS via an online survey tool.

Strategy I

Prior to data collection, a research assistant utilized a computer-generated random numbers table to label envelopes containing the two outcome measures (ICI and HADS), labelled with the same unique subject identification (ID) number to code to ensure confidentiality for all subjects. After consenting to participate, the subjects underwent a brief interview with the PI to determine if they had undergone genetic testing, and if so, what the results were (negative or positive). Those with a positive genetic test were additionally asked if they had been diagnosed with HD by their physician. This information was used to allocate the subject into the appropriate group.

Subjects were provided with a coded envelope and led to a private area to complete the questionnaires and were instructed to return their completed surveys to the PI with the envelope sealed. Following the completion of the questionnaire, the PI assessed each subject via the UHDRS and recorded their scores. All data for each subject were gathered during one session lasting approximately one hour.

An independent research assistant scored both paper outcomes, and documented the UHDRS scores. The assistant then used a second computer-generated random numbers table to reassign subject numbers and provided the PI with the total scores for each of the three outcome measures for data analysis. The second random number served to further ensure anonymity for participants.

The PI was blinded to the participants' survey responses as described above. Compliance was assessed by recording the number of eligible subjects who participated, the number who provided demographic information and the number not willing to participate.

Strategy II

Strategy II was introduced to increase the number of eligible participants and allow for the recruitment of subjects across Canada. All data (including demographic information) were collected online. Identical subject inclusion and exclusion criteria applied in both strategies. All subjects consenting to participate were provided survey questions as described above from the demographic questionnaire, the ICI and the HADS. The same randomized coding procedure as described in Strategy I was undertaken by an independent research assistant. All data were returned to the PI for analysis.

Sample size estimation

The sample size, based on Cohen's f for an ANOVA⁴⁰ and calculated via the R-Project software (R Project, version 2.10.0)⁴¹, was 15 per group for a total of 60 subjects. Standard deviations (SD) obtained from previously cited studies^{5,42} suggested a great degree of variability. Therefore, the pooled SD (13.52) from the Smith *et al.* study⁴² was used to calculate the sample size based upon methodological similarity to our study. This calculation was based on an effect size (f) of 0.49, accounting for a 10% change that was arbitrarily deemed as clinically relevant. To compensate for non-compliance and errors in completing the outcome measures, an additional 20% was added to each group, for a total sample size of 18 per group and a total study size of 72.

Data analysis

In order to assess for any differences in responses between the subjects who completed Strategy I (in-person), and those who completed online assessment (Strategy II) of the ICI and HADS, analysis of variance was performed. The ICI scores for each group were descriptively analyzed to provide means, standard deviations, minimum and maximum values. To address the primary aim of this study, a one-way ANOVA with Bonferroni correction was used to assess mean differences in the ICI scores. All calculations were based on a Type I error of 0.05 and a Type II error of 0.2. To address the secondary aim of this study, the scores for the HADS were analyzed using a Kruskal-Wallis test, as the data violated the assumptions for ANOVA use (assessed by the Bartlett test).

We calculated Spearman's correlation test to assess the relationships between the outcome measures. Specifically,

Table 1.
Demographic characteristics of subjects.

	Negative	At-risk	Pre-symptomatic	Early Symptomatic
Total number	8	10	9	7
Average age (yrs)	30.0 (3.8)	29.0 (5.2)	35.4 (7.2)	38.7 (6.1)
Female subjects (%)	87.5	70	66.7	42.9
Subjects from Strategy 1 (%)	50.0	60.0	44.4	57.1
Avg years from genetic result (sd)	2.0 (2.3)	N/A	8.1 (6.5)	7.6 (5.1)
Avg years from clinical diagnosis (sd)	N/A	N/A	N/A	3.6 (2.9)

we assessed the relationship between HADS-anxiety and HADS-depression scores, ICI score and length of time in years from HD clinical diagnosis, ICI score and length of time in years from HD genetic result, and depression and length of time in years from genetic result, and the length of time in years from the clinical diagnosis. All data were analyzed with STATA (STATA, version 10.0) and SPSS (IBM SPSS Statistics for Windows, Version 26.0) statistical software.

Results

Thirty-four subjects completed all the questionnaires, while 16 completed the UHDRS assessment (Strategy 1). Subjects were similarly distributed across each of the four groups. They ranged in age from 25 to 45, with greater proportion being female; however, this finding is consistent with other HD studies (Table 1).³⁸ There were no significant differences in ICI and HADS scores of subjects recruited in-office and on-line. Therefore, the data sets were merged and all scores used in the analysis.

The grouped ICI scores are summarized in Table 2. All groups demonstrated mean scores above the scale midpoint of 84, indicating increased levels of internal LOC.⁴ Post-hoc Bonferroni-corrected contrasts demonstrated a significant difference between the at-risk and early symptomatic groups ($p < 0.01$), and the early and pre-symptomatic groups ($p < 0.02$) (Table 3).

Results from the Kruskal-Wallis test of the HADS-D indicated a significant difference between groups (Adjusted $H = 12.2$, $df=3$, $p < 0.01$). Table 4 provides the means and standard deviations of these scores. Subjects in the at-risk category had the lowest scores (fewest depres-

Table 2.
Descriptive statistics of ICI scores by group.

Group	Mean	SD	Minimum	Maximum
Negative	104.4	12.8	80	124
At-risk	109.3	5.5	101	118
Pre-symptomatic	107.0	9.4	92	117
Early symptomatic	90.6	12.0	74	107

Table 3.
ICI scores contrasted between groups as calculated by Bonferroni-corrected comparison.

Group	Negative	At-risk	Early symptomatic
At-risk	4.9		
Pre-symptomatic	2.6	-2.3	16.4 ($p < 0.02$)
Early symptomatic	-13.8	-18.7 ($p < 0.01$)	

Table 4.
Median scores and ranges for HADS-D.
Adjusted $H = 12.2$, $p < 0.0$

Group	Score
Negative	2.0 (0-10)
At-risk	0.5 (0-3)
Pre-symptomatic	3.0 (0-9)
Early symptomatic	6.0 (1-10)

Table 5.
Median scores and ranges for HADS-A.
 $H = 6.8, p=0.08$

Group	Score
Negative	5.5 (3-9)
At-risk	5.0 (1-8)
Pre-symptomatic	6.0 (3-15)
Early symptomatic	8.0 (4-15))

sive symptoms), while the early symptomatic subjects averaged the highest scores. There were no significant differences between groups (Adjusted $H = 6.8, df=3, p = 0.08$). Descriptive results by group are provided in Table 5.

Secondary analysis demonstrated HADS-D was significantly related to HADS-A, $r_s = 0.63$, 95% Bias Corrected accelerated (BCa) Confidence Interval (CI) [0.42, 0.74], $p < 0.01$. The ICI scores were not significantly related to the length of time (in years) from the time of genetic testing, $r_s = -0.59$, 95%BCa CI [-0.43, 0.31], $p = 0.78$. The ICI scores were significantly related to length of time (in years) of clinical diagnosis $r_s = 0.89$, 95%BCa CI [0.16, 1.00], $p < 0.01$. The HADS-D scores were not significantly related to the length of time (in years) from the time of genetic testing, $r_s = 0.02$, 95%BCa CI [-0.41, 0.44], $p = 0.93$, nor were the HADS-D scores significantly related to the length of time (in years) from the time of clinical diagnosis, $r_s = 0.22$, 95%BCa CI [-0.50, 0.73], $p = 0.63$.

Discussion

Our data suggests differences in LOC may exist between groups of subjects affected by HD, such that those in the early symptomatic group had greater internal LOC than the at-risk and pre-symptomatic groups. Additionally, we observed a significant difference in depression scores between the at-risk and early symptomatic groups. We observed a trend of increasing anxiety from the at-risk to the pre-symptomatic groups, which appeared to decrease in the early symptomatic groups. We also observed increased anxiety levels in the negative group compared to those at-risk.

Locus of control

Our results suggest that differences in LOC may exist within groups of subjects affected by HD. Specifically; the at-risk group had a significantly lower LOC score than the early symptomatic group. Although we did not evaluate changes in LOC as the disease progresses within an individual with HD, one may hypothesize that because those in the at-risk group have not yet received the genetic test results, they may perceive a lack of ability to control their future. Alternatively, those in the early symptomatic group may recognize the disease process has begun and may adopt active coping behaviours, as demonstrated in those with greater internal LOC.^{2,4,6,31-33}

Helder *et al.* identified a similar trend in their cross-sectional study.³⁴ They found that a sample of patients with clinically diagnosed HD scored significantly higher on the “acceptance” subscale of the COPE inventory compared to a convenience sample of healthy adults.³⁴ Interestingly, these patients scored significantly lower on other subscales such as “suppression of competing activities” and “mental disengagement”.³⁴ This may suggest a difference in coping strategies related more to personality traits (such as LOC) rather than the cognitive decline associated with HD.^{8,34}

Furthermore, our secondary analysis demonstrated a relationship between higher ICI scores and time from first clinical diagnosis of HD in the early symptomatic group. Again, this is consistent with the findings of Helder *et al.*, where their subjects had a meantime elapsed of 5.1 years from first diagnosis.³⁴ Although we had a small group ($n = 7$) in our study, a future longitudinal study is warranted to confirm this trend.

The early symptomatic group also had a significantly higher internal LOC than the pre-symptomatic group. Again, it is possible that those in the pre-symptomatic stage of HD experience a sense of uncertainty in waiting for the onset of their disease and thus are more likely to feel that future health outcomes are related to luck or fate.^{2,4,6} McAllister *et al.* reported that anxiety is commonly experienced in patients affected by genetic conditions; however, it often becomes more intense during times of disease change, such as a new diagnosis or genetic testing.⁴⁵ With respect to those specifically affected by HD, they commonly experience greater levels of distress following a genetic diagnosis in anticipation of the onset of HD, due to previous experiences of observing the effects

of HD and concern for how such effects will affect their own life.^{34,45}

Depression

Since up to 63% of patients with HD demonstrate depressive symptoms that may be related to passive coping and poorer health outcomes^{8,28}, we also examined the scores of the HADS-D scale. Previous studies suggest that depressive symptoms in patients with HD are associated with impaired function and decreased quality of life.²⁸ Paulsen *et al.* found that those with an expanded CAG repeat upon genetic testing demonstrated greater levels of distress on psychiatric testing than those without the expansion.⁴⁴ We observed a significant difference in depression scores between the at-risk and early symptomatic groups. Our findings reinforce the presence of depressive symptoms in patients with early symptomatic HD, and encourage early evaluation and treatment.²⁸

While our findings support the presence of depression in patients with HD, we cannot comment upon how it is affected by the progression of symptoms over time. We found a trend toward a negative correlation between the HADS-depression score and the elapsed time since receiving a genetic result. It is important to interpret these results with caution as this relationship was observed within a small and specific group. If confirmed in a larger study, these results may suggest that within the pre-symptomatic stage of HD, individuals may become less affected by depressive symptoms the longer they have to accept the result. When dealing with patients with terminal illnesses, Kubler-Ross described a dynamic cycle involving denial, anger, bargaining, depression and acceptance states of grief.⁴⁵ In consideration of this process, it may be hypothesized that simply the confirmation of a genetic diagnosis may evoke similar emotional responses and cause an individual to progress through the process of grief, eventually resulting in greater acceptance.

Anxiety

Anxiety is considered a common neuropsychiatric symptom in patients with HD, and the evaluation of its presence is considered important in a comprehensive examination of a patient with the disease.²¹ Although we observed no significant differences between groups, our findings suggested a trend of increasing anxiety from the at-risk to the pre-symptomatic, which then decreased in the early

symptomatic groups. As hypothesized above, perhaps the distress associated with receiving a positive genetic test leads to greater anxiety than the actual development of the disease due to LOC and the belief that future health will be determined by fate.^{2,4,6,34}

Increased anxiety levels were also observed in the negative group compared to the at-risk. In addition to the challenges that may present to all members of an affected family¹⁴⁻¹⁶, those who receive a negative genetic test result are commonly affected by “survivor’s guilt”⁴⁵. This concept speaks to the guilt associated with not inheriting the CAG expansion, while other family members may not yet know about their future, may have tested positive, and/or may already demonstrate symptoms.⁴⁵ Studies suggest those receiving negative genetic test results demonstrate this phenomenon, in addition to a period of emotional numbness and difficulties developing new perspectives for life.^{45,47-51} Future studies with a larger sample may elucidate if this finding changes over time.

Finally, when examining the pooled data from all subjects, a positive relationship was observed between depression and anxiety, suggesting that these symptoms may be linked. This finding is supported by previous research that has identified a wide spectrum of neuropsychiatric symptoms and disorders in patients with HD.^{10,11,27,28}

Limitations and future research

Our study included a small convenience sample that may not be representative of the general HD population. It is important, therefore, to interpret the results with caution. It is also possible that data collected online were completed by participants with assistance from a second party. In the future, a study involving multiple sites to recruit a larger sample may confirm the observed results of this study.

Recruitment was limited to individuals prior to the onset of moderate to severe symptoms in order to ensure that all subjects were competent to provide consent. While this limitation was intended to protect the rights of the research subjects and increase internal validity, it may have resulted in the recruitment of a younger population and limited the generalizability to the entire population affected by HD. In patients without neurodegenerative disease, internal LOC was associated with hippocampal volume in young and elderly subjects⁵²; however, this relationship has not been examined in patients with HD or other neurodegenerative diseases. Future research may

attempt to measure this. It should also be noted that there was a high percentage of females who participated in this study. Given there is no relationship between inheritance and patient sex, the findings of this study must be interpreted with caution with respect to the generalizability to the entire population affected by HD.

It is important to consider that disease progression in patients with HD has historically been evaluated based on motor dysfunction; however, changes in behaviour and cognition may be observed as a patient approaches symptomatic diagnosis.⁵³ Duff *et al.* have found a greater prevalence of apathy, disinhibition and executive dysfunction (“frontal behaviours”) in patients with the CAG expansion, and also noted that these behaviours are associated with motor and cognitive markers of HD progression.⁵³ These findings reinforce the need for further research with a more robust population and may also suggest that the group stratification could be modified given the association between the Frontal System Behavioral Scale scores and the probability of diagnosis within five years.⁵² It is possible that those in the pre-symptomatic category could perhaps be considered in the early stage using expanded diagnostic criteria. Future longitudinal research with a more robust population and diverse outcome measures may provide further insight into the role of LOC as the disease progresses.

Despite numerous attempts using various forms of communication through both the PI and the HSC, subject recruitment was a significant challenge. The HSC has documented that many Canadians affected by HD prefer to remain independent from affiliations with the disease, avoid participation with volunteer or support groups, and decline to participate in research studies.⁴ Utilizing Dillman’s method as it is described may have resulted in a greater response rate; however, we felt our modified approach balanced the need for email communications and respecting the time and willingness of a charitable organization and its volunteers, donors and members to participate.³⁵

Given the difficulties with recruitment, we recognize our study was underpowered based on our sample size estimation. This may have impacted our results and increased the potential for type II error, thus our findings should be cautiously interpreted. Given the lack of current studies on LOC in patients with HD, we used the pooled SD of a study with similar methodology⁴², as well as arbitrarily

assuming a 10% difference in scores would be clinically relevant. In the initial estimation of required number of subjects, it was decided to include an additional percentage (20%) to account for the possibility of non-compliance and errors in completing the outcome measures. In retrospect this appeared to be a gross over-estimation of the actual number of incomplete returns, which was zero. Despite these comments, the authors acknowledge the failure to recruit the number of subjects recommended by the power calculation, and encourage care in interpretation of the results. Future studies should consider a multi-site strategy in effort to facilitate subject recruitment.

Further, it is also important to recognize the potential for selection bias in the current study. It is possible that the participants in our study may have had greater levels of internal LOC compared to the general population affected by HD. Those who demonstrate more active coping strategies (including those with greater levels of internal LOC) commonly seek out information regarding their disease process³⁰, thus they may be more likely to participate in research. It may also be possible that those with fewer depressive symptoms and lower levels of anxiety would also be more likely to participate in research. However, it can be assumed that selection bias would affect each group equally, and thus relationships observed between groups may be realistic.

Finally, our study was cross-sectional in design and did not allow for evaluation of change in LOC over time. It does provide the basis for hypothesis generation and future longitudinal studies. In particular, this study aimed to inform future research that examines an individual’s LOC as it relates to coping ability and strategies, quality of life and/or functional abilities.

Conclusion

As patients with chronic pain and neurodegenerative diseases such as HD are likely to present for chiropractic care, it is important that chiropractors recognize the psychosocial factors that may affect patients’ clinical presentation and response to care. In addition to manual care, chiropractors may consider evaluating LOC, screening for symptoms of anxiety and depression, and/or identifying passive coping strategies, which may be associated with poorer outcomes in chronic health conditions. Assisting patients with the development of active coping strategies (or referring for this when appropriate)

may benefit patients and their prognoses. Future research could have important implications in informing disease management programs and coping strategies for individuals affected by HD.

Acknowledgements

The authors wish to thank Dr. Marion McGregor for her assistance with the development of this study and statistical analysis, and the Canadian Memorial Chiropractic College for funding for this study.

References

1. Bourgaize S, Newton G, Kumbhare D, Srbely J. A comparison of the clinical manifestation and pathophysiology of myofascial pain syndrome and fibromyalgia: implications for differential diagnosis and management. *J Can Chiropr Assoc.* 2018;62(1): 26-41.
2. Rotter JB. Some problems and misconceptions related to the construct of internal vs. external control of reinforcement. *J Consult Clin Psych.* 1975; 48: 56-67.
3. Rotter JB. Some implications of a social learning theory for the prediction of goal directed behaviour from testing procedures. *Psychol Rev.* 1960; 67: 301-316.
4. Duttweiler PC. The Internal Control Index: a newly developed measure of locus of control. *Educ Psychol Meas.* 1984; 44: 209-221.
5. Combs DR and Livingston RB. Psychological sub-types among persons with HIV infection: an empirical study. *AIDS Care.* 2001;13: 157-162.
6. Gruber-Baldini AL et al. Effects of optimism/pessimism and locus of control on disability and quality of life in Parkinson's disease. *Parkinsonism Related D.* 2009; 15: 665-669.
7. Oberle K. A decade of research in locus of control: What have we learned? *Journal Adv Nurs.* 1991;16: 800-806.
8. Fishbain DA, Cole B, Cutler RB, Lewis J, Rosomoff HL and Rosomoff RS. Chronic pain and the measurement of personality: Do states influence traits? *Pain Med.* 2006;7(6): 509-529.
9. Beliveau PJH, Wong JJ, Sutton DA, Simon NB, Bussieres AE, Mior SA and French SD. The chiropractic profession: a scoping review of utilization rates, reasons for seeking care, patient profiles, and care provided. *Chiropr Man Therap.* 2017;25: 35-52.
10. Tabrizi SJ, Langbehn DR, Leavitt BR, Roos RA, Durr A, Craufurd D et al. Biological and clinical manifestations of Huntington's disease in the longitudinal TRACK-HD study: cross-sectional analysis of baseline data. *Lancet Neurol.* 2009; 8: 791-801.
11. Paulsen J. Early detection of Huntington disease. *Neurol.* 2010;5(1): 85-104.
12. Hedreen JC, Peyser CE, Folstein SE, Ross CA. Neuronal loss in layers V and VI of cerebral cortex in Huntington's disease. *Neurosci Lett.* 1991;133: 257-261.
13. Huntington Society of Canada [homepage on the Internet]. Kitchener, ON; c2010 [cited 2010 January 24]. Available from: <http://www.huntingtonsociety.ca>.
14. Etchegary H. Coping with genetic risk: living with Huntington Disease (HD). *Curr Psychol.* 2009;28: 284-301.
15. Forrest Keenan K, Miedzybrodzka Z, van Teijlingen E, McKee L, Simpson SA. Young people's experiences of growing up in a family affected by Huntington's disease. *Clin Genet.* 2007;71: 120-129.
16. Vamos M, Hambridge J, Edwards M, and Conaghan J. The impact of Huntington's disease on family life. *Psychosomatics.* 2007; 48: 400-404.
17. Statistics Canada [homepage on the Internet]. Address; c2010 [cited 2010 January 24]. Available from <http://www.statcan.gc.ca>
18. Witjes-Ane MW, Mertens B, van Vugt JP, Bachoud-Levi AC, van Ommen GJ, and Roos RA. Longitudinal evaluation of "presymptomatic" carriers of Huntington's disease. *J Neuropsychiatry Clin Neurosci.* 2007; 19: 310-316.
19. The Huntington's Disease Collaborative Research Group. A novel gene containing a trinucleotide repeat that is expanded and unstable on Huntington's disease chromosomes. *Cell.* 1993; 72: 971-983.
20. Li JL, Hayden MF, Almqvist EW, Brinkman RR, Durr A, Dode C et al. A genome scan for modifiers of age at onset in Huntington Disease: The HD MAPS study. *Am J Hum Genet.* 2003; 73: 682-687.
21. Paulsen JS, Zhao H, Stout JC, Brinkman RR, Guttman M, Ross CA et al. Clinical markers of early disease in persons near onset of Huntington's disease. *Neurol.* 2001; 57: 658-662.
22. Duff K, Paulsen JS, Beglinger LJ, Langbehn DR, Stout JC et al. Psychiatric symptoms in Huntington's Disease before diagnosis: The Predict-HD Study. *Biol Psychiatry.* 2007; 62: 1341-1346.
23. Babul R, Adam S, Kremer B, Dufasne S, Wiggins S, Huggins M et al. Attitudes toward predictive testing for the Huntington disease gene: relevance for other adult-onset disorders. The Canadian Collaborative Group on Predictive Testing for Huntington Disease. *J Am Med Assoc.* 1993;270: 2321-2325.
24. Fox et al. Predictive testing for Huntington disease: description of a pilot project in British Columbia. *Am J Med Genet.* 1989; 32: 211-216.
25. Pakenham KI, Goodwin VA and MacMillan JC. Adaptation to being at-risk for Huntington's disease and the availability of genetic testing: application of a stress and coping model. *Psycho Health Med.* 2004; 9: 380-397.
26. Hayden MR. Predictive testing for Huntington's disease: the calm after the storm. *Lancet.* 2000; 356: 1944-1945.

27. Kiebertz K et al. The Unified Huntington's Disease Rating Scale: reliability and consistency. *Mov Dis.* 1996;11: 136-142.
28. De Souza J, Jones LA and Rickards H. Validation of Self-Report Depression Rating Scales in Huntington's Disease. *Mov Dis* 2010; 25: 91-96.
29. Bombard Y et al. Managing genetic discrimination: Strategies used by individuals found to have the Huntington disease mutation. *Clin Genet.* 2007; 71: 220-231.
30. Bombard Y et al. Engagement with genetic discrimination: concerns and experiences in the context of Huntington disease. *Eur J H Genet.* 2008;16: 279-289.
31. Snow-Turek AL, Norris MP and Tan G. Active and passive coping strategies in chronic pain patients. *Pain.* 1996;64: 455-462.
32. Brown GK, Nicassio PM, and Wallston KA. Pain coping strategies and depression in rheumatoid arthritis. *J Consult Clin Psych.* 1989;57(5): 652-657.
33. Carroll LJ, Cassidy JD, and Cote P. The role of pain coping strategies in prognosis after whiplash injury: Passive coping predicts slowed recovery. *Pain.* 2006;124: 18-26.
34. Helder DI, Kaptein AA, van Kempen GMJ, Weinman J, van Houwelingen HC and Roos RAC. Living with Huntington's disease: Illness perceptions, coping mechanisms and patients' well-being. *Br J Health Psych.* 2002; 7: 449-462.
35. Hoddinott SN and Bass MJ. The Dillman Total Design survey method: A sure-fire way to get high survey return rates. *Can Fam Phys.* 1986;32: 2366-2368.
36. Meyers LS and Wong DT. Validation of a new test of locus of control: the Internal Control Index. *Educ Psychol Meas.* 1988; 48: 753-761.
37. Jacobs KW. Psychometric properties of the Internal Control Index. *Psychol Rep.* 1993;73: 251-255.
38. Zigmond AS and Snaith RP. The Hospital Anxiety and Depression Scale. *Acta Psychiatr Scand.* 1983;67: 361-370.
39. Djukanovic I, Carlsson J and Arestedt K. Is the Hospital Anxiety and Depression Scale (HADS) a valid measure in a general population 65-80 years old? A psychometric evaluation study. *Health Qual Life Out.* 2017; 15: 193.
40. Cohen J. (1998) *Statistical power analysis for the behavioural sciences* (2nd ed), New Jersey: Lawrence Erlbaum Associates.
41. R project
42. Smith VL. Analysis of locus of control and education level utilizing the Internal Control Index. Marshall University Graduate College 2003.
43. Amrhein PC et al. Locus of control and the age difference in free recall from episodic memory. *J Gen Psychol.* 1999, 126:149-164.
44. Paulsen JS, Magnotta VA, Mikos AE, Paulson HL, Penziner E, Andreasen NC and Nopoulos PC. Brain structure in preclinical Huntington's disease. *Biol Psychiatry.* 2006;59: 57-63.
45. McAllister M, Davies L, Payne K, Nicholls S, Donnai D and MacLeod R. The emotional effects of genetic diseases: Implications for clinical genetics *Am J Med Genetics.* 2007;143A: 2651-2661.
46. Kubler-Ross E (1969). *On death and dying*, Routledge.
47. Meiser B and Dunn S. Psychological impact of genetic testing for Huntington's disease: an update of the literature. *J Neurol Neurosurg Psychiatry.* 2000;69: 574-578.
48. Maat-Klerit A, Vegter-van der Vlis M, Zoetewij M, Losekoot M, van Haeringen A and Roos R. Paradox of a better test for Huntington's disease. *J Neurol Neurosurg Psychiatry.* 2000;69: 579-583.
49. Tibben A, Duivenvoorden HJ, Vegter-van der Vlis M, Niermeijer MF, Frets PG, van de Kamp JJP et al. Presymptomatic DNA testing for Huntington disease: identifying the need for psychological intervention *Am J Med Genet.* 1993;48: 137-144.
50. Tibben A, Timman R, Bannink EC and Duivenvoorden HJ. Three-year follow-up after presymptomatic testing for Huntington's disease in tested individuals and partners. *Health Psychol.* 1997;16(1): 20-35.
51. Wiggins S, Whyte P, Huggins M, Adam S, Theilmann J, Bloch M et al. The psychological consequences of predictive testing for Huntington's disease. *N Engl J Med.* 1992;327: 1401-1405.
52. Pruessner J, Baldwin MS, Dedovic K et al. Self-esteem, locus of control, hippocampal volume, and cortisol regulation in young and old adulthood. *NeuroImage.* 2005;28: 815-826.
53. Duff K, Paulsen JS, Beglinger LJ et al. "Frontal" behaviors before the diagnosis of Huntington's disease and its relationship to markers of disease progression: Evidence of early lack of awareness. *J Neuropsychiatry Clin Neurosci.* 2010; 22(2): 196-207.

Superficial venous thrombosis of the upper limb presenting to a chiropractic clinic: a case report

Paul Mastragostino, HBSc.Kin, DC, FCCS(C)¹

Chadwick Chung, BSc, DC, FCCS(C)²

Objective: *To present the clinical case of a patient with an upper extremity superficial venous thrombosis (SVT), and to highlight the importance for clinicians working in musculoskeletal care settings, to consider non-musculoskeletal causes for their patients' presentations.*

Clinical Features: *A 31-year-old male presented to an academic chiropractic clinic with progressive left sided tension over the medial arm, extending to the anterior aspect of his proximal forearm.*

Intervention and Outcome: *The patient was initially diagnosed with possible biceps/brachialis muscular strain and peripheral entrapment of the median nerve. A course of treatment involving soft tissue therapy was initiated. Unfortunately, the patient's symptoms worsened, and on further evaluation, near full occlusion and phlebitis of the left cephalic vein was discovered. Symptoms dissipated over the next few days with conservative medicinal efforts.*

Summary: *Although not often viewed as a serious*

Objectif : *Présenter le cas d'un patient ayant une thrombose veineuse superficielle (TVS) du membre supérieur et souligner l'importance pour le clinicien dans un établissement de traitement de troubles musculosquelettiques de prendre en compte les causes qui ne sont pas d'origine musculosquelettique dans l'étude du tableau clinique du patient.*

Caractéristiques cliniques : *Un homme de 31 ans s'est présenté à une clinique chiropratique universitaire en raison d'une tension progressive ressentie à partir de la partie médiane du bras gauche jusqu'à la face antérieure de l'avant-bras proximal.*

Intervention et résultat : *On a d'abord évoqué la possibilité d'une foulure du biceps ou du muscle brachial antérieur et d'un englobement périphérique du nerf médian. Une thérapie des tissus mous a été amorcée. Mais malheureusement, les symptômes du patient se sont aggravés et, après une évaluation plus approfondie, on a découvert une occlusion presque complète de la veine céphalique gauche et une phlébite. Les symptômes se sont dissipés au cours des jours qui ont suivi le début d'un traitement conservateur par des médicaments.*

Résumé : *La TVS n'est pas souvent considérée comme*

¹ Department of Graduate Education and Research Programs, Canadian Memorial Chiropractic College

² Division of Clinical Education, Canadian Memorial Chiropractic College

Corresponding author: Paul Mastragostino, Department of Graduate Education and Research Programs, Canadian Memorial Chiropractic College, Toronto, Ontario, Canada

E-mail: paul.mastragostino@gmail.com

© JCCA 2020

The authors have no disclaimers, competing interests, or sources of support or funding to report in the preparation of this manuscript.

The involved patient provided consent for case publication.

condition, or factored in the differential diagnoses of musculoskeletal practitioners, to not consider SVT as a cause of a patient's symptoms may lead to a protracted clinical course and increased discomfort for the patient, and in rare cases, more serious consequences.

(JCCA. 2020;64(1):76-81)

KEY WORDS: chiropractic, diagnosis, management, superficial venous thrombosis, upper limb

Introduction

Superficial venous thrombosis (SVT) is a relatively common condition, yet many cases are not recognized or reported as it is generally considered to have a benign and self-limiting course. That being said, SVT has been linked to more serious conditions such as deep vein thrombosis (DVT) and pulmonary embolism (PE).¹⁻³ Although the true incidence is uncertain, in the United States, the annual incidence of SVT has been roughly estimated to be over 123,000 cases.^{4,5} Patients often present with a painful “cord-like” mass along the course of the affected superficial vein, and clinical features may also include swelling, erythema, and/or warmth of the overlying skin.^{4,6,7}

In the early stages, some patients with venous thrombosis lack signs of acute inflammation and may present with non-discriminatory symptoms mirroring musculoskeletal or other biomechanical conditions.^{8,9} This may result in a delay in diagnosis or improper treatment options, which may lead to a progression of the disease process, poorer prognosis, and prolonged pain and discomfort for the patient. Therefore, it is important that SVTs are effectively identified by all primary care practitioners to allow for proper management and the mitigation of unfavourable outcomes. However, pathology associated with vascular diseases are not often considered as differential diagnoses by musculoskeletal practitioners such as physical therapists and chiropractors.¹⁰ We present the case of a 31-year-old male who presented to a chiropractic clinic with a chief complaint of anterior left arm tension, in-

une affection grave et n'est pas prise en compte dans les diagnostics différentiels posés par les praticiens de l'appareil locomoteur. En ne la considérant pas comme une possible cause des symptômes d'un patient, on risque de prolonger les interventions et d'aggraver la gêne du patient, et dans de rares cas, de causer des conséquences plus graves.

(JCCA. 2020;64(1):76-81)

MOTS CLÉS : chiropratique, diagnostic, prise en charge, thrombose veineuse superficielle, membre supérieur

itially attributed to a muscular strain/nerve entrapment, which was later identified as being the result of an SVT. An overview of the etiology, risk factors, diagnosis, and management of SVT as they pertain to this case will also be discussed.

Case presentation

History

A 31-year-old male presented to a chiropractic teaching clinic with a four-week history of progressive left sided arm tension which extended from the medial aspect of his arm to the anterior aspect of his proximal forearm. He indicated that he was generally healthy and had not changed any of his regular physical activity or exercise routines over the preceding year. He did not participate in any competitive training or repetitive overhead activities. He described his arm symptoms as tension along the medial aspect of the arm when full outstretched, and did not otherwise have any discomfort. The symptoms had been mildly progressive as they were becoming present with a reduced amount of extension to the elbow and they were not resolving with time. The patient indicated that the tension in the arm had begun to limit his ability to participate in physical activity and that he was becoming increasingly concerned and anxious. The patient denied any constitutional signs or symptoms, his medical history was unremarkable, and he was generally well. In addition, there were no red flags indicative of serious pathology identified during the assessment.

Physical examination

The patient was 5'4" and weighed 120 pounds. There was no observable swelling, discolouration, or skin lesion visualized over the area of complaint. Ranges of motion of the elbow were full with a report of tension over the anterior aspect of the left elbow in full extension. His blood pressure in the left arm was 110/65 mmHg and his heart rate was 64 bpm. An upper limb neurological examination was unremarkable bilaterally. Orthopedic testing of the left elbow was negative. Examination of the cervical spine was unremarkable and did not reproduce any of the patient's arm symptoms. On palpation, mild tenderness was elicited over the left biceps and brachialis musculature as well as over the cubital fossa. There was a thin rope-like structure that was tender to palpation along the anterior medial aspect of the forearm that coursed along the path of the median nerve. The differential diagnoses for the patient included biceps/brachialis muscular strain and peripheral entrapment of the median nerve.

Management

A treatment plan that included soft tissue therapy (specifically myofascial release techniques) over the area of complaint was initiated. Following a single session of manual therapy, the patient reported a gradual increase in tension in the anterior arm, and pain when performing full extension of the elbow. At this point, further manual therapies were discontinued and a trial of watchful rest was advised. After approximately one week of rest, the patient reported that the symptoms were progressively worsening and the patient was instructed to consult with his general physician who subsequently requisitioned a doppler ultrasound of the left elbow. The results of the ultrasound identified a near full occlusion and phlebitis of the mid-portion of the left cephalic vein, above the cubital fossa. The patient was then advised to avoid strenuous activity and to apply heat to the area until his symptoms dissipated. The patient began to experience relief to his arm within the five days that followed. On further questioning, it was discovered that approximately one month prior to the patient's initial clinic visit, he had undergone routine venipuncture for serology testing, which may have been the inciting event to the patient's condition.

Discussion

SVT characteristically involves a sequence of inflamma-

tion and thrombosis in the lumen of a superficial vein(s) as a consequence of one, or more, mechanical, chemical, biological, or rarely, infectious factors.^{1,11} The degree of thrombosis within a vein can vary, with more severe cases extending into the deep venous system.¹² The etiology of SVT has often been attributed to the components of Virchow's triad, which describes three broad categories of contributory factors for the development of thrombosis: 1) endothelial injury, 2) hemodynamic changes, such as venous stasis and turbulent flow, and 3) hypercoagulable states due to underlying conditions (i.e. factor V Leiden, prothrombin G mutation, protein C and protein S deficiency, antithrombin II, etc.).^{13,14} Endothelial damage to the vessel wall can result from either external trauma, such as from blunt force injuries or compression of the vein, or internal trauma, due to intravenous injections or drawing blood, as depicted in the presented case.¹

Varicose veins are thought to be a major predisposing factor for SVT and have been found to be associated in as high as 62% of cases due to their increased susceptibility to local external injury, as well as their propensity for venous stasis and altered flow rates.^{1,14-16} Additional risk factors that have been identified for the development of SVT include prolonged immobilization, obesity, use of oral contraceptives or hormone replacement therapy, pregnancy, recent surgery, vein stripping, the use of certain drugs, a history of SVT or DVT, intravenous catheter use, and active malignancies.^{1,11,13} Autoimmune diseases may also increase susceptibility to the development of SVT, such as Behçet's disease and Buerger's disease.^{17,18}

Although the epidemiology of SVT has not been well established, the prevalence of SVT in the lower extremity in the general population has been estimated to be around 3% to 11%, with an increased prevalence in women and older adults.^{5,19} This is approximately two-times higher than both DVT and PE combined.²⁰ SVT in the upper extremity is less common and typically involves the cephalic and basilic veins.^{13,21}

Clinical features of SVT can include the presence of erythema, warmth, and/or swelling over the affected vein. On palpation the vein is usually tender and may be hard along its distribution.^{6,22,23} In the absence of characteristic signs and symptoms, recognizing potential instigating factors from the patient's history, such as possible traumas or systemic conditions, becomes imperative for identifying the correct diagnosis.

Compared to the lower extremity, the risk of progression of upper extremity SVT to DVT or PE is believed to be less.¹³ Nevertheless, pulmonary emboli have been reported to be present in up to one-third of patients with upper extremity DVT.²⁴ Additionally, although a rare occurrence, the development of PE arising from thrombosis of the superficial veins of the arm, such as the basilic vein, has been documented.²⁵ The risk also seems to be greater when an SVT is in close proximity to a junction with the deep venous system, in particular, the saphenofemoral and the saphenopopliteal junctions.^{1,22} In fact, migration of an SVT to within three centimetres of the saphenofemoral junction, where the great saphenous vein joins the femoral vein, is considered to be as dangerous as a proximal femoral DVT.⁴

In the present case, the patient's chief complaint was that of tension over the anteromedial arm and forearm with no obvious signs of swelling or discolouration. His medical history was unremarkable. However, he did recall routine blood work being performed approximately one month prior to the onset of his left arm complaints that was conducted on the same side of his presenting symptoms. This key piece of information was initially not considered during the initial assessment, and only deemed relevant after an adverse response to soft tissue therapy was experienced by the patient. As stated above, SVTs occurring in the upper extremities are not as common as those in the lower extremities, but specific details of the patient's history such as the use of intravenous catheters, intravenous injections or drug use, or venipuncture for blood work or donations, should increase the suspicion of an upper extremity SVT as these are believed to be chief causative factors.^{13,26} In addition to asking the patient about potential causes for vessel trauma, inquiring about other risk factors discussed above should be included in the patient's initial evaluation (Table 1). In the absence of a local or obvious causative factor, the presence of an underlying occult condition should be considered and the patient may be required to undergo additional screening for coagulation abnormalities, particularly in cases where the thrombosis is extensive, migrant, or recurrent.^{22,27} Likewise, the presence of fever or leucocytosis may indicate that the patient is suffering from a systemic infection.¹³

In an otherwise healthy, young patient, where there is clinical suspicion of venous thrombosis in the upper ex-

Table 1.
Factors that should increase a practitioner's suspicion of SVT.

<ul style="list-style-type: none"> • Intravenous catheters • Intravenous injections or drug use • Venipuncture for blood work or donations • Prolonged immobilization • Recent surgery 	<ul style="list-style-type: none"> • Pregnancy • Use of oral contraceptives or hormone replacement therapy • Varicose veins or vein stripping • Inherited hypercoagulable condition • History of SVT or DVT
---	--

tr emity, consideration should also be given to a diagnosis of Effort Thrombosis, or Paget-Schroetter Syndrome. This is a relatively infrequent disorder, and refers to axillary-subclavian vein thrombosis associated with strenuous and repetitive or sustained upper extremity movements, such as those involved in sporting activities.^{28,29} Swelling and arm discomfort are the most frequent presenting problems associated with this condition, and unlike those suffering from upper extremity DVT secondary to central venous catheters, patients with effort thrombosis are usually symptomatic.²⁸

In most uncomplicated cases of SVT, a clinical diagnosis is established based on presenting signs and symptoms, however, due to the associated risk of DVT and PE, further evaluation may be necessary. Concomitant DVT has been reported in up to 44% of individuals with a diagnosis of SVT and is usually associated with extensive limb swelling.^{22,27} Furthermore, up to 33% of DVT's have been reported to have asymptomatic PE, and up to 13% have symptomatic PE.²⁷ In a systematic review and meta-analysis on the prevalence of DVT and PE in patients with SVT, roughly 18% and 7% of patients with SVT were found to have DVT and PE respectively.²⁰ For this reason, asking patients who present with confirmed or suspected SVT about symptoms of PE, such as dyspnea, pleuritic chest pain, and fever is important. Given that a diagnosis of SVT on the basis of clinical features alone is not always straightforward, as well as taking into consideration the supplemented risk of an associated DVT or PE, venous ultrasonography is now considered essential for both confirming the clinical diagnosis of SVT and for

ruling out concomitant DVT.^{4,23} Ultrasound may also be useful for excluding other differential diagnoses, such as cellulitis, panniculitis, erythema nodosum, insect bites, and lymphangitis.²⁴

Treatment of an SVT will depend on a number of factors, including the location and the underlying cause of the SVT, as well as the presence of concomitant DVT, varicose veins, and hypercoagulability disorders.^{1,13} The majority of patients are treated symptomatically with the goal of relieving pain, reducing erythema and swelling, and preventing potential complications or re-occurrence.^{1,13} These treatments may include local heat, anti-inflammatory agents, and compression.¹ In cases of upper extremity SVT secondary to an intravenous catheter, the device is first removed followed by conservative measures. Anticoagulation is seldom indicated for SVT unless there is progression or involvement of the deep venous system³⁰, inflammation is persistent in the affected area³¹, or venous reflux is demonstrated on duplex ultrasound at the saphenofemoral junction/ saphenopopliteal junction in association with varicose veins¹. If a hypercoagulable disorder is identified, treatment with long term anticoagulation may be warranted.¹

In this case, the patient underwent a course of soft tissue therapy over his area of complaint due to an initial differential diagnosis of muscular strain and peripheral nerve entrapment. This treatment exacerbated the patient's condition after a single session and prompted ancillary investigation. Local massage or manual therapy is generally contraindicated over an area of venous disease involvement (this may include phlebitis, thrombophlebitis, DVT and/or SVT) due to potential for worsening accompanying inflammation and pain.³² In the case of varicose veins, the area with varicosities should be avoided if applying pressure is pain provoking or if the patient has risk factors of clot formation.³² Although this patient's symptoms eventually resolved within five days once the appropriate diagnosis was made, unnecessary pain and discomfort could have been avoided.

Summary

This case demonstrates the importance for clinicians that work primarily in a musculoskeletal care setting to consider non-musculoskeletal causes for their patients' presentations when formulating differential diagnoses. With respect to SVT, the initial signs, symptoms, and

examination may not always point to a definitive diagnosis. Therefore, ensuring that key elements from the patient's history are acknowledged, such as possible trauma or underlying medical conditions, becomes essential. Due to the associated risks of DVT and PE, ultrasound evaluation of a suspected SVT may need to be performed. As such, communication with the patient's primary care physician, or other health care providers, may be necessary to help facilitate further diagnostic procedures. In the case of a confirmed SVT contributing to a patient's symptoms, it may be within the practitioner's scope to provide symptomatic relief and/or coordinate with other practitioners to provide interventions that may fall outside their abilities or legal restrictions. For chiropractors and other manual therapists, the main goal should be to identify these conditions early, to prevent unnecessary and potentially harmful treatments, and to ensure that the appropriate work-up is carried out.

References

1. Litzendorf ME, Satiani B. Superficial venous thrombosis: disease progression and evolving treatment approaches. *Vasc Health Risk Manag.* 2011;7: 569-575.
2. Roach RE, Lijfering WM, van Hylckama Vlieg A, Helmerhorst FM, Rosendaal FR, Cannegieter SC. The risk of venous thrombosis in individuals with a history of superficial vein thrombosis and acquired venous thrombotic risk factors. *Blood.* 2013;122(26): 4264-4269.
3. Decousus H, Quéré I, Presles E, Becker F, Barrellier MT, Chanut M, Gillet JL, Guennevez H, Leandri C, Mismetti P, Pichot O. Superficial venous thrombosis and venous thromboembolism: a large, prospective epidemiologic study. *Ann Intern Med.* 2010;152(4): 218-224.
4. Decousus H, Frappé P, Accassat S, Bertoletti L, Buchmuller A, Seffert B, Merah A, Becker F, Quéré I, Leizorovicz A. Epidemiology, diagnosis, treatment and management of superficial-vein thrombosis of the legs. *Best Pract Clin Haematol.* 2012;25(3): 275-284.
5. Coon WW, Willis PW, Keller JB. Venous thromboembolism and other venous disease in the Tecumseh community health study. *Circulation.* 1973;48(4): 839-846.
6. Cesarone MR, Belcaro G, Agus G, Georgiev M, Errichi BM, Marinucci R, Errichi S, Filippini A, Pellegrini L, Ledda A, Vinciguerra G. Management of superficial vein thrombosis and thrombophlebitis: status and expert opinion document. *Angiology.* 2007;58(1S): 7S-14S.
7. Ellis MH, Fajer S. A current approach to superficial vein thrombosis. *Eur J Haematol.* 2013;90(2): 85-88.

8. Kopansky-Giles DR, Grod JP, Crowther ER. Acute proximal deep vein thrombosis: presentation in a chiropractic office. *J Can Chiropr Assoc.* 1995;39(4):205-209.
9. Theiss JL, Fink ML, Gerber JP. Deep vein thrombosis in a young marathon athlete. *J Orthop Sports Phys Ther.* 2011;41(12): 942-947.
10. Christensen MG, Hyland JK, Goertz CM, Kollasch MW, Shotts BL, Blumlein NL, Johnson JM, Day AA, Smith MM. Practice analysis of Chiropractic 2015: A project report, survey analysis, and summary of chiropractic practice in the United States. National Board of Chiropractic Examiners; 2015.
11. Kalodiki E, Stvrtinova V, Allegra C, Andreozzi GM, Antignani PL, Avram R, Brkljacic B, Cadariou F, Dzsinič C, Fareed J, Gaspar L. Superficial vein thrombosis: a consensus statement. *Int Angiol.* 2012;31(3): 203-216.
12. Sobreira ML, Yoshida WB, Lastória S. Superficial thrombophlebitis: epidemiology, physiopathology, diagnosis and treatment. *J Vasc Bras.* 2008;7(2):131-143.
13. Cronenwett JL, Johnston KW. Rutherford's vascular surgery. 8th. ed. Philadelphia: Elsevier Health Sciences, 2014: 174, 209-211, 599-610, 833-839.
14. Poredoš P, Spirkoska A, Ježovnik MK. In patients with superficial vein thrombosis the inflammatory response is increased and related to the recanalization rate. *Arch Med Sci.* 2019;15(2): 393.
15. Leon L, Giannoukas AD, Dodd D, Chan P, Labropoulos N. Clinical significance of superficial vein thrombosis. *Eur J Vasc Endovasc Surg.* 2005;29(1): 10-17.
16. Meissner MH, Wakefield TW, Ascher E, Caprini JA, Comerota AJ, Eklof B, Gillespie DL, Greenfield LJ, He AR, Henke PK, Hingorani A. Acute venous disease: venous thrombosis and venous trauma. *J Vasc Surg.* 2007; 46(6):S25-S53.
17. Sarica-Kucukoglu R, Akdag-Kose A, Kayabali M, Yazganoglu KD, Disci R, Erzenin D, Azizlerli G. Vascular involvement in Behçet's disease: a retrospective analysis of 2319 cases. *Int J Dermatol.* 2006;45(8): 919-921.
18. Olin JW, Shih A. Thromboangiitis obliterans (Buerger's disease). *Curr Opin Rheumatol.* 2006; 18(1):18-24.
19. Decousus H, Epinat M, Guillot K, Quenet S, Boissier C, Tardy B. Superficial vein thrombosis: risk factors, diagnosis, and treatment. *Curr Opin Pulm Med.* 2003;9(5): 393-397.
20. Di Minno MN, Ambrosino P, Ambrosini F, Tremoli E, Di Minno G, Dentali F. Prevalence of deep vein thrombosis and PE in patients with superficial vein thrombosis: a systematic review and meta-analysis. *J Thromb Haemost.* 2016;14(5): 964-972.
21. Heil J, Miesbach W, Vogl T, Bechstein WO, Reinisch A. Deep vein thrombosis of the upper extremity: a systematic review. *Deutsches Ärzteblatt Int.* 2017;114(14): 244.
22. Nasr H, Scriven JM. Superficial thrombophlebitis (superficial venous thrombosis). *BMJ.* 2015;350: h2039.
23. Marchiori A, Mosena L, Prandoni P. Superficial vein thrombosis: risk factors, diagnosis, and treatment. *Semin Thromb Hemost.* 2006;32(7): 737-743.
24. Prandoni P, Polistena P, Bernardi E, Cogo A, Casara D, Verlato F, Angelini F, Simioni P, Signorini GP, Benedetti L, Girolami A. Upper-extremity deep vein thrombosis: risk factors, diagnosis, and complications. *Arch Intern Med.* 1997;157(1): 57-62.
25. Sassu GP, Chisholm CD, Howell JM, Huang E. A rare etiology for PE: basilic vein thrombosis. *J Emerg Med.* 1990;8(1): 45-49.
26. Bleker SM, van Es N, Kleinjan A, Büller HR, Kamphuisen PW, Aggarwal A, Beyer-Westendorf J, Camporese G, Cosmi B, Gary T, Ghirarduzzi A. Current management strategies and long-term clinical outcomes of upper extremity venous thrombosis. *J Thromb Haemostas.* 2016;14(5): 973-981.
27. Di Nisio M, Wichers IM, Middeldorp S. Treatment for superficial thrombophlebitis of the leg. *Cochrane Database Syst Rev.* 2013(4).
28. Roche-Nagle G, Ryan R, Barry M, Brophy D. Effort thrombosis of the upper extremity in a young sportsman: Paget-Schroetter syndrome. *Br J Sports Med.* 2007;41(8): 540-541.
29. Alla VM, Natarajan N, Kaushik M, Warriar R, Nair CK. Paget-Schroetter syndrome: review of pathogenesis and treatment of effort thrombosis. *Western J Emerg Med.* 2010;11(4): 358.
30. Stevens SM. Review: Fondaparinux reduces VTE and recurrence in superficial thrombophlebitis of the leg. *Ann Intern Med.* 2012;157(4): JC2-JC4.
31. Prandoni P, Tormene D, Pesavento R. High vs. low doses of low-molecular-weight heparin for the treatment of superficial vein thrombosis of the legs: a double-blind, randomized trial. *J Thromb Haemost.* 2005;3(6): 1152-1157.
32. Salvo SG. Mosby's pathology for massage therapists. Elsevier Health Sciences; 2013 Dec

Potential publication bias in chiropractic and spinal manipulation research listed on clinicaltrials.gov

Breanne M. Wells, DC, MS¹

Dana Lawrence, DC, MMedEd, MA²

Introduction: Clinical trial registries are used to help improve transparency in trial reporting. Our study aimed to identify potential publication bias in chiropractic and spinal manipulation research by assessing data drawn from published studies listed in clinicaltrials.gov.

Methods: We searched the clinicaltrials.gov registry database for completed trials tagged with the key indexing terms chiropractic or spinal manipulation. We assessed if the trial registry had been updated with data, then searched for publications corresponding to the registered trials. Finally, the frequency of positive or negative results was determined from published studies.

Results: For the term ‘chiropractic’, 63% of studies supported the intervention and 52% supported the intervention for the term ‘spinal manipulation’.

Discussion: Publication bias in chiropractic and spinal manipulation research listed in clinicaltrials.gov

Introduction : Les registres des essais cliniques servent à accroître la transparence des rapports sur les essais. Notre étude visait à trouver les éventuels partis pris de publication dans les travaux de recherche sur la chiropratique et les manipulations vertébrales à l’aide de données tirées d’études publiées et répertoriées dans clinicaltrials.gov.

Méthodologie : Dans la base de données du registre clinicaltrials.gov, nous avons cherché des essais terminés marqués par les termes d’indexation « chiropratique » et « manipulation vertébrale ». Nous avons cherché à savoir si le registre des essais avait été mis à jour par l’ajout de données, puis nous avons recherché les publications correspondant aux essais répertoriés. Enfin, nous avons établi la fréquence des résultats positifs et négatifs à l’aide des études publiées.

Résultats : Avec le terme « chiropratique », 63 % des études étaient en faveur des interventions; avec l’expression « manipulation vertébrale », 52 % des études étaient en faveur des interventions.

Discussion : Il semble y avoir un parti pris de publication dans les travaux de recherche sur la chiropratique et les manipulations vertébrales

¹ Palmer College of Chiropractic

² Parker University

Corresponding author: Breanne Wells, 1000 Brady St. Davenport, IA 52803

Tel: 563-884-5334

E-mail: breanne.wells@palmer.edu

© JCCA 2020

The authors have no disclaimers, competing interests, or sources of support or funding to report in the preparation of this manuscript.

appears to occur. Further work may help understand why this happens and what may be done to mitigate this moving forward.

(JCCA. 2020;64(1):82-87)

KEY WORDS: chiropractic, publication bias, scientific journals

Introduction

Publication bias is defined as “the tendency to publish reports of research that appears to support a hypothesis and to refrain from publishing findings that do not, thereby creating opinions about the truth of the intervention that may be unduly optimistic”.¹ This may result from fear of rejection, failure to submit findings with negative results, failure to accept such papers by journal editors, or failure to submit information by those with vested interest in the results. This is important because exclusion of studies at a review or study level may not provide an accurate representation of aggregate study results, which could impact recommendations drawn from systematic reviews and meta-analysis.² Additionally, stakeholders need complete information to make decisions about the effectiveness of a given intervention.³

Clinical trials are essential for providing information on how treatments compare to one another for a given condition.⁴ Researchers in the United States who initiated studies after September 2007 are required to register clinical trials onto registries such as the clinicaltrials.gov website.⁵ The International Committee of Medical Journal Editors also recommends this to all authors conducting clinical trials.⁶ Trials databases are specifically designed to prevent selective publication and selecting reporting of research outcomes. Trial registries also provide a venue where information from study results can be made public. They are, further, a source of information for non-published, yet completed, clinical trials.⁷ However, doubts exist that trial registries are an effective method for reducing publication bias.⁸ Approximately half of trials fail to report results in a clinical trial registry.⁹

To study publication bias, one can search an electronic clinical trial registry to identify relevant studies for a

répertoriés dans clinicaltrials.gov. D'autres travaux pourraient aider à comprendre la cause de ce phénomène et à trouver des moyens de le réduire dans l'avenir.

(JCCA. 2020;64(1):82-87)

MOTS CLÉS : chiropratique, biais de publication, revues scientifiques

given area of research. This type of search examines what is known as *gray literature*, which includes unpublished studies and studies never submitted to peer review.² In one meta-analysis of 28 special education journals, less than half included gray literature and only 33% addressed publication bias.² Researchers of this study concluded that not including the gray literature is associated with an increased risk of publication bias.²

Goldacre encourages others to explore publication bias for specific interventions to identify its prevalence.⁹ We could locate no information about the prevalence of publication bias in chiropractic and spinal manipulation clinical trials. The primary objective of our study was to identify potential publication bias related to *chiropractic* and *spinal manipulation* trials.

Methods

We used a 4-step process to meet our primary objective. The first step determined the number of completed trials in clinicaltrials.gov listed under the key indexing terms *chiropractic* and *spinal manipulation* research. A second step determined if any results were posted on the clinicaltrials.gov database. Third, we looked at the publication section on clinicaltrials.gov to determine if there were any publications associated with the studies. Finally, we determined the proportion of publications whose results favored the intervention.

Search strategy

Data collection took place from May 2018-August 2018 for the term *chiropractic* and from February 2019-April 2019 for the term *spinal manipulation*. To complete our first objective, identifying potential publication bias, we searched clinicaltrials.gov looking for studies with

the term *chiropractic* or *spinal manipulation*, using the *advanced search* category. In the 'other terms' box, we first searched the term *chiropractic* and then conducted a second search for the term *spinal manipulation*. In the *study type and study results* box, we choose *all studies*. In the targeted search box, for intervention/treatment, we searched *chiropractic* and then *spinal manipulation*. For locations, we selected studies in the USA. We searched all funder types, phases, ages, genders, and start dates. We included all studies that provided results using this search strategy. We restarted the search in clinicaltrials.gov each time we began to search for articles to ensure all studies available were included by the end of data collection. Finally, the results of the spreadsheet and the article abstracts were compared between two reviewers.

Article eligibility criteria were based on criteria from a prevalence study of clinical trials on clinicaltrials.gov by Fleming and Goldacre.⁷ We considered a clinical trial still in progress if it had one of the following statuses: 'Active, not recruiting,' 'Available,' 'Enrolling by invitation,' 'Not yet recruiting,' 'Recruiting,' or 'Suspended.' Articles in progress were excluded from the additional comparison search for published articles but were still tallied. Studies with a status of 'Withdrawn,' 'Withheld,' 'No longer available,' and 'Temporarily not available' were also excluded.⁷ Trials were considered completed if they had a status of 'Completed' or 'Terminated.'⁷

Updated results

To determine if results were posted on clinicaltrials.gov database, we looked under the results tab to see if it had results or if it said, 'no results posted'. If there was any information in the results section, we considered it to have provided results.

Search for publications

To find publications linked to the studies posted in the database, we examined the publication section of clinicaltrials.gov. If there were any publications listed, we tallied the number on the spreadsheet. If the publication was a study protocol, we still counted it as a publication, but did not use it in our assessment of publication bias.

Assessment of potential publication bias

After completing the clinicaltrials.gov search for eligible articles, we determined if results from included trials were

published. To do this, we analyzed all articles associated with those trials posted on the clinicaltrials.gov database. We developed a spreadsheet that noted if the trial had posted results, the number of published papers related to the trial, and whether or not the published paper favored the effectiveness of the experimental intervention. We also had a category called *mixed results*, for when the results of the study did not appear to favor or reject the intervention. Thus, the conclusions were classified as in favor of, against, or mixed results. This was determined by reading the abstract, results and conclusions of each study. Results were tallied and verified by 2 independent coders and placed on an Excel spreadsheet.

Results

For our primary objective, we found 65 studies under the key term *chiropractic*. Six studies had been terminated; 59 were complete. Ten of those studies posted results on clinicaltrials.gov. There were 64 total published articles, eight of which were study protocols. As noted above, we did not include the study protocols in the analysis, leaving 56 articles to analyze. Thirty-five studies supported the intervention, eight did not, and 13 had mixed results.

Searching *spinal manipulation* showed similar results. There were 76 total studies. Five studies had been terminated, and 71 were complete. Thirteen of the 71 studies posted results on clinicaltrials.gov. There were 97 total publications from the 71 studies. Twenty-five of the studies were study protocols, and were not included, leaving 72 total articles. Forty-nine of the studies supported the intervention, five did not and 18 had mixed results. Complete search results are shown in Table 1.

Discussion

The existence of publication bias in chiropractic and spinal manipulation research cannot be ruled out. There are trials with results not yet posted on clinicaltrials.gov, as well as completed studies without published results. Additionally, results often skew in favor of the intervention. Our results harmonize with existing literature that also show publication bias exists in research for other professions.⁸ A study investigating the evidence of publication bias in oncology research found that it was more likely to report positive findings when the trial was registered in advance. Larger sample sizes, with non-stringent blinding, were more likely to report that the drug had favorable results⁸

Table 1.
Results from searching chiropractic and spinal manipulation on clinicaltrials.gov.

	Chiropractic	Spinal Manipulation
Total studies	65	76
Completed	59	71
Terminated	6	5
Results posted	10	13
Studies that published	22	38
Total published articles from studies that published	64	97
Studies that only published study protocols	8	3
Total study protocols	19	25
Articles in favor of hypothesis	35	49
Articles against hypothesis	8	5
Mixed conclusions	13	18

Publication bias occurs across disciplines. It has been seen in anesthesiology¹⁰, gastroenterology¹¹, dermatology¹², again in oncology¹³, and even organizational sciences¹⁴. Hermann and colleagues examined publication bias in clinical oncology reviews. They examined systematic reviews published in the top five highest impact factor oncology journals, for the years 2007-2015. Out of 182 reviews, only 57 reported publication bias evaluations.¹⁵ It is fair to say this is now both an endemic and epidemic problem.

In addition to publication bias present in research from other professions, it is also present in other countries. A retrospective study analyzing data in the European Clinical Trials Register revealed that out of 7274 trials, only 49.5% reported results. To comply with the European Commission, trials are required to post results within 12 months of the completion date, but half of trials fail to do this. Large studies with a commercial sponsor were more likely to post positive results than those without a commercial sponsor.¹⁶ This correlates with another study stating that for-profit funded research is associated with publication bias as well as with non-publication of trial results.¹⁷

Possible reasons for failure to publish are that researchers need time to analyze and report their data. Studies that have not completed recruiting would not be expected to publish. Goldacre and Powell Smith argue that live, ongoing monitoring of trials and the imposition of negative

consequences for withholding trial results may help decrease publication bias.⁹ A study exploring methods to reduce publication bias found that editors thought mandatory publication would be the most effective method, while researchers thought a two-stage review would be more effective.^{2,18} As early as 1990, Kay Dickersin argued in *JAMA* that publication bias was a growing problem.¹⁹

The presence of publication bias is responsible for the growth of clinical trials registries.²⁰ The continued reporting of trials with statistically significant results and not those without skews the results of a systematic review or meta-analysis.^{21,22} Some have argued that the prestigious Cochrane Collaboration- which is well aware of the many issues surrounding publication bias- may actually help amplify the effects of bias. Jefferson has argued that one can identify “subtle distortions, discrepancies and missing information” when reading a short synopsis of a huge data set, which Cochrane creates for each report.²³ However it may be, what is clear is that if nonsignificant or unfavorable findings are withheld from publication, results of any review or meta-analysis will skew in favor of the intervention under study.

The International Committee of Medical Journal Editors recommends that editors should be careful when assessing research to ensure the results are valid and there are no additional outcomes added. They also urge editors to avoid not publishing articles because of lack of statistical significance.²⁴ Negative studies are every bit as

important as positive ones; publishing null findings can identify ineffective practices and inform and produce new theories and research. It also is a more accurate representation of the current state of knowledge. Thus, being aware of null findings is crucial when examining the effectiveness and limitations of a given intervention. Authors and editors should publish their research, whether or not the results are statistically significant. Publishing null findings are important because they help shape the knowledge base and guide clinical practice.³

Limitations

We limited our assessment of publication bias in chiropractic and spinal manipulation to the sole US trials database, clinicaltrials.gov.²⁵ In the United States, it is a legal requirement to update a registry after completion within one year of completion of a trial. We studied abstracts of articles, rather than the full paper. We searched the terms *chiropractic* and *spinal manipulation* and did not exclude studies from other professions; thus, the results are not specific to just chiropractic. Additionally, only two reviewers analyzed the data; more reviewers might provide a more well-rounded picture of publication bias.

Conclusion

There is evidence of possible publication bias in chiropractic and spinal manipulation research. Action steps should be taken to reduce publication bias, including publishing completed research regardless of the outcome and timely posting of results to clinicaltrials.gov. Future research should focus on the reasons why this bias exists and what may be done to mitigate its presence in our literature.

References

1. Collins Dictionary of Medicine. 2005; <https://medical-dictionary.thefreedictionary.com/publication-bias>. Accessed April 2, 2018.
2. Gage N, Cook B, Reichow B. Publication bias in special education meta-analyses. *Except Child*. 2017;83: 428-445.
3. Cook B, Therrien W. Null effects and publication bias in special education research. *Behav Disord*. 2017;42: 149-158.
4. Heneghan C, Goldacre B, Mahtani K. Why clinical trial outcomes fail to translate into benefits for patients. *Trials*. 2017;18(1): 122.
5. ClinicalTrials.gov FDAAA 801 and the Final Rule. <https://>

- clinicaltrials.gov/ct2/manage-recs/fdaaa#PedPostmarket. Accessed August 20, 2019.
6. Journals stating that they follow the ICMJE Recommendations.2018; <http://www.icmje.org/journals-following-the-icmje-recommendations>. Accessed August 20, 2019.
7. Fleming B. Prevalence of clinical trial status discrepancies: A cross-sectional study of 10492 trials registered on both ClinicalTrials.gov and the European Union Clinical Trials Register. *PLoS One*. 2018;3: 1-8.
8. Rasmussen N, Lee K, Bero L. Association of trial registration with the results and conclusions of published trials of new oncology drugs. *Trials*. 2009;10:116.
9. Goldacre B. The TrialsTracker: automated ongoing monitoring of failure to share clinical trial results by all major companies and research institutions. *F1000 Res*. 2016;5:1-14.
10. Hedin R, Umberham B, Detweiler B, Kollmorgen L, Vassar M. Publication bias and nonreporting found in majority of systematic reviews and meta-analyses in anesthesiology journals. *Anesth Analg*. 2016;123: 1018-1025.
11. Heavener T. A review of publication bias in the gastroenterology literature. *Indian J Gastroenterol*. 2018;37: 58-62.
12. Atakpo P. Publication bias in dermatology systematic reviews and meta-analyses. *J Dermatol Sci*. 2016;82:69-74.
13. Lundh A, Knijnenburg L, Jorgensen AW, van Dalen EC, Kremer LC. Quality of systematic reviews in pediatric oncology-a systematic review. *Cancer Treat Rev*. 2009;35: 645-652.
14. Kepes S, Banks G, McDaniel M, Whetzel D. Publication bias in the organizational sciences. *Organ Res Methods*. 2012;15: 624-662.
15. Herrmann D, Sinnott P, Holmes J, Khan S, Koller C, Vassar M. Statistical controversies in clinical research: publication bias evaluations are not routinely conducted in clinical oncology systematic reviews. *Ann Oncol Off J Eur Soc Med Oncol*. 2017; 28: 931-937.
16. Goldacre B, Devito N, Heneghan C et al. Compliance with requirement to report results on the EU Clinical Trials Register: cohort study and web resource. *Br Med J*. 2018;362: K3218.
17. Falk Delgado A. Outcome switching in randomized controlled oncology trials reporting on surrogate endpoints: a cross-sectional analysis. *Sci Rep*. 2017;7: 9206.
18. Carroll H, Toumpakari Z, Johnson L, Betts J. The perceived feasibility of methods to reduce publication bias. *PLoS One*. 2017;12: e0186472.
19. Dickersin K. The existence of publication bias and risk factors for its occurrence. *J Am Med Assoc*. 1990;263: 1385-1389.

20. Abaid LN, Grimes DA, Schulz KF. Reducing publication bias through trial registration. *Obstet Gynecol.* 2007; 109:1434-1437.
21. Unger J, Barlow W, Ramsey S, LeBlanc M, Blanke C, Hershman D. The scientific impact of positive and negative phase 3 cancer clinical trials. *JAMA Oncol.* 2016; 2(7): 875-881.
22. Onishi A, Furukawa T. Publication bias is underreported in systematic reviews published in high-impact-factor journals: metaepidemiologic study. *J Clin Epidemiol.* 2014;67: 1320-1326.
23. T J. How Cochrane is doing pharma a good turn. <https://blogs.bmj.com/bmj/2018/11/12/tom-jefferson-cochrane-pharma-good-turn/>, accessed April 24, 2019.
24. ICMJE T. Uniform requirements for manuscripts submitted to biomedical journals: writing and editing for biomedical publication updated October 2007. *J Bangladesh Soc Physiol.* 2007;2: 60-88.
25. Protections for human research. Office for human research protections, listing of clinical trial registries. <https://www.hhs.gov/ohrp/international/clinical-trial-registries/index.html>. 2015. Accessed October 4, 2019.

Tibialis anterior herniation – a rare clinical entity: a case report and review of the literature

Ali Masoumi, BSc, DC, MSc, RCCSS(C) resident^{1,2}
Gerry Ramogida, BSc, DC¹

Objectives: *To present a case of a tibialis anterior muscle herniation in a soccer player.*

Clinical features: *A 28-year-old male soccer player presented with a trauma-induced injury to his right anterior shin. After assessment and due to his clinical signs and symptoms, a high suspicion of tibialis anterior muscle herniation was suspected.*

Intervention and outcomes: *Diagnostic ultrasound confirmed this diagnosis, and a trial of conservative therapy was recommended. After eight weeks of treatment, he was able to return to sport.*

Summary: *A trial of conservative treatment for the tibialis anterior muscle hernia should be included as a part of general treatment strategy prior to any surgical interventions.*

(JCCA. 2020;64(1):88-91)

KEY WORDS: tibialis anterior herniation, muscle herniation, soft tissue herniation, chiropractic

Objectifs : *Présenter un cas d'une hernie tibiale antérieure chez un joueur de soccer.*

Caractéristiques cliniques : *Joueur de soccer de 28 ans présentant une blessure traumatique. Après examen et en raison de ses signes et symptômes cliniques, on a fortement suspecté une hernie tibiale antérieure.*

Intervention et résultats : *L'échographie a confirmé ce diagnostic et un essai de traitement conservateur a été recommandé. Après huit semaines de traitement, il a pu reprendre une activité sportive.*

Résumé : *Avant toute chirurgie, un essai de traitement conservateur de la hernie tibiale antérieure doit être inclus dans la stratégie générale de traitement.*

(JCCA. 2020;64(1):88-91)

MOTS CLÉS : hernie tibiale antérieure, hernie musculaire, hernie des tissus mous, chiropratique.

¹ Private practice

² Royal College of Chiropractic Sports Sciences (Canada)

Corresponding author:

Ali Masoumi, 126 – 15850 26th Ave, Surrey, BC, V3Z 2N6

E-mail: ali@drmasoumi.com

Tel: 604-721-4881

© JCCA 2020

The authors have no disclaimers, competing interests, or sources of support or funding to report in the preparation of this manuscript.

The involved patient provided consent for case publication.

Introduction

Muscle herniation, also known as myofascial defect, is the protrusion of a muscle through the surrounding fascia. This type of herniation, which is a relatively atypical clinical entity, has been rarely discussed in the dermatological and musculoskeletal literature.¹ The clinician's ability to differentially diagnose and treat this type of injury from other similar looking pathologies such as lipomas, hematomas and fibromas is of great importance.² Correct diagnosis will prevent unnecessary skin biopsy and the potential psychological side effects for patients due to a mistakenly diagnosed serious pathology.

Tibialis anterior muscle herniation often presents as a distinct palpable swelling or nodule over the muscle especially with weight bearing and muscle contraction.¹ The swelling tends to shrink in size with muscle inactivation or when the patient is non-weightbearing or supine.³⁻⁵ Even though clinical signs and symptoms have been the gold standard for the diagnosis of this condition, the recent use of dynamic ultrasound for the diagnosis of different muscle herniation has surged in popularity among healthcare professionals.²

The conservative management of tibialis anterior muscle herniation has not yet been discussed or researched in depth. The purpose of this case report is to highlight the successful conservative management of a tibialis anterior muscle herniation.

Case presentation

A 28-year-old male soccer player was referred by his family physician for his persistent localized swelling and pain over his right anterior shin of three months duration. His injury was the result of a direct trauma (slide tackling) from the left side in a soccer match. He was unable to continue playing due to the severity of pain. The pain was described as a dull, localized pain with an intensity of 7/10 on a Numerical Pain Rating Scale (NPRS). Weight bearing and intense physical training increased his symptoms. There was no numbness, tingling or weakness in the lower extremities. His medical and social histories were unremarkable.

Plain film imaging ordered by his family physician to rule out a potential fracture, and a complete neurovascular examination were within normal limits. There was palpable swelling over the belly of the tibialis anterior muscle, which would decrease in size when the patient was in a



Figure 1.
An example of Fencer's lunge position causing tibialis anterior herniation.

supine position. Fencer's lunge position (Figure 1), which will increase the strain on the tibialis anterior muscle, increased the localized swelling and pain. During gait analysis, using a treadmill, a mild bilateral subtalar overpronation during mid-stance and a five degree decrease in the ankle dorsiflexion was noted. Other orthopedic and functional assessments of the lower extremity were unremarkable.

Due to the high positional variability of the lesion, presenting history and the absence of red flags, muscle herniation was highly suspected and a diagnostic ultrasound was ordered.

The diagnostic ultrasound confirmed a 1 cm hypoechoic lesion of the tibialis anterior muscle in the transverse plane, showing a loss of continuity in the surrounding fascia. Conservative management of this condition was recommended to the patient with a possibility of a surgical referral if there were no significant changes in his symptomology within a month.

Based on the available scientific literature, conserv-

ative treatment was limited to load modifications and compression stockings. Due to the pathophysiology of this condition, supine isometric contraction of the tibialis anterior was recommended for the first two weeks, with eventual progression to concentric and eccentric exercises. Home stretches and mobility exercises targeting the ankle range of motion especially in dorsiflexion were prescribed. Custom made orthotics to alleviate the tensile load and contraction of the tibialis anterior during heel strike was also prescribed.

After eight weeks of conservative management, his reported pain intensity was substantially reduced to 1/10 on the NPRS and he had a small reduction in the observed size of his muscle hernia. At this time, he was cleared to return to sport and to resume previous activities. A two-month follow-up after his medical release was unremarkable as he continued to enjoy playing soccer with a visible muscle hernia, pain free.

Discussion

Muscular hernia, which is the protrusion of the muscle through its surrounding fascia, is a rare clinical finding.² There have been about 200 cases of extremity muscle herniation reported in the literature with the majority occurring in the tibialis anterior muscle.¹ The extensor digitorum muscle of the forearm is the other common reported site of herniation.¹

Due to its rare occurrence and the limited literature on the topic, this condition is routinely misdiagnosed as a serious pathology such as a lipoma, hematoma or fibroma.² A lack of clinical and orthopedic red flags and the often traumatic nature of this condition should guide the clinician away from unnecessarily procedures such as skin biopsy.²

In the past, clinical findings such the appearance of a focal swelling with weight bearing and the disappearance or the decrease in its size when supine was sufficient criteria for the diagnosis of tibialis anterior muscle herniation. Currently the use of diagnostic ultrasound is the gold standard for such diagnosis.⁴ MRI is the imaging modality of choice if conservative treatments fail and surgical treatment is recommended.¹⁰ MRI allows better visualization of the musculofascial demarcation, determination of herniated muscle volume, and will assist with surgical planning by assessing the neighboring neuromusculoskeletal tissues.¹⁰

Muscle hernias can be classified into traumatic and constitutional in origin. In this case, a single slide tackle from the opposing player and the direct trauma to the anterior shin presumably disrupted the superficial and deep fascia membrane surrounding the tibialis anterior muscle, causing localized herniation. Constitutional muscle hernia origins can either be congenital or due to the increased intracompartmental pressure from excessive muscular exertion and exercise.^{1,6,7}

Treatments for muscle herniation are controversial. Asymptomatic tibialis anterior herniation typically requires no specific treatment except patient assurance and education.³ The only conservative management for painful hernias cited in the literature are rest, load modification, and compression stockings.⁸

In this case, isometric contraction of the tibialis anterior muscle, which will decrease pain and increase motor neural recruitment, was added for the first two weeks. Bement *et al.*¹¹ demonstrated the analgesic effect of isometric contraction as they suggested that the activation of high-threshold motor units is involved in exercise-induced analgesia. These exercises were recommended in the supine position to minimize any potential intracompartmental pressure from weight bearing.

Concentric contraction in supine and in weight bearing were recommended to the patient for the following two weeks, followed by eccentric exercises to generate force at greater length and to stimulate maximal tissue adaptation to elastic force.¹² The elastic energy stored during the lengthening phase of the eccentric contraction can be used during the shortening phase of muscular contraction to amplify force and power production during exercise.¹² In the last stage of rehabilitation, sports-specific plyometric exercises were introduced to generate multidirectional force and stability through neural adaptation which enhances proprioception and kinesthesia required when playing soccer.¹³

If conservative treatments fail, surgery is an option. Traditionally, the surgical technique was the direct closure of the fascia defect by tightening the area.³ However this procedure has lost its popularity due to a high muscular herniation recurrence rate and increased intracompartmental pressure which predisposed the patient to compartment syndrome.⁹ Recent studies suggest a newer approach of longitudinal decompressive fasciotomy or repair with synthetic patches.³ Hegde recommended the

closure of the facial defect with an autologous tensor fascia lata graft.⁵

In this case, conservative management had a very good outcome in eight weeks allowing full recovery and participation in sport. These results, however, should be interpreted with caution, as many important variables such as the natural history of this condition may have played a role in this case. Furthermore, there are no randomized control studies comparing different types of conservative management in tibialis anterior herniation.

Summary

Tibialis anterior muscle hernia is a rare clinical condition and can be a differential diagnosis of patients with localized leg pain and focal swelling. Direct trauma, size variability of the lesion with and without weight bearing, and the absence of red flags in the history should assist the clinician with this diagnosis. Diagnostic ultrasound can confirm this diagnosis, which will prevent the unnecessary use of other imaging modalities such as MRI and CT scan. Conservative management including the use of therapeutic exercises, load modifications, and compression stockings alongside optimal functional movements in the lower extremity can be prescribed prior to potential surgical referral.

References

1. Kim M, Hong SP, Hawng SM, Park H, Ahn SK. Tibialis anterior muscle herniation development after trauma. *Int J Dermatol.* 2008;47: 845-847.
2. Zeiss J, Ebraheim NA, Woldenberg LS. Magnetic resonance imaging in the diagnosis of anterior tibialis muscle herniation. *Clin Orthop Rel Res.* 1989; 244: 249.
3. Donmez G, Evrenos M, Cereb M, Karanfil Y, Doral M. Double layer repair of tibialis anterior muscle hernia in a soccer player: a case report and review of the literature. *Muscle Ligament Tendon J.* 2015;5(4): 331-334.
4. Artul S, Habib G. The importance of dynamic ultrasound in the diagnosis of tibialis anterior muscle herniation. *Crit Ultrasound J.* 2014; 6(14).
5. Hegde AS. An interesting case of post traumatic tibialis anterior muscle herniation. *Kathmandu Univ Med J.* 2013; 11(44):332-334.
6. Bergmann G, Ciritsis BD, Wanner GA, Simmen HP, Werner CM, Osterhoff G. Gastrocnemius muscle herniation as a rare differential diagnosis of ankle sprain. *Patient Saf Surg.* 2012; 14:6(1): 5.
7. Angadi DS, Rampaul RS, Akthar I, Makdhoomi K. Bilateral muscle hernias of the anterior tibialis muscle. *Foot Ankle Int.* 2007;28(4): 520.
8. Marchalik D, Lipsky A, Petrov D, et al. Dermatological presentations of orthopedic pathologies: a review of diagnosis and treatment. *Am J Clin Dermatol.* 2012;13: 293-310.
9. Miniaci A, Rorabeck CH. Compartment syndrome as a complication of repair of a hernia of the tibialis anterior. *J Bone Joint Surg.* 1986;68: 1444-1445.
10. Ulu M, Cakmak M, Nas K. A case of tibialis anterior muscle herniation diagnosed by ultrasonography. *Turk J Physical Med Rehab.* 2013;59: 73-75.
11. Bement H, Dicapo J, Rasiarmos R, Hunter SK. Does response of isometric contractions on pain perception in healthy adults. *Med Sci Sports Exerc.* 2008;40(11): 1880-1889
12. LaStayo P, Woolf JM, Lewek MD, Mackler L, Reich T, Lindstedt SL. Eccentric muscle contractions: their contribution to injury, prevention, and sport. *J Orthopedic Sport Phys Ther.* 2003;33: 557-571.
13. Behrens M, Mau-Moeller A, Bruhn S. *Int J Sports Med.* Effect of plyometric training on neural and mechanical properties of the knee extensor muscles. 2014;35(2).