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Chiropractic integration within a community health centre: a cost description and partial analysis of cost-utility from the perspective of the institution

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Objective: *To evaluate costs and consequences of a new back pain service provided by chiropractors integrated into a Community Health Centre in Cambridge, Ontario. The study sample included 95 consecutive patients presenting between January 2014 to January 2016 with a mixture of sub-acute and chronic back pain.*

Methods: *A secondary cost-utility analysis was performed and conducted from the perspective of the healthcare institution. Cost-utility was calculated as cost per quality-adjusted life year (QALY) gained over a time horizon of 90 days.*

Results: *According to the EuroQol 5 Domain questionnaire, nearly 70% of patients improved. The*

Objectif : *Évaluer les coûts et les conséquences d'un nouveau service pour soulager les maux de dos offert par des chiropraticiens intégrés à un centre de santé communautaire à Cambridge, en Ontario. L'échantillon de l'étude comprenait 95 patients qui se sont présentés consécutivement entre janvier 2014 et janvier 2016 et qui étaient atteints de diverses douleurs dorsales subaiguës et chroniques.*

Méthodes : *Une analyse coût-utilité secondaire a été effectuée du point de vue de l'établissement de santé. Le coût-utilité a été calculé en tant que coût par année de vie ajustée en fonction de la qualité (AVAQ) obtenu sur une période de 90 jours.*

Résultats : *Selon les résultats du questionnaire EuroQol 5 Domain, près de 70 % des patients ont*

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mean number of treatment sessions was 8.4, and an average of 0.21 QALYs were gained at an average cost per QALY of $1,042. Seventy-seven percent of patients did not visit their primary care provider over the 90-day period, representing potential cost savings to the institution of between $2,022.23 and $6,135.82.

Conclusion: *Adding chiropractic care to usual medical care was associated with improved outcomes at a reasonable cost in a sample of complex patients with sub-acute and chronic back pain. Future comparative cost-effectiveness studies are needed.*

(JCCA. 2019;63(2):64-79)

KEY WORDS: health services research, cost analysis, community health centres, chiropractic

observé une amélioration de leurs symptômes. Le nombre moyen de séances de traitement était de 8,4 et une moyenne de 0,21 AVAQ a été obtenue à un coût moyen par AVAQ de 1 042 $. Soixante-dix-sept pour cent des patients n'ont pas consulté leur fournisseur de soins primaires au cours de la période de 90 jours, ce qui représente des économies potentielles de l'ordre de 2 022,23 $ à 6 135,82 $ pour l'établissement.

Conclusion : *L'ajout de soins chiropratiques aux soins médicaux habituels a entraîné une amélioration des résultats à un coût raisonnable pour un échantillon de patients ayant des besoins complexes et présentant des douleurs dorsales subaiguës et chroniques. De futures études comparatives coût-efficacité sont nécessaires.*

(JACC. 2019;63(2):64-79)

MOTS CLÉS : recherche sur les services de santé, analyse des coûts, centres de santé communautaire, chiropratique.

Introduction

The efficacy for chiropractic care including manipulative therapy in treating patients with spine-related pain has been established.[1-3] Moreover, clinical trials have shown greater efficacy when chiropractic treatment is added to usual medical care in managing patients with these conditions.[4,5] Chiropractic care also has the potential to be cost-effective, as chiropractors typically use a conservative (i.e. non-pharmacological, non-surgical) approach. In fact, studies have shown that when managed by a chiropractor, even when controlling for confounding factors, patients tend to have fewer advanced medical procedures including opioids and referrals for diagnostic imaging, injections, or spinal surgery.[6-8]

Although chiropractic care has been suggested as a cost-effective alternative to the medical management of spine-related pain disorders[9], the evidence for economic evaluations of chiropractic treatment compared with, or added to, medical care is inconclusive[10]. Regardless, back and neck pain remain prevalent and costly in society. The socioeconomic burden of these disorders, in terms of health resource utilization and economic costs (e.g. lost productivity), is particularly high among vulnerable populations.[9,11] In Canada, such populations commonly receive health care services at the primary care level within Community Health Centres (CHCs).[12]

CHCs are non-profit, publicly funded and community-governed organizations that promote health, illness prevention, and community development.[12] These centres employ multidisciplinary teams of health professionals that traditionally include medical doctors, nurse practitioners, registered nurses, dieticians, social workers, and community health workers. CHCs are distinct from other primary care centres in that they serve the needs of complex patient populations, typically including younger individuals, the socioeconomically disadvantaged, people with severe mental health or addiction issues (e.g. opioid over-use), and those with various other co-morbidities.[12,13] Musculoskeletal disorders including chronic back pain are also prevalent among these groups.[9,14,15] To improve the management of these patients, particularly in the clinical area of low back pain, a growing number of Ontario CHCs have been adding chiropractic care to their existing medical services. Some of these programs have been voluntary[16] and some have been funded as pilot projects by the Ontario Ministry of Health and Long-Term Care[14,17].

In line with such integration, the examination of costs and cost-effectiveness of chiropractic care has become one of the top research priorities for the chiropractic profession in Canada.[18] In 2017, a pilot project was conducted to evaluate a new integrated chiropractic back pain program at the Langs CHC in Cambridge, Ontario.[16,19] The main findings included patient-reported improvements in back pain intensity, disability and health-related quality of life, high levels of satisfaction, and reduced primary care provider visits and analgesic use with this service.[16] Although such findings suggest potential for health cost savings, a formal economic evaluation of this project was not undertaken. Others have found similar outcomes[14,15,17] yet few studies have examined the costs associated with chiropractic integration into primary healthcare settings, including within CHCs. Moreover, no studies have explored whether the implementation of these programs is cost-effective from the institutional point of view. As such, the purpose of the current study was to conduct a secondary analysis of the aforementioned program[16] using an economic evaluation design completed from the perspective of the Langs CHC[19]. Resource utilization and costs at the CHC from the health system perspective will be measured in a future study.

Research Question

In adult patients with back pain presenting within a CHC setting, is the addition of chiropractic care to usual medical care cost-effective from the perspective of the healthcare institution?

Methods

Study Design

This was a secondary cost-utility analysis[20] of a prospective, single-cohort observational study of a new back pain service provided by chiropractors integrated into a primary care CHC setting. The service was evaluated using patient-reported outcome measures, and data were collected prospectively on consecutive patients throughout the first two years of the program between January 2014 and January 2016.[16] For the current study, cost-utility was calculated as cost per quality-adjusted life year (QALY) gained. All costs are presented in 2019 Canadian dollars, and the perspective of the analysis was the Langs CHC (i.e. healthcare institution). The time horizon was 90 days (i.e. the length of time used for a care episode of low back pain in the previous study[16]). With a time horizon of less than one year, discounting of costs and effects was not performed.

Chiropractic Care

The chiropractic service provided at the CHC has been described previously.[16] Briefly, treatment sessions were evidence-based[21] and included high-velocity, low-amplitude spinal manipulation, soft-tissue therapy, education, reassurance, and home advice (e.g. icing, spinal stretching, and core strengthening exercises). Initial visits were typically 60 minutes in length and follow-up visits were 15 minutes. The service was provided on a rotating basis by four chiropractors (PCE, ALB, DFC, AFP), and was accessible on Tuesdays and Thursdays at the CHC from 12pm to 2pm, for a total of four hours per week.

Usual Medical Care

At the time of referral for chiropractic treatment all patients had already been under the management of their primary care provider for a back pain-related complaint. This was termed as 'usual medical care' and defined as any and all medical care received by patients with back pain at the Langs CHC, including but not limited to: primary care provider consultation visits (approximately 30 minutes in length), prescription medications (i.e. muscle relaxants, anti-inflammatories, analgesics), referrals for specialty care (e.g. spine surgical consultation), and referrals for diagnostic testing (e.g. lab work, imaging).

Patients

To be eligible, participants were: rostered patients at the Langs CHC, 18 years of age or older, seen and referred by their primary care provider (i.e. medical doctor or nurse practitioner) for a back pain-related complaint, unable to privately pay for chiropractic care, and suitable for manual therapy (i.e. absence of "red flags" [21]). Patients were screened by their primary care provider for eligibility and referred for chiropractic treatment. Referred patients included both naïve chiropractic patients and those with prior experience with chiropractic care. Patients completed an initial questionnaire before their first appointment, and then again at or before 90-day follow-up.

Baseline and Follow-up Instruments

Outcome measures included the Bournemouth (BQ)[22] and Bothersomeness[23] questionnaires, the Patient Global Impression of Change (PGIC),[24] and the EuroQol 5 Domain (EQ-5D).[25] The EQ-5D (3L version) is a widely used generic measure of health status commonly used in economic evaluations such as cost-utility analyses.[26] It assesses patient health in five dimensions (mobility, self-care, usual activities, pain/discomfort, and anxiety/depression). Each of these dimensions has three levels of response: no problems, some problems, and extreme problems. The EQ-5D also includes a visual analogue scale (VAS), on which the patient rates their perceived health from 0 (worst health imaginable) to 100 (best health imaginable). Responses from the five EQ-5D domains were used in the current study to generate a descriptive profile and a single index value of health status (i.e. utility) for each patient.[26] The pre-treatment and follow-up questionnaires also inquired about patient satisfaction, work status, details of medication usage, and other health care utilization. All instruments were administered at baseline and at (or before) 90-day follow-up.

Data Analysis

Descriptive statistics were used to summarize the demographic and clinical characteristic data from the pre-treatment questionnaires. For follow-up data, comparisons were made between responders and non-responders using the chi-squared test (or Fisher's exact test when appropriate) and independent t-test for categorical and continuous variables, respectively. The distributions for cost and effect data were analyzed for normality by examining data histograms, probability plots, and quantile-quantile plots, and then confirmed with the Kolmogorov-Smirnov test. For normal distributions, average values were calculated as the mean with 95% confidence intervals (CIs). For non-normal distributions, non-parametric bootstrapping (using 10,000 samples taken with replacement) was used to estimate 95% CIs. Because cost data are typically skewed[20], average values were also presented as the mean (standard deviation) and median (inter-quartile range) for comparison. Statistical significance was set at $p < 0.05$, and SAS® (SAS Institute Inc., Cary, North Carolina), Version 9.4, was used for analysis.

Analyses of the BQ, Bothersomeness, and PGIC outcome scores for the study have been described elsewhere.[16] For the EQ-5D, each patient's index (or utility) score was calculated using the EuroQol societal preference weights for Canada.[27] In general these scores range from less than 0 (where 0 is a health state equivalent to death, and negative values are valued as worse than death) to 1 (perfect health), with higher scores indicating higher health utility.[26] These were then used to calculate QALYs, as the change in EQ-5D index scores from baseline to follow-up multiplied by the estimated length of effect.[28,29] These values were then adjusted to a 90-day time horizon (by dividing each by four) for the current study. In addition, individual EQ-5D domain change scores were calculated and presented as recommended by EuroQol.[25,26]

A Paretian method[26,30] was also used to compare patients' EQ-5D health states pre- and post-chiropractic treatment. With this method, patients were categorized into one of four categories: worse (i.e. worse in at least one dimension and no better in any other), same (i.e. no change in pre-post health state profiles), mixed (i.e. better on one dimension, but worse on another), and improved (i.e. better on at least one dimension and no worse in any other).

For the cost analysis, cost estimates were obtained directly from the Langs institution. CHCs including Langs are unique in that they, unlike most Canadian primary care centres, which use fee-for-service reimbursement from provincial health plans for physician services, employ health professionals on a salaried basis.[13] For the purposes of this study, despite chiropractic services only being partially subsidized at the Langs CHC[16] we examined cost-utility of the program in a fully funded scenario. In addition, resource use for the program was determined by combining hourly wages and overhead expenses with the total number of hours of clinic operation over a 90-day period. For example, if the chiropractic low back pain program operated four hours per week this would result in a total of 52 hours over a 90-day period. Cost per patient was then calculated as the number of treatment visits (converted into total hours of use) multiplied by the unit costs of the program. For example, if a patient came to the clinic for a total of nine chiropractic treatment visits (one 60-minute initial visit and eight 15-minute follow-up visits) this would equal a total of three hours of chiropractic service use. If the total hourly cost of the chiropractic low back pain program (including hourly chiropractor salary, shared reception, and overhead expenses) was $95.04 then

Table 1.
Unit costing (hourly rate) of resource items for the low back pain program at the healthcare institution.

Resource Item	Unit Cost ($/hr)
Medical services [a]	
General physician	120.20
Nurse practitioner	52.63
Chiropractic services [b]	81.00
Reception services [c]	20.96
Examination room (rent) [d]	
Common charges (e.g. utilities, taxes, insurance, maintenance)	2.54
Shared business services expense (e.g. phone, internet, cleaning)	1.02

[a] These values are the current hourly wages paid to the respective medical providers at the healthcare institution.
[b] This value is based on the current hourly rate paid by the Ontario Ministry of Health and Long-Term Care for chiropractic services in the Waterloo Region.
[c] This value is the average hourly wage paid to reception staff at the healthcare institution.
[d] These values are based on current rental figures for a 140 sq ft examination room at the healthcare institution (operating 54 hours per week,[19] 52 weeks per year).

the total program cost for that patient would be $285.12. Resource use items and unit costs from the perspective of the healthcare institution are provided in Table 1.

Cost-utility of the chiropractic program was calculated as the average cost per QALY gained per patient during the 90-day period and plotted graphically on a cost-effectiveness plane (Figure 1). We performed a one-way sensitivity analysis[20] comparing mean and median cost per QALY gained values to investigate the impact of any differences between these values on study results. We also tested for potential cost savings of the program in a scenario[20] sensitivity analysis (see below).

Ethical Considerations

This study was approved by the AECC University College Policy and Procedures on Research Ethics.[16] All patients were also informed in writing at their first visit that the information given in the questionnaires would be used anonymously. Informed consent to chiropractic treatment was performed by the attending clinician.

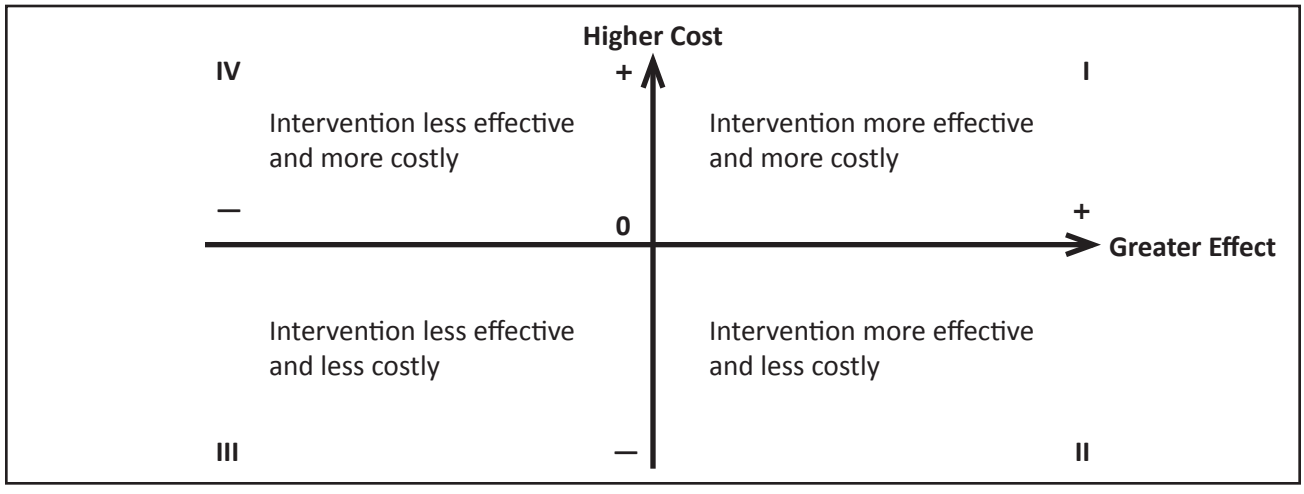

Figure 1.
The cost-effectiveness plane. The horizontal axis represents the difference in effect between the intervention of interest (e.g. chiropractic care) and the relevant alternative (e.g. the status quo or a competing program).[20] The vertical axis represents the difference in cost. If the cost-utility ratio of the intervention of interest lies in the south-east quadrant (II) it is both more effective and less costly than the alternative (i.e. it dominates the alternative), making it an obvious choice for program adoption. The opposite is true in quadrant IV. The most common scenario is when the intervention of interest lies within the north-east quadrant (I) indicating that the program is more effective but also more costly. In quadrant III the program is less costly but also less effective. In the latter two cases a value judgement is used (within the context of scarce resources and opportunity cost[20]) in deciding whether or not to adopt the new program.

Results

Patient Characteristics

In total, 95 consecutive patients reporting a back pain-related complaint were included in the service evaluation. The mean age of the cohort was 49 (±16) years and two-thirds (66%) were unemployed. Of those in paid employment, most (88%) were on sick leave because of their back pain. Eighty percent of patients had sub-acute (24%) or chronic (56%) back pain, with most describing it as constant (68%) and disabling (90%). Only a minority (19%) of patients had had a pain-free month in the past six months, and nearly four out of every 10 (37%) rated themselves in poor general health. Despite being a mostly chronic back pain population, the large majority (84%) expected to improve with treatment. In general, patients were also high users of health care services with two-thirds (65%) taking daily prescription analgesics and a similar proportion (67%) utilizing other health care providers in addition to their primary care physician for pain management. Baseline demographic and clinical characteristics of the study sample are summarized in Table 2.

Table 2.
Baseline demographic and clinical characteristics of patients (n = 95).

Variable	Value[a]	Missing Data
Age, mean (SD)	48.7 (16.1)	1
Sex (female)	52 (54.7)	0
Smoker	38 (40.4)	1
In paid employment	32 (34)	1
If employed, time off work for current back pain episode: 1-2 days 3-7 days 1-3 weeks > 3 weeks	 5 (20)[b] 7 (28)[b] 1 (4)[b] 9 (36)[b]	7
Same or similar condition in the past	73 (78.5)	2
Pain-free for a whole month in the past six months	18 (19)	0
Duration of current back pain episode: < 1 week 1-4 weeks 1-3 months > 3 months	 5 (5.4) 14 (15.1) 22 (23.7) 52 (55.9)	2
Constant pain	65 (68.4)	0
Limited usual activities > one day	83 (90.2)	3
Medication on a daily basis for back pain	60 (65.2)	3
Other practitioner use	62 (66.7)	2
Expectation of recovery/improvement	75 (84.3)	6
Physically active in comparison to others	47 (51.1)	3
In good general health	59 (62.8)	1
EQ-5D VAS, mean (SD)	54.1 (57.9)	14
EQ-5D Index [c], mean (SD)	0.50 (0.24)	3

SD = standard deviation, *VAS* = visual analogue scale.
[a] Values are expressed as the number (%) unless otherwise noted.
[b] Percentage values are calculated from the total number employed in the sample with complete data (i.e. 25).
[c] Single imputation methods (i.e. mean values) were used for missing EQ-5D data.

Follow-Up

There were a substantial number of patients who after completing the baseline assessment dropped out or were lost to follow-up. Out of 95 patients, 45 completed both the initial and follow-up assessments (47.4% response rate). Baseline data between responders and non-responders were analyzed for bias but no differences in age, sex, and severity of their condition (as measured by baseline total BQ scores) were found between the two groups.[16] Baseline EQ-5D scores were nearly all identical as well, with the exception that fewer responders reported extreme problems with anxiety and depression compared to non-responders (Appendix 1). No information was available regarding loss to follow-up of patients under usual medical care.

Patient-Reported Outcome Measures

Health outcomes for the BQ, Bothersomeness, PGIC, and patient satisfaction measures have been reported elsewhere.[16] EQ-5D scores as categorized using the Paretian method indicated that two-thirds (67%) of patients at follow-up improved and fewer than one in 10 (8.9%)

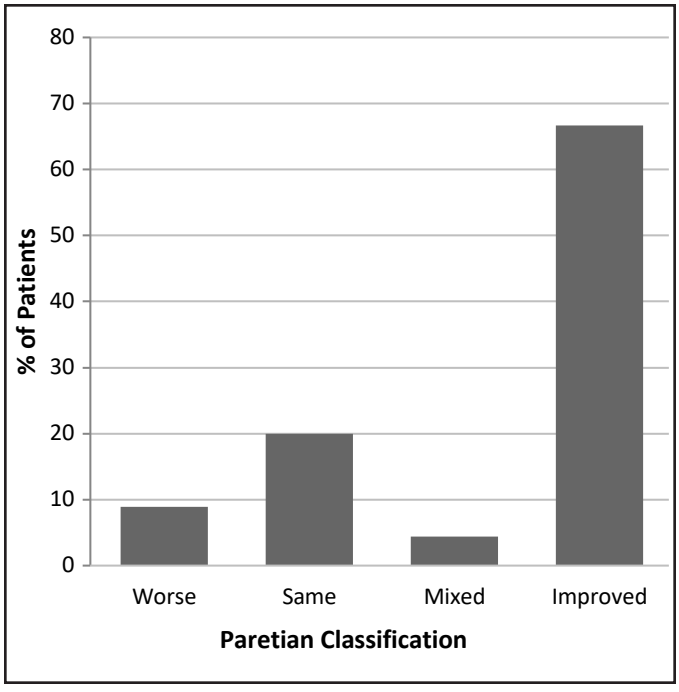

Figure 2.
Paretian classification of EQ-5D patient profiles (post-chiropractic treatment).

had worsened (Figure 2). The remainder stayed about the same (20%) or had mixed improvement (4.4%). These results were similar to those obtained in the study sample using the BQ, Bothersomeness, and PGIC measures reported previously.[16] An analysis of the five EQ-5D domains revealed that the highest reported changes for improvement (in decreasing order) were in self-care, anxiety/depression, usual activities, and mobility, whereas the lowest changes in improvement reported by patients were in pain/discomfort (Table 3).

Cost Analysis

The mean time between completing the pre-treatment and follow-up questionnaires over the 90-day period was 10 ± 8.3 weeks (range, 1-47 weeks). The mean number of chiropractic treatment sessions that patients received during this time was 8.4 ± 3.8 (range, 2-16 visits). Among this group, nearly half (46%) of those who were on sick leave for their back-pain complaint returned to work post-chiropractic treatment. With regards to other health care utilization, more than three-quarters (77%) had not sought help from any other practitioner for their back pain during the follow-up period, and more than eight out of

Table 3.
Numbers and proportions of patients reporting levels within each of the EQ-5D dimensions: pre- and post-chiropractic care for back pain.

	Mobility		Self-Care		Usual Activities		Pain / Discomfort		Anxiety / Depression	
	Pre	**Post**	**Pre**	**Post**	**Pre**	**Post**	**Pre**	**Post**	**Pre**	**Post**
Level 1	10 (22.2%)	23 (51.1%)	30 (66.7%)	37 (82.2%)	5 (11.1%)	21 (46.7%)	2 (4.4%)	11 (24.4%)	15 (33.3%)	28 (62.2%)
Level 2	33 (73.3%)	22 (48.9%)	14 (31.1%)	8 (17.8%)	36 (80%)	23 (51.1%)	25 (55.6%)	32 (71.1%)	27 (60.0%)	16 (35.6%)
Level 3	2 (4.4%)	0 (0%)	1 (2.2%)	0 (0%)	4 (8.9%)	1 (2.2%)	18 (40%)	2 (4.4%)	3 (6.7%)	1 (2.2%)
Total [a]	45 (100%)	45 (100%)	45 (100%)	45 (100%)	45 (100%)	45 (100%)	45 (100%)	45 (100%)	45 (100%)	45 (100%)
Reporting some problems [b]	35 (77.8%)	22 (48.9%)	15 (33.3%)	8 (17.8%)	40 (88.9%)	24 (53.3%)	43 (95.6%)	34 (75.6%)	30 (66.7%)	17 (37.8%)
Change in numbers reporting problems	–13		–7		–16		–9		–13	
% change reporting problems	–37%		–47%		–40%		–21%		–43%	
Rank of dimensions in terms of % changes	4		1		3		5		2	

[a] Results are for those who responded to both the pre- and post-treatment questionnaires. Forty-seven percent of respondents to the pre-treatment EQ-5D also responded to the post-treatment EQ-5D.
[b] Some problems = levels 2 + 3.

Table 4.
Summary of healthcare institution costs ($) of chiropractic care per back care episode[a].

Cost	$, Mean (SD)	$, Median (IQR)	% Total Costs
Direct costs [b]			
Chiropractic consultations	232.65 (77.19)	243 (101.25)	85.2
Reception services [c]	30.10 (9.99)	31.44 (13.1)	11.0
Overhead cost [b]			
Examination room	10.23 (3.39)	10.68 (4.45)	3.8
Total costs [b,d]	272.98 (90.56)	285.12 (118.8)	100.0

IQR = inter-quartile range, *SD* = standard deviation.
[a] Equivalent to 90 days.
[b] Kolmogorov-Smirnov test was not significant ($p > 0.15$), confirming the approximation to a normal distribution.
[c] This service was valued as a shared cost between the medical and chiropractic services (i.e. half of the $20.96 hourly wage).
[d] Results are for patients who responded to both the pre- and post-treatment questionnaires (n = 45).

10 (82%) were either not taking any medication or had managed to significantly reduce their medication usage for their pain.

An analysis of the cost of care during the 90-day follow-up period is shown in Table 4. The mean total cost of direct care was approximately $263 (95% CI, $237 to $289) per patient, while the overhead cost for use of an examination room was only around $10 (95% CI, $9 to $11) per patient, leading to a total mean cost of care of about $273 (95% CI, $246 to $300). Chiropractic consultation visits, fully subsidized by the Langs institution at a rate of $81 per hour, would have contributed to 85% of total costs. Other costs including shared reception services and overhead expenses would constitute around 11% and 4%, respectively.

The costs to the healthcare institution per QALY gained with chiropractic care are summarized in Table 5. Over the 90-day period an average of 0.21 (95% CI, 0.14 to 0.29) QALYs were gained at an average cost per QALY (including overhead expenses) of $1,042 (95% CI, $768 to $1,340). This represents a trade-off between higher costs and improved outcomes for patients treated with

Table 5.
Average healthcare institution costs per QALY gained with chiropractic care (per back care episode [a]).

	EQ-5D Scores				Cost of Care ($)[b]		
	Pre	Post	Change	QALY Gained[b]	Direct	Overhead	Cost/QALY ($)[c]
Mean (SD)[d]	0.55 (0.24)	0.76 (0.16)	0.21 (0.25)	0.21 (0.25)	263 (87)	10.2 (3.4)	1,042 (947)
Median (IQR)[d]	0.59 (0.32)	0.77 (0.14)	0.16 (0.36)	0.16 (0.36)	274 (114)	10.7 (4.5)	905 (733)

IQR = inter-quartile range, *QALY* = quality-adjusted life years, *SD* = standard deviation.
[a] Equivalent to 90 days.
[b] Kolmogorov-Smirnov test was not significant ($p > 0.15$), confirming the approximation to a normal distribution.
[c] Kolmogorov-Smirnov test was significant ($p < 0.01$) with quantile-quantile plot indicating long tails at both ends of the data distribution.
[d] Results are for patients who responded to both the pre- and post-treatment questionnaires (n = 45).

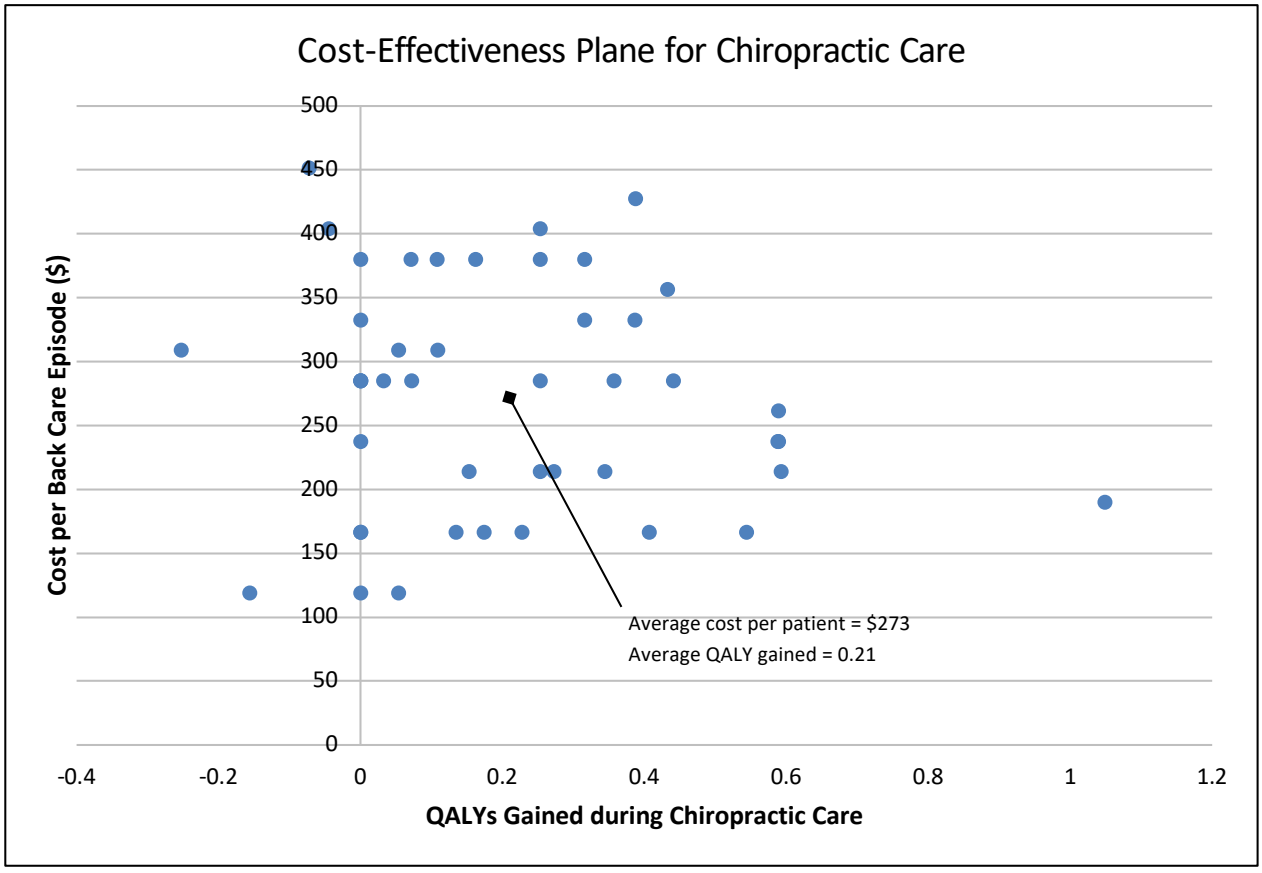

Figure 3.
Quality-adjusted life years gained and associated cost for individual patients during chiropractic care (pre- to post-treatment). QALY = quality-adjusted life years.

chiropractic care (compared with no chiropractic care).[20] This trade-off is illustrated in Figure 3 as the distribution of cost versus QALYs gained during chiropractic care. The average cost per QALY gained would fall within the north-east quadrant of the cost-effectiveness plane. When median cost per QALY gained was used, the value reduced (i.e. $905 vs. $1,042) and remained in a trade-off position yet would fall at a more south-easterly position (not shown in Figure 3).

Cost Savings

Studies from the literature have reported the mean number of annual visits to a primary care physician for the management of chronic back pain to be between 5.4 and 16.4 per patient.[31,32] Assuming similar rates of utilization, if 77% of patients in the current study did not visit their primary care provider during the 90-day period while under chiropractic care this would result in cost savings for the institution. For instance, if primary care visits at the Langs CHC were 30 minutes in length and if an average wage of $86.42 for medical services was used (see Table 1), the reduced total number of hours of primary care visits in this cohort of patients over a 90-day period would have ranged between 23.4 and 71 hours, representing a potential cost savings from this program of between $2,022.23 and $6,135.82. (This was assuming a linear relationship between cost reduction and time in extrapolating these figures from 10 weeks [i.e. the average number of weeks patients were under this service] to three months.) Taking these cost savings into account, in a worst-case scenar-

io the average cost per QALY gained during chiropractic care would have remained in a trade-off position between higher costs and improved outcomes, but at a lower cost per QALY gained (i.e. $517 [95% CI, $466 to $568] vs. $1,042 [95% CI, $768 to $1,340]). However, in a best-case scenario there would have been an average cost savings of $212 (95% CI, $193 to $231) per QALY gained per patient, moving the cost-utility ratio into the south-east quadrant of the cost-effectiveness plane (Appendix 2). Median cost per QALY gained values were slightly lower in each scenario (i.e. $431 and –$177, respectively) but did not change the qualitative conclusions of the results.

Discussion

This study aimed to evaluate the potential cost-utility of chiropractic services integrated within a Canadian primary care CHC setting from the perspective of the healthcare institution. Nearly 70% of patients who were referred into this service pathway and followed through to discharge reported improvement with chiropractic care. This finding is consistent with those of other studies involving the integration of chiropractic services in the management of patients with low back pain and other musculoskeletal disorders.[14-17,33] On average, patients also reported a gain of 0.21 QALYs over a 90-day period. In other words, each patient gained an average of 0.21 years (i.e. greater than two and a half months) in full health during this time as a result of receiving chiropractic treatment. This is an important finding as the majority of patients in this study reported persistent back pain symptoms at baseline despite being under usual medical care.

Without taking potential cost savings of the current program into consideration, the average cost per QALY gained per patient over the 90-day treatment period was $1,042. When compared to a threshold of $20,000 per QALY recommended in Canada as a cut-off for accepting health care programs as being cost-effective[34], this program appears to represent very good value-for-money. In essence, needed resources that would be displaced from other treatments or programs at the healthcare institution in order to fund the chiropractic program (i.e. the opportunity cost[20]) would be comparatively low. Although this threshold is arbitrary and has been challenged in the literature[35] many healthcare programs including pharmaceutical and surgical interventions with equivocal long-term outcomes to chiropractic treatment[36-38] have been universally adopted in healthcare systems at much higher cost-utility ratios than the current program[39].

The findings of the current study suggest there may also be potential for cost savings to the healthcare institution when chiropractic care is added to usual medical care in the management of complex patient populations such as those who present within CHCs. For instance, over three-quarters of patients in this study did not visit their primary care provider while under chiropractic care. Similar reductions in physician visits in association with chiropractic integration have also been reported elsewhere.[15,40] When taken in to account this translates into an estimated average cost reduction (in terms of hourly services in primary care provider visits) of between greater than $2,000 and $6,000 over the course of three months. (Exact values calculated were $2,022.23 and $6,135.82, respectively.) In a fully funded scenario, if the chiropractic program were to cost the healthcare institution $4,942.08 over three months, the opportunity cost (i.e. resources that would need to be displaced from other programs to fund the chiropractic program) over this time period would range from a high of around $3,000 to a low of nearly –$1,200. In other words, in the first case, and in the context of scarce resources, a choice would need to be made by the healthcare institution in order to decide if the value of what is being added is greater than the value of what is being given up.[20] For instance, would it be worth taking resources from other institution programs (e.g. youth and teen, diabetes education, midwifery[19]) to fund low back pain services? Alternatively, the institution could generate the required resources through, for example, additional fundraising and/or government grants. In the second case, however, the potential cost savings alone would more than subsidize the chiropractic service. Although we did not consider the costs to the institution for patients who were lost to follow-up, we also did not consider the cost savings these patients may have created for the institution by not visiting their primary care provider while under chiropractic care. For instance, these patients had an average of 3.6 ± 2.8 (range, 1-11) visits at the institution at the point they were lost to follow-up. Although this group would have generated less in terms of potential cost savings to the institution, they also would have created less costs as they attended for fewer visits than the group of patients not lost to follow-up (i.e. aver-

age of 3.6 vs. 8.4 visits). Notwithstanding, the estimated reductions in primary care provider visits for this study were based on rates of physician utilization by chronic back pain patients observed in other research[31,32], as actual rates were not tracked in the current study. Until such measures are conducted, caution is warranted in applying these estimates to the current context.

In addition to reduced primary care provider visits, the majority of patients in this service pathway at follow-up reported reductions in the use of analgesic medications. Reductions in pain medication usage with access to chiropractic services have also been reported in other studies.[5,6,8,17,33,40] In light of the current opioid crisis, further investigation regarding the impact of chiropractic integration on opioid use among chronic back pain patients in the healthcare system is warranted.

In this study almost half of respondents who were employed but on sick leave at baseline returned to work after the 90-day treatment period. As unemployment rates and disability are characteristically high among CHC patient populations[13-16], this is also a potentially important finding. The return-to-work of even a small number of such cases has indirect cost savings implications from the societal perspective. From the health system perspective there may have been other reductions in health service utilization, such as emergency department visits, orders for advanced diagnostic imaging, referrals for injections, and/or referrals for spinal surgery, that were not measured in the current study. Spine-related pain is often disabling and recurrent[41], particularly in complex patients and those of low socioeconomic status[9,11,13-16,41]. Such patients also tend to be high users of healthcare services.[11,13] By adding chiropractic care to usual medical care within the CHC setting, the findings of the current study and others[14-16,33,40] suggest there may be potential for cost savings outside of those obtainable through the healthcare institution. As such, the effect of chiropractic integration on these and other direct and indirect health system and societal costs and consequences should be measured in future investigations. Future studies should include a comparator group so as to properly evaluate these programs within the economic context.[20]

Limitations

This study had several important limitations. First, this was a single-cohort study conducted on a relatively small patient sample. A complete economic evaluation would have included a comparison between at least two health care programs or alternatives.[20] Although the current study design allowed for the evaluation of the addition of a new program to the existing back care services provided at the CHC (i.e. chiropractic care plus usual medical care) data were not collected on a control group of patients not receiving chiropractic treatment. As such, incremental cost-utility (i.e. the added cost per additional QALY gained with the new program compared to the existing one) could not be calculated. At best, cost-utility could only be examined from a 'with' versus 'without' chiropractic care scenario as part of a partial economic evaluation.[20]

Second, there was a high loss to follow-up. In all pragmatic studies, which, by their nature, are not conducted in a highly controlled research environment, there is likely to be a relatively high number of dropouts, as was the case here. However, we are not aware of any systematic differences between those patients who did and did not complete follow-up questionnaires as baseline characteristics were similar between the two groups. Nevertheless, caution is warranted in extrapolating the findings to other CHC populations.

Third, QALYs were measured for this study using a time trade-off approach.[26] In brief, with this method 243 potential individual health states from the EQ-5D (3L) were transformed into single indexes. The difference between the pre- and post-treatment indexes were then multiplied by the estimated length of effect (and then adjusted to a 90-day time horizon) to obtain the QALYs gained for each patient from chiropractic treatment.[28] Although a complete set of these indexes has been obtained from a representative sample of the Canadian general population,[27] and this value set was used in calculating QALYs for the current study, these measures were based on societal preferences and not that of the individual patient. For this and other reasons (such as the restrictive assumptions that underlie utility theory[20]) QALYs have been criticized in the literature[42] and measures such as healthy-years equivalents (HYEs) and willingness-to-pay methods have been suggested as superior alternatives.[42,43] Although this value set was obtained from a Canadian population[27] there may have also been systematic differences in terms of socio-demographics between this group and the current study sample. For instance, when con-

trasted with the current study cohort participants in the aforementioned sample[27] were, on average, older, more well-educated, and of a higher socioeconomic class (i.e. higher income levels). As such, these preference weights may not have been completely generalizable to the current study group.

Fourth, we used methods similar to those proposed by Newell *et al*.[44] to estimate the length of effect for the benefits of chiropractic care in the current study. However, the cited rate of reoccurrence[44] was based on a study involving patients with acute back pain symptoms.[29] Moreover, there are limitations to assuming that the benefit from chiropractic care will be the same for every patient. The majority of CHC patients in the current study presented with chronic back pain symptoms, which may have resulted in an over-estimation of the length of effect of chiropractic care for these patients. Furthermore, it was assumed that individual patients' baseline QALYs would not have changed *without* chiropractic treatment. The follow-up period was also less than ideal as a follow-up of one to two years is more commonly recommended for spine pain patients.[45] Although the majority of patients in the current study at baseline presented with chronic and persistent back pain, those with acute pain may have been more likely to recover regardless of treatment,[29,44] thereby also contributing to an inflated benefit of the chiropractic service.

Finally, for this study we estimated primary care provider visit utilization from the literature as these rates were not measured in the original study.[16] Although these utilization rates were not directly measured for the current study they may still have been an accurate proxy as they were obtained from chronic back pain patient populations who were similar in many respects to the current study sample. Nevertheless, other health care utilization measured previously[16] was also conducted using patient-reported questionnaires. The non-utilization of primary care services reported by 77% of patients from that study was not validated (e.g. with data from electronic medical charts) and therefore could have been subject to measurement bias.

Conclusion

This study evaluated the cost-utility of chiropractic integration for low back pain services within a primary care CHC setting from the perspective of the healthcare institution. Among the subjects followed in this study, the addition of chiropractic care to usual medical care was associated with improved outcomes at a reasonable cost. These outcomes, along with the potential cost savings of such integration, may have important implications for healthcare institutions and their patients, as well as for policy decision-makers and other health stakeholders. Future comparative cost and effectiveness studies with control of confounding are nevertheless needed to evaluate the impact of chiropractic care with or without usual medical care in these settings.

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Appendix 1.

Comparison of reported levels within baseline EQ-5D dimensions of responders (n = 45) and non-responders (n = 50).

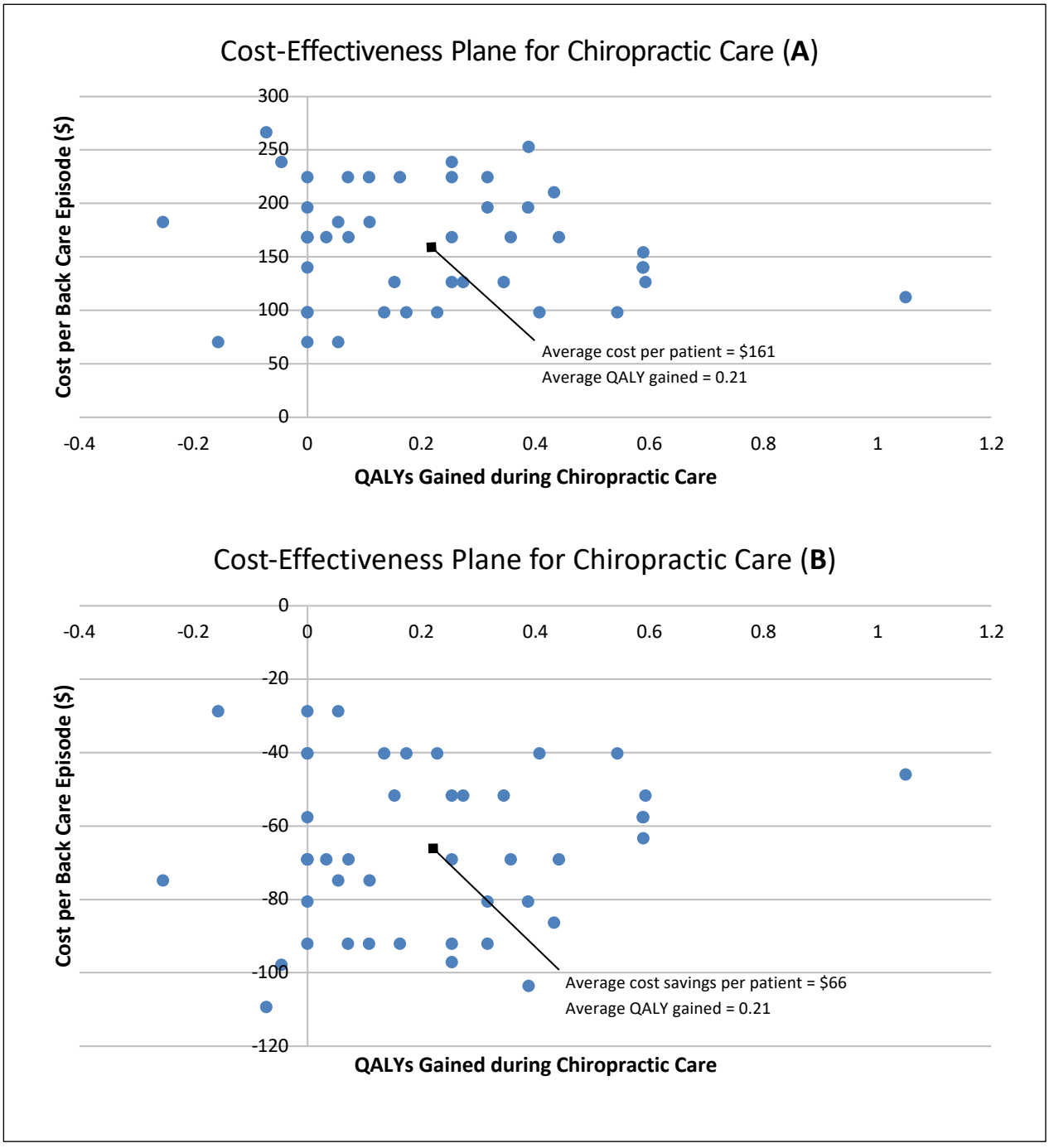
	Mobility			Self-Care			Usual Activities			Pain / Discomfort			Anxiety / Depression		
	R	NR	p [a]	R	NR	p [a]	R	NR	p [a]	R	NR	p [a]	R	NR	p [a]
Level 1	10 (22.2)	14 (28)	0.52	30 (66.7)	30 (60)	0.50	5 (11.1)	6 (12)	0.89	2 (4.4)	0 (0)	0.22[b]	15 (33.3)	10 (20)	0.14
Level 2	33 (73.3)	34 (68)	0.57	14 (31.1)	19 (38)	0.48	36 (80)	35 (70)	0.26	25 (55.6)	23 (46)	0.35	27 (60)	28 (56)	0.69
Level 3	2 (4.4)	2 (4)	0.38[b]	1 (2.2)	1 (2)	0.50[b]	4 (8.9)	9 (18)	0.20	18 (40)	27 (54)	0.17	3 (6.7)	12 (24)	*0.02
Total	45 (100)	50 (100)		45 (100)	50 (100)		45 (100)	50 (100)		45 (100)	50 (100)		45 (100)	50 (100)	
Reporting some problems [c]	35 (77.8)	36 (72)	0.52	15 (33.3)	20 (40)	0.50	40 (88.9)	44 (88)	0.89	43 (95.6)	50 (100)	0.22[b]	30 (66.7)	40 (80)	0.14

NR = non-responder, *p* = p-value, *R* = responder.
Values for responders and non-responders are expressed as the number (%).
[a] All p-values were calculated using the chi-squared test unless otherwise noted.
[b] Calculated using Fisher's exact test.
[c] Some problems = levels 2 + 3.

* Statistically significant.

Appendix 2.

Quality-adjusted life years gained and associated potential cost savings to the healthcare institution per individual patient with chiropractic care. (A) Worst case scenario. (B) Best case scenario. QALY = quality-adjusted life year



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Lifetime prevalence of concussion among Canadian ice hockey players aged 10 to 25 years old, 2014 to 2017

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Background: *The primary objective of this study was to identify the self-reported lifetime prevalence of diagnosed concussions among Canadian ice hockey players aged 10 to 25 years old.*

Method: *Medical records were identified for n=5223 athletes whom completed comprehensive baseline assessments with a Canada-wide network of private concussion management clinics. Variables extracted included: sex, age, diagnosed history of and number of prior concussions, diagnosed health condition(s), and Post-Concussion Symptom Scale scores.*

Results: *Approximately 22% of all athletes, 21.7% of females and 21.8% of males reported that they had sustained at least one diagnosed concussion. Age was significantly associated with history of concussion as was having an additional health condition. Sex was not significantly associated with a history of concussion.*

Conclusion: *Lifetime history of concussion prevalence*

Contexte : *L'objectif principal de cette étude était de déterminer la prévalence autodéclarée au cours de la vie des commotions cérébrales diagnostiquées chez les joueurs canadiens de hockey sur glace âgés de 10 à 25 ans.*

Méthode : *Des dossiers médicaux ont été identifiés pour 5223 athlètes qui ont effectué des évaluations de base complètes avec un réseau pancanadien de cliniques privées de gestion des commotions cérébrales. Les variables extraites comprenaient : le sexe, l'âge, les antécédents diagnostiqués et le nombre de commotions cérébrales antérieures, le ou les problèmes de santé diagnostiqués et les scores de l'échelle des symptômes après la commotion cérébrale.*

Résultats : *Environ 22 % de tous les athlètes, 21,7 % des femmes et 21,8 % des hommes ont déclaré avoir reçu au moins un diagnostic de commotion cérébrale. L'âge était associé de façon significative aux antécédents de commotion cérébrale tout comme le fait d'avoir un autre problème de santé. Le sexe n'était pas associé de façon significative à des antécédents de commotion cérébrale.*

Conclusion : *Les estimations de la prévalence des*

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estimates aligned closely with estimates previously published. Future investigations should seek to establish the prevalence of concussions that occur during ice hockey games and practices alone.

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KEY WORDS: concussion history, ice hockey, prevalence, youth, young adult

antécédents de commotion cérébrale au cours de la vie concordent étroitement avec les estimations publiées antérieurement. Les enquêtes futures devraient chercher à établir la prévalence des commotions cérébrales qui surviennent uniquement pendant les parties et les pratiques de hockey sur glace.

(JACC. 2019;63(2):80-91)

MOTS CLÉS : antécédents de commotion cérébrale, hockey sur glace, prévalence, jeunes, jeunes adultes.

Introduction

Over the last 10 to 15 years, annual incidence rates of sport-related concussion (SRC) among youth athletes has increased dramatically. A recent article published by Zemek *et al*. indicated that the number of concussions treated in emergency departments and physician offices across the province of Ontario demonstrated a 5.5-fold increase (per 100 000 children) from 2003 to 2013.[1] Zemek attributed this steep increase to improvements in stakeholder concussion education and awareness, improved diagnostics, and stricter return to play medical clearance requirements. However, generating annual incidence and prevalence estimates of concussion based on emergency department visits may provide a gross underestimate of the actual number of concussions, as many athletes do not present to a hospital for diagnosis or follow-up treatment.[2] To that extent, a large body of evidence suggests that head injuries often go unrecognized and underreported by adolescent aged athletes.[3-7] Estimates suggest that approximately half of all suspected injuries go unreported in this population.[8] Further investigation is required to provide more accurate estimates of lifetime prevalence of concussion among young athletes.

Incidence of SRC by sport is well documented among youth athlete populations.[9] Findings indicated that athletes in certain sports face a greater risk for SRC than others, especially those athletes involved in high speed contact sports such as rugby, ice hockey, football and lacrosse.[9, 10] Ice hockey is not only Canada's national winter sport, but also one of our most popular recreational activities for children and youth behind that of soccer and swimming.[11] In the 2017-18 Annual Report released by Hockey Canada, over 340 000 individuals between the ages of 10 and 25 were enrolled in a hockey program at any given level.[12] As such, the popularity of ice hockey in Canada places a large number of athletes at risk for SRC. Examining athlete-reported histories of diagnosed concussions at the time of preseason baseline assessment may be useful in gauging cumulative injury prevalence. Despite being retrospective in nature, asking an athlete to recall their concussion history at a time when their current playing status is not at stake (i.e., when the athlete is not at risk to be removed from play due to a suspected SRC), may provide a more accurate assessment of concussion prevalence in this population.

Examining factors related to concussion history, such as age, sex and health history, may provide an indication as to those athletes at greater risk for an additional concussion. At present, younger age has yet to be clearly established as a risk factor for concussion.[13] With respect to recovery however, evidence suggests that younger athletes take longer to recover following concussion compared to older athletes.[14-16] With respect to sex, evidence suggests that the events surrounding concussion (i.e., incidence or risk for concussion by sport, initial symptom severity, time until functional recovery, etc.) diverge between males and females.[17-19] Following concussion, females typically take longer to achieve functional recovery than do males and are also more likely to experience persisting symptoms.[20-23.] A pre-morbid history of learning disability, psychiatric illness (i.e., depression and/or anxiety), sleep disorder, and history of headache/migraine is also thought to increase an athlete's risk for persisting post-concussion symptoms.[24]

The primary objective of this study was to identify the lifetime prevalence of self-reported concussion diagnoses among Canadian ice hockey players aged 10 to 25 years old, whom presented to Complete Concussion Management Inc. (CCMI) clinics between 2014 and 2017, by age and sex. The secondary objective(s) were to: i) examine the relationship between history of concussions (including number of concussions), and an additional diagnosed health condition (i.e., learning exceptionality, history of psychiatric illness, sleep disorder, and history of headache) by age and sex, and ii) examine the relationship between history of concussion and Post-Concussion Symptom Scale (PCSS) severity score at baseline (i.e., in the absence of injury), by sex.

Methods

Study design, participants and data extraction

This was a cross-sectional study that utilized a convenience sample of youth and young adult ice hockey players aged 10 to 25 years old, involved in various levels of hockey (i.e., house league, rep, provincial, etc.). A retrospective review of electronic medical records were identified and extracted from athletes whom had participated in comprehensive preseason concussion baseline testing at any one of 107 CCMI clinics across Canada between October 2014 and October 2017. The CCMI comprehensive pre-season testing protocol consisted of a clinical interview regarding medical and concussion histories as well as the PCSS, Sport Concussion Assessment Tool 3 (SCAT3), oculomotor function testing, postural sway measuring centre of pressure area (COP), clinical reaction time, grip strength, and computerized neurocognitive testing (for athletes 13 or older). Each province had at least one clinic and clinic locations varied from urban to rural settings. Most clinics offered a myriad of clinical services beyond that of concussion management and comprehensive baseline assessments (i.e., sport medicine [MD], physiotherapy [PT], chiropractic [DC], etc.). All clinicians were licenced health care professionals (i.e., MD, PT, DC), and were trained by CCMI via the completion of a 36-hour continuing education training course for evidence-based concussion care. CCMI partnered clinics are also required to undergo a 15-hour recertification course every 24-months to retain certification. Any individual in need of assessment following a suspected concussion, or whom was interested in obtaining a comprehensive baseline assessment, is eligible to visit a CCMI clinic. CCMI network clinics do not exclusively treat athletes.

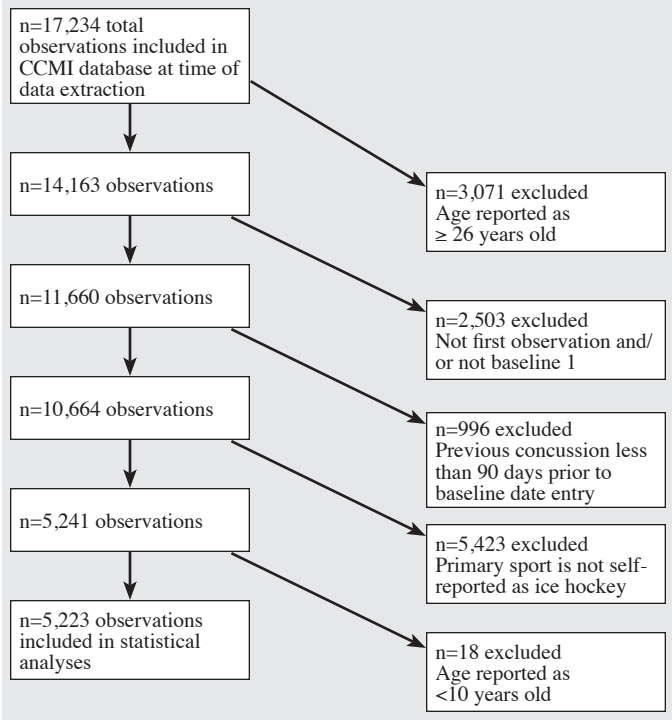

Figure 1.
CCMI database medical record exclusion flowchart.

At time of data extraction (October 2017), the CCMI database contained n=17 234 observations. First, an age filter was applied to include records from individuals aged ≤ 25.9 years old at time of record entry (n=3071 excluded; age reported as ≥ 26 years old). Duplicate records for an athlete were removed (n=2503 excluded); baseline assessments are conducted on an annual basis, and concussion assessments on an as needed basis. Only the first entry within an athlete's medical record was included in analysis. Athletes who had sustained a concussion and then participated in a concussion baseline assessment < 90 days from day of injury were also removed (n=996 excluded). This criterion was applied to ensure that PCSS severity scores had returned to baseline (i.e., pre-injury levels) following a diagnosed concussion. Athletes who did not identify ice hockey as their primary sport were also removed (n=5423 excluded). Finally, athletes < 10 years old were removed (n=18 excluded). Refer to Figure 1 for med-

ical record exclusions. All medical records identified for inclusion in this study were screened by a biostatistician (SHJ) with whom the authors consulted.

Medical records were generated and saved within the CCMI database for each athlete who participated in a baseline assessment or concussion assessment at the time of their appointment. Although clinicians collected health histories (i.e., presence of a diagnosed psychiatric illness or learning exceptionality, etc.) and concussion injury data across several domains (i.e., cognitive and oculomotor functioning, reaction time, postural sway, handgrip strength, etc.), only data pertaining to athlete age, sex, health and concussion history, and PCSS severity scores were extracted from medical records.

Health and concussion histories were self-reported by athletes during an in-person interview with a CCMI certified clinician. Each athlete was asked to respond to the following questions: i) "have you ever been *diagnosed* with a concussion?" ii) "if yes, how many?", iii) "when was the most recent?", iv) "how long did it take you to recover from the most recent concussion?", and v) "do you still experience symptoms from your most recent concussion?". Only responses pertaining to questions i) and ii) were used within analyses. Data pertaining to the following health history variables were also extracted: *diagnosed* learning exceptionality, ADD/ADHD, depression, anxiety, sleep disorder, and history of headache. Concussion-like symptom severity scores (as reported on the PCSS, a 22-item measure with a total possible score of 132) were extracted and stratified by number of concussions for comparison between sex groups. The PCSS has been found to have a high internal consistency in both healthy (Cronbach's α = 0.89-0.94) and concussed samples (α = 0.92-0.93)[25], and consists of a four factor structure: somatic, cognitive, emotional and sleep-related symptoms[26]. For the purposes of age comparison, athletes aged 10 to 12 years old were defined as "youth", 13 to 18 years old as "adolescent" and 19 to 25 as "young adult".

Pre-participation consent was provided by athletes and/or their guardians for the collection and use of de-identified data for research purposes. This investigation was approved by the Research Ethics Board at the Canadian Memorial Chiropractic College, Toronto, Ontario (protocol #162009).

Data analysis

De-identified athlete medical records were extracted from the CCMI database and exported into an encrypted excel spreadsheet. The spreadsheet was then moved into Statistical Package for the Social Sciences software (SPSS) in January 2018. Mean age (SD) was calculated for the total sample and each sex group. Chi square statistics were calculated for the lifetime prevalence of concussion among sex and age groups (see Table 1). To allow for comparison of the effect size, odds ratios were calculated for a history of concussion for both sex and age groups (see Table 2; note that history of concussion was dichotomized as "yes" or "no"). Chi square statistics were also calculated

Table 1.

Lifetime prevalence of self-reported diagnosed concussions among Canadian ice hockey players aged 10 to 25 years old presenting to CCMI clinics between 2014 and 2017, by sex and age. Note: percentages reflect the proportion of the row total.

	Number of Prior Concussions								χ^2	P-Value
	0		1		2		3+			
	N	%	N	%	N	%	N	%		
Total Sample	4085	78.2	743	14.2	269	5.2	126	2.4	—	—
Sex	**N**	**%**	**N**	**%**	**N**	**%**	**N**	**%**		
Male	3265	78.2	602	14.4	212	5.1	97	2.3	1.44	0.70
Female	820	78.3	141	13.5	57	5.4	29	2.8		
Age Category	**N**	**%**	**N**	**%**	**N**	**%**	**N**	**%**		
Youth (10 to 12 yrs.)	1602	83.4	238	12.4	67	3.5	13	0.7	206.8	<0.001
Adolescent (13 to 18 yrs.)	2352	76.5	466	15.2	173	5.6	82	2.7		
Young Adult (19 to 25 yrs.)	131	57.0	39	17.0	29	12.6	31	13.5		

Table 2.

Association between history of self-reported diagnosed concussion (vs. none) by age, sex and select medical histories among Canadian ice hockey players aged 10 to 25 years old presenting to CCMI clinics between 2014 and 2017. Legend: N= no, Y= yes, OR = odds ratio

		No History Concussion	History of Concussion	OR	95% CI	P-Value
Age Category						
Younger (10 to 18 yrs.)		3954	1039	—	—	—
Older (19 to 25 yrs.)		131	99	2.88	2.20-3.77	<0.001
Sex						
Male		3265	911	—	—	—
Female		820	227	0.99	0.84-1.17	0.93
Medical History						
Learning Exceptionality	N	3882	1047	—	—	—
	Y	203	91	1.66	1.29-2.15	<0.001
Prior Psychiatric Diagnosis	N	4003	1068	—	—	—
	Y	82	70	3.20	2.31-4.43	<0.001
Sleep Disorder	N	4066	1128	—	—	—
	Y	19	10	1.88	0.88-4.09	0.12
History of Headache	N	3406	873	—	—	—
	Y	679	265	1.52	1.30-1.79	<0.001

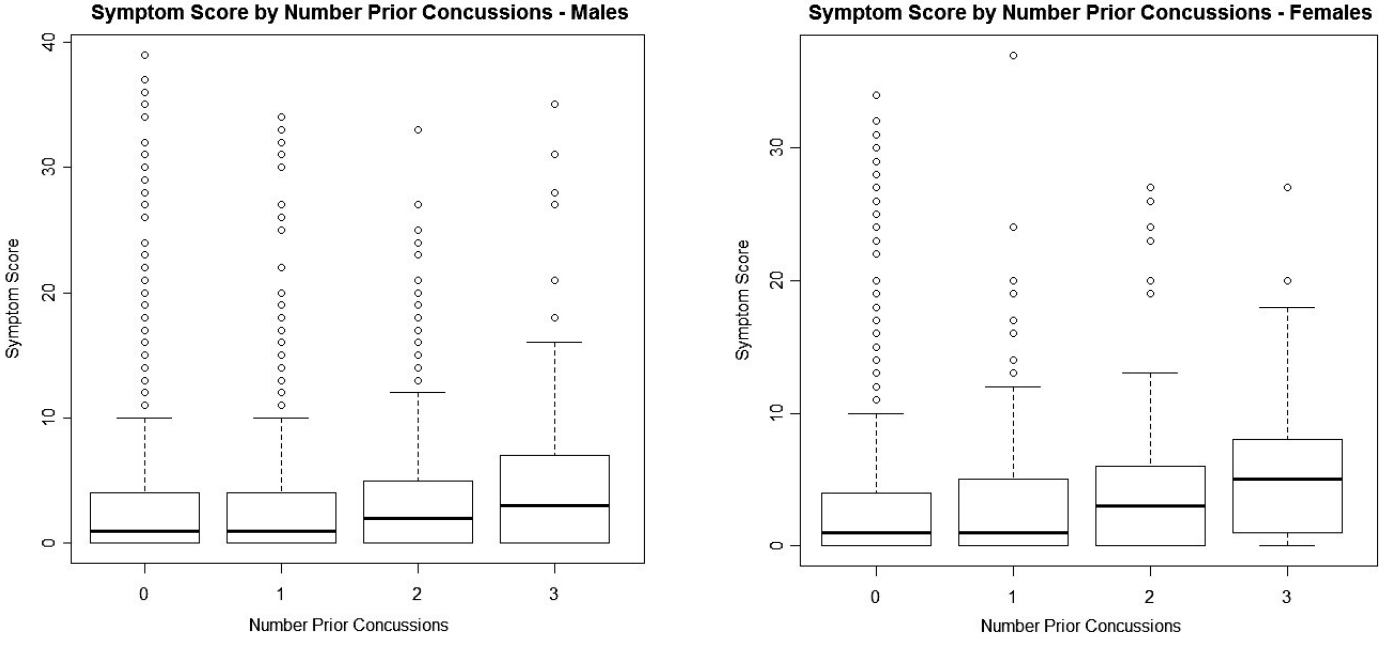

Figure 2.

Median PCSS score by prior number of concussions and sex.

within each sex group for prevalence of concussion(s) by age and health history (see Tables 3 and 4). Lastly, box and whisker plots were used to compare PCSS severity scores for males and females by number of previous concussions (see Figure 2). Statistical significance was set at p=0.05 for all tests.

Results

A total of n=5223 medical records from ice hockey players between the ages of 10 and 25 years old were included in this study. This sample represented approximately 1.5% of the entire youth, adolescent and young adult population enrolled in Hockey Canada programs in the 2017-2018 season (approx. n= 345 000).[12] Males (n=4176) comprised 79.9% of the entire sample. The mean age was 13.6 years old (SD 2.67) for the entire sample, 13.4 years old (SD 2.25) for males, and 14.3 years old (SD 3.81) for females. Approximately 22% of all athletes had indicated they had been diagnosed with at least one prior concussion. The number of males and females whom reported a history of

Table 3.

Cross tabulation of number of concussions by age and select medical diagnoses among male Canadian ice hockey players aged 10 to 25 years old presenting to CCMI clinics between 2014 and 2017.
*Legend: learning exceptionality = aggregate variable containing athletes who self-reported a learning disability, ADD or ADHD diagnosis; prior psychiatric diagnosis = aggregate variable containing athletes self-reporting a diagnosis of depression or anxiety; * cells had lower than expected counts – interpret p-value with caution.*
Note: percentages reflect the proportion of the row total.

	Number of Prior Concussions									
Age Category	**0**		**1**		**2**		**3+**		χ^2	**P-Value**
Youth (10 to 12 yrs.)	1264	82.4	202	13.2	57	3.7	11	0.7	106.7	<0.001
Adolescent (13 to 18 yrs.)	1935	76.3	385	15.2	144	5.7	71	2.8		
Young Adult (19 to 25 yrs.)	66	61.7	15	14.0	11	10.3	15	14.0		
Medical History										
Learning Exceptionality	180	69.0	38	14.5	21	8.1	22	8.4	52.6	<0.001
Prior Psychiatric Diagnosis*	56	58.3	13	13.6	14	14.6	13	13.5	75.7	<0.001
Sleep Disorder *	16	69.6	3	13.0	1	4.4	3	13.0	11.7	0.008
History of Headache	528	73.1	118	16.3	50	6.9	27	3.7	18.5	<0.001

Table 4.

Cross tabulation of number of concussions by age and select medical diagnoses among female Canadian ice hockey players aged 10 to 25 years old presenting to CCMI clinics between 2014 and 2017.
*Legend: * cells had lower than expected counts – interpret p-value with caution.*
Note: percentages reflect the proportion of the row total.

	Number of Prior Concussions									
Age Category	**0**		**1**		**2**		**3+**		χ^2	**P-Value**
Youth (10 to 12 yrs.)	338	87.6	36	9.3	10	2.6	2	0.5	103.2	<0.001
Adolescent (13 to 18 yrs.)	417	77.5	81	15.1	29	5.4	11	2.0		
Young Adult (19 to 25 yrs.)	65	52.9	24	19.5	18	14.6	16	13.0		
Medical History										
Learning Exceptionality*	23	69.7	2	6.0	6	18.2	2	6.1	13.2	0.004
Prior Psychiatric Diagnosis*	26	46.4	13	23.2	9	16.1	8	14.3	52.5	<0.001
Sleep Disorder*	3	50.0	0	0	2	33.3	1	16.7	14.3	0.003
History of Headache	151	68.4	42	19.0	16	7.2	12	5.4	18.8	<0.001

concussion declined as the number of prior concussions increased (i.e., fewer individuals reported multiple concussions compared to a single concussion). Sex and number of prior concussions were not significantly associated with one another ($\chi^2(3) = 1.44$, p=0.70). Stratified by age, a similar trend was identified; within each age group, fewer athletes reported multiple prior concussions compared to zero or one prior concussion. The association identified between age and number of prior concussions was significant ($\chi^2(6) = 206.8$, p=<0.001).

Calculated odds ratios provided an indication as to the effect size between age, sex, health history and history of concussion. Like above, athletes in the older age group were 2.88 times more likely (CI 95%, 2.20 to 3.77) to have had a history of concussion compared to younger athletes. The odds of having a history of concussion was not significantly associated with an athlete's sex. Having had at least one prior concussion was significantly associated with a history of learning disorder (OR=1.66, p<0.001), a previously diagnosed psychiatric illness (OR=3.20, p<0.001) and a history of headache (OR=1.52, p<0.001). History of sleep disorder was the only health condition not found to increase an athlete's odds of having a history of concussion (p=0.12).

Tables 3 (males only) and 4 (females only) stratify number of prior concussions by age and medical history. Among male athletes, number of prior concussions was significantly associated with an increase in age ($\chi^2(6) = 106.7$, p <0.001). Irrespective of age, fewer males reported a history of multiple concussions (i.e., two and three or more) compared to males who reported zero or one. One exception was noted among young adult males; a greater proportion of males reported three or more prior concussions (14.0%) as compared to two (10.3%). Significant associations between number of prior concussions and each health condition were also identified. Note the low cell counts for prior psychiatric diagnosis and sleep disorder categories.

Similar to findings among males, data from female athletes indicated there was a significant association between number of prior concussions and age ($\chi^2(6) = 103.2$, p <0.001). Again, fewer females reported multiple concussions compared to zero or one. Significant associations between each health condition and number of concussions was identified, but due to low cell counts (except for a history of headache) findings should be interpreted with caution. When comparing concussion histories across sex groups, a greater proportion of males reported a history of concussion (of any number) within each of the age categories. Two exceptions were noted; a greater percentage of females in the young adult age category reported one (19.5% vs. 14%) and two prior concussions (14.6% vs. 10.3%) compared to males in the same age category.

A positive association was identified between PCSS severity score and prior number of concussions as seen in Figure 2; symptom severities increased with each additional concussion that was reported. Symptom severity score appeared to be similar among the sexes for zero and one prior concussion. However, male and female symptom severities diverged in the two and three or more concussion categories, with females having higher symptom severity scores than their male counterparts. Pairwise comparisons and tests of significance were not calculated.

Discussion

With respect to the entire sample, 21.8% of athletes reported that they had been diagnosed with a least one prior concussion. Estimates were comparable within each sex group (21.8% and 21.3% for males and females, respectively). Prevalence declined as number of concussions increased (i.e., fewer athletes reported two or three or more concussions as compared to one or none). A similar trend was identified within each sex group, with the exception of males in the young adult category where more athletes reported three or more concussions (14%) than two (10.3%). Sex was not significantly associated with number of prior concussions, but age was. Categorized as a young adult and having an additional health condition was associated with an increase in the odds of having a history of concussion. Similar trends of association were noted within each sex group. With respect to baseline PCSS severity scores, females with a history of two and three or more concussions reported greater symptom severities at baseline than did males with the same concussion histories.

Four previously published investigations (three pertaining to high school aged athletes and one to college aged athletes) on the prevalence of concussion in ice hockey were identified and compared with our findings. Similar to our investigation, Brooks *et al*. also examined concussion prevalence among Canadian youth ice hock-

ey players aged 13 to 17.9 years old (n=615).[27] Among their total sample, 62.1% of athletes reported no history of concussion, 30.1% reported one (males = 32.9%; females = 20.4%), 5.9% reported two (males = 6.2%; females = 4.1%), and 1.1% reported three prior concussions (males = 1.2%; females = 1.0%). Notably, the proportion of athletes within Brooks' sample whom reported a history of at least one prior concussion was more than double that of our athletes in the same age group (i.e., 13 to 18 years old; 30.1% vs. 14.2%). Larger discrepancies in prevalence were also noted among Brooks' athletes than ours when comparing number of previous concussions by sex. Kontos *et al*. also calculated prevalence of concussion among ice hockey players aged 12 to 18 years old (n=397), but included only American athletes.[28] Findings indicated that 73.5% of their athletes had no history of concussion, 18.6% reported one, 6.3% reported two and 0.8% reported three prior concussions; prevalence estimates for zero and one prior concussion were much closer to ours than Brooks' were. In a 2018 multi-sport study conducted by Veliz *et al*. on American 8th, 10th and 12th grade students (n=25 048), 81.6% of students reported no prior concussion and 18.4% reported at least one.[29] Again, this was similar to the prevalence for the same concussion categories within our investigation (15.2%). Looking exclusively at the students whom were ice hockey players (n=391), 23.0% reported one and 15.9% reported a history of multiple concussions, both of which were much greater than the estimates generated from our sample of the same age. Overall, prevalence estimates generated from our investigation for the adolescent age group alone (i.e., 13 to 18 years old) were similar but slightly more conservative than those referenced above. For example, 15.2% athletes in our adolescent age group (i.e., 13 to 18 years old) reported one prior concussion as compared to 30.1%[27], 18.6%[28], and 23.0%[29]. When considering all athletes included in our sample (regardless of age) who reported a least one prior concussion, our prevalence estimates (~22%) come much closer to that of the estimates referenced above. One important consideration for readers to note here is that athletes with a history of concussion face an increased risk for subsequent concussion.[30,31] In other words, once an athlete has sustained one concussion, they are more likely to sustain another.

The final study for comparison included college aged athletes. Katz *et al*. captured n= 15 681 National Collegiate Athletic Association (NCAA) athletes, of which n=221 were ice hockey players.[32] They found that 58% of all players, 57% of males and 64% of females reported no prior concussion. Prevalence estimates declined with each additional concussion (i.e., one prior concussion; total sample: 28%, females: 25%, males: 29% vs. two prior concussions; total sample: 9%, females: 8%, males: 10% vs. three or more prior concussions; total sample: 5%, females: 4%, males: 5%), which was the same trend identified within our investigation and both Brooks' and Kontos'. Prevalence estimates from our investigation were more conservative for athletes within the same age range (i.e., young adult; 19 to 25 years old) whom had zero (57.0%) or one prior concussion (17.0%), but inflated for athletes with two (12.6%) or three or more concussions (13.5%). Similar to our findings, Katz' prevalence estimates declined as the number of prior concussions increased. In general, prevalence of concussion (of any number) was greater among Katz' collegiate aged athletes than our high-school aged athletes and those in the studies described above. This trend is likely explained by greater cumulative athletic exposure hours that accompany older age (i.e., a 21-year-old will have participated in and accumulated a greater number of hours playing in ice hockey games and practices than a 15-year-old who started playing hockey at the same age).

Readers may wish to note that any discrepancies identified with respect to lifetime concussion prevalence estimates between the referenced studies may be explained by methodological differences in how history of concussion was assessed and reported (i.e., diagnosed concussions as reported in a medical record vs. self-reported diagnosed concussions only vs. self-reported diagnosed and undiagnosed concussions vs. parent and/or athlete self-reports); each of the above referenced studies, including our own, assessed athlete concussion histories in a slightly different manner. In the case that concussion histories were generated based on athlete self-report, poor memory recall and limited knowledge of concussion and the associated symptoms may also explain variability in prevalence estimates. Although each of the above studies included ice hockey players, concussions reported by athletes did not appear to be limited to only those that occurred whilst playing ice hockey.

Within our investigation, athletes who reported an additional health condition (other than a sleep disorder)

were significantly more likely to have had a history of concussion than athletes who did not. However, timing of diagnosis in relation to the occurrence of concussion was not acquired from athletes. As such, definitive statements pertaining to if these conditions increased the athlete's risk for concussion cannot be made. Health conditions, like those assessed within our investigation, may have important implications on recovery outcomes following a SRC. One systematic review summarizing predictors of clinical recovery following a SRC in athletes found several pre-injury health conditions (including but not limited to ADHD, a history of headache, familial and personal psychiatric history, as well as previous concussion) were associated with poor post-concussion outcomes (i.e., persisting post-concussive symptoms) [24]. Refuting evidence was also cited.

At time of baseline assessment, our athletes were also asked to report on the presence and severity of concussion-like symptoms via the PCSS. Both males and females reported similar symptom severities when stratified by prior number of concussions (see Figure 2). This trend is both supported[33, 34] and refuted[35-37] by the existing literature. Findings from one large scale meta-analysis (n=42 493) on high school and college aged athletes (inclusive of ages 12 to 26 years old) indicated that females were 43% more likely to report at least one concussion-like symptom at baseline compared to males. Specifically, females had significantly higher odds of reporting: vision/ hearing problems, headache/migraine, difficulties concentrating, energy/sleep and emotional disturbances.[38] Irrespective of sex, a history of concussion has been associated with greater concussion-like symptom reports in the absence of injury (i.e., at baseline).[27, 39] The take home message here being that clinicians must consider the impact that single and multiple prior concussions have on the short and long-term health of young athletes.[31]

Examining injury reporting behaviours specific to young athletes is critical to interpreting prevalence trends. This is especially important given the lack of an objective test for diagnosing a concussion. Unless observed by a parent/coach/trainer/game official, athletes must come forward to seek medical attention if they suspect a concussion has occurred. As noted above, approximately half of all suspected concussions go unreported by youth athletes.[8] Sex has also been shown to impact reporting behaviours; in one study, male high school aged athletes were 4 to 11 times more likely than females to not report a suspected concussion to their coach or parent.[40] Despite sex differences, athletes cited the following reasons for not reporting a SRC: 1) "I thought my coach would get mad", 2) "I thought my teammates would think I'm weak", 3) "I didn't want to let my team mates down", 4) "I thought my coach would think I'm weak", and 5) "I thought my parents would get upset".[40] In a separate study, being female was associated with an increase in SRC knowledge and improved concussion reporting behaviours as compared to males.[41] With respect to age, younger athletes may not be as familiar with concussion symptoms or the negative consequences associated with non-reporting behaviours, thus resulting in fewer injury reports. Clinicians should note that athletes who fail to report a SRC may face an increased risk for a subsequent concussion and poor long-term health outcomes.[42]

Strengths and limitations

To our knowledge, this study offers the largest sample of ice hockey players compared to the other prevalence studies referenced. However, athlete records included in our study represent a very small proportion (~1.5%) of all Hockey Canada registrants. As such, the sample captured and prevalence estimates generated may not be representative of the population as a whole. Our study also offers comparison of concussion prevalence rates across a wide age range (i.e., middle school to collegiate aged athletes). Noting however that college aged athletes were largely underrepresented within our sample (4.4%). Caution should be used if generalizing study findings to a young adult age group. One unique aspect of our study was that we examined the association and calculated odds ratios between several health conditions and history of concussion. Due to the limited health history information that was collected from athletes, the identified relationships lack temporality; we do not know if a reported health condition occurred before or after a concussion occurred (if any). Furthermore, athletes were only asked to report *diagnosed* health conditions. We did not cross reference athlete self-reports with official medical records. Finally, diagnosed concussions reported by the athletes were not limited to those that occurred whilst playing hockey. As such, we cannot make any conclusions regarding prevalence of concussion within the sport of ice hockey exclusively. Athletes also provided retrospective injury self-re-

ports which can be problematic and subject to poor memory recall. Athletes may have inflated or underreported their history of concussion due to any number of reasons, some of which are discussed above. Reported diagnosed concussions were not cross-referenced with official medical records.

Conclusions

Approximately 22% of ice hockey players presenting to CCMI clinics reported at least one prior concussion, which was similar to previously published prevalence estimates. A smaller proportion of athletes (~7.6%) reported a history of two or more prior concussions.

Sex was not found to be significantly associated with an increase in the odds of having a history of concussion. Although young adults made up a very small percentage of the total sample, a greater proportion of these athletes reported a history of concussion (of any number) compared to younger age groups. Older age was significantly associated with having a history of concussion. This was attributed to a greater overall athletic exposure (i.e., greater cumulative number of hours spent in games, practices and training) to injury. Athletes whom reported an additional health condition (other than a sleep disorder) were significantly more likely to have a history of concussion compared to those athletes without an additional diagnosis. Lastly, number of prior concussions appears to be positively associated with concussion-like symptoms reported via the PCSS in the absence of injury.

Although risk for SRC cannot be removed entirely from the sport of ice hockey, revision of player-to-player contact policies in youth hockey (such as barring body checking under the age of 13) has consistently demonstrated a protective effect in reducing athlete risk for SRC.[31] Given the proportion of ice hockey players whom reported a previous concussion, stakeholders are encouraged to consider the availability of educational, injury management and injury prevention strategies that are in place and accessible at the athlete level. Moving forward, subsequent investigations should seek to establish prevalence estimates of SRC that occur *while playing* ice hockey. Acquiring information from athletes with respect to timing of an additional health condition (i.e., present pre- or post-concussion) would help to better define any risk and recovery implications that may exist. In turn, clinicians can offer a more informed approach to managing and protecting athletes at risk for prolonged recovery and subsequent SRCs.

Acknowledgments

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Multimodal non-surgical intervention for individuals with knee osteoarthritis: a retrospective case series

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Introduction: *The purpose of this study is to review the pain and functional outcomes of a multimodal intervention in three patients with knee osteoarthritis (OA). This study explores how manual therapy can be delivered within an evidence-based framework for the management of knee OA.*

Methods: *Medical records were reviewed for three patients with knee OA who underwent a standardized multimodal intervention including education, exercise, and manual therapy. Changes in pain intensity and function from baseline to post-intervention were calculated and compared to thresholds for minimal clinically important differences.*

Results: *One participant met the threshold for clinically significant improvement in pain and two*

Introduction : *Le but de cette étude est d'examiner les résultats en matière de douleur et de fonction d'une intervention multimodale chez trois patients atteints d'arthrose du genou. Cette étude explore comment la thérapie manuelle peut être dispensée dans un cadre fondé sur des données probantes pour la prise en charge de l'arthrose du genou.*

Méthodes : *On a examiné les dossiers médicaux de trois patients atteints d'arthrose du genou qui ont subi une intervention multimodale normalisée comprenant l'*éducation, l'*exercice et la thérapie manuelle. Les changements concernant l'intensité de la douleur et la fonction entre le début et la fin de l'intervention ont été calculés et comparés aux seuils pour connaître les différences minimales importantes sur le plan clinique.*

Résultats : *Un participant a atteint le seuil d'amélioration significative du point de vue clinique*

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participants for function. No adverse events were reported.

Conclusion: *Combined education, exercise, and manual therapy delivered over a 6-week period improved function in two of the three patients reviewed. Higher quality research is required to explore whether this multimodal intervention may improve outcomes in individuals with knee OA.*

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KEY WORDS: chiropractic, knee, osteoarthritis, multimodal, treatment

pour ce qui est de la douleur et deux participants pour ce qui est de la fonction. Aucun effet indésirable n'a été signalé.

Conclusion : *L'éducation, l'exercice et la thérapie manuelle combinés sur une période de six semaines ont amélioré les fonctions chez deux des trois patients examinés. Des recherches de meilleure qualité sont nécessaires pour déterminer si cette intervention multimodale peut améliorer les résultats chez les personnes atteintes d'arthrose du genou.*

(JACC. 2019;63(2):92-99)

MOTS CLÉS : chiropratique, genou, arthrose, multimodal, traitement.

Introduction

Osteoarthritis (OA) is the 12th leading cause of disability in the world and 10th leading cause in Canada.[1] It is estimated that 13% of Canadians[2] and over 300 million people worldwide have OA[1]. Due to the increasing age of the population and rates of obesity[3], the prevalence of OA in Canada is expected to increase to 25% by the year 2040, placing a large burden on the health care system[2]. The 2011 report from the Arthritis Alliance of Canada[2] recommends the development of pain management interventions to help reduce the growing burden of knee OA.

The Canadian Chiropractic Guideline Initiative (CCGI) has adopted the National Institute for Health and Care Excellence (NICE) OA guidelines.[4] The NICE guidelines recommend education and self-management, including exercise, should be offered as core treatments to individuals with OA.[4] These recommendations are also supported in guidelines from the Osteoarthritis Research Society International (OARSI) and European League Against Rheumatism.[5,6] A recent Cochrane Review found that self-management education programs may improve pain, function and symptoms for people with OA[7], while a second Cochrane Review concluded that therapeutic exercise can reduce knee pain and improve physical function among people with knee OA[8].

Manual therapy is not considered a core treatment for knee OA due to the lack of high quality evidence that currently exists.[9] However, almost 50% of individuals with knee OA use some form of complementary and alternative medicine, which includes manual therapy.[10] The existing evidence base supporting the use of manual therapy for knee OA is comprised of only a small number of trials and review papers.[11] The most recent systematic review found short-term benefits favouring manual therapy over exercise alone.[12]

International guidelines differ on the use of manual therapy for knee OA. The CCGI-adopted NICE guidelines state that joint manipulation and stretching can be considered, but particularly for those with hip OA.[4] The American College of Rheumatology guidelines and a review of guidelines by the U.S. Bone and Joint Initiative recommend the consideration of manual therapy only when in combination with exercise.[13,14] These are generally weak recommendations, as little evidence is available to evaluate the efficacy of manual therapy as part of multimodal treatments for patients with knee OA.

The objective of this report is to describe the pain and functional outcomes of a multimodal, non-surgical intervention comprised of education, exercise, and manual therapy in three patients with knee OA.

Methods

Design

This was a retrospective case series aimed to describe the multimodal non-surgical management of three patients

with knee OA. The consenting patients' medical records were retrospectively reviewed to extract basic demographic information and outcome measure data from the time of assessment and upon completion of a standardized treatment program.

Protection of Human Participants

The Unity Health Toronto Research Ethics Board (18-192[C]) and Canadian Memorial Chiropractic College Research Ethics Board (1902X01) gave approval for this study. Medical charts were screened for eligibility by the research team. The current treating chiropractor explained the study to eligible participants and referred interested participants to the research team. The research team met with eligible participants to provide detailed information of the study and obtain informed consent. All participants in this study agreed to participate and provided informed consent.

Participants

Medical records were reviewed for three consecutive patients from the Department of Family and Community Medicine chiropractic clinic at St. Michael's Hospital presenting in November 2017. Individual records were considered eligible for review if they fulfilled the following inclusion criteria: 1) clinical examination evidence of knee osteoarthritis[4], 2) ability to read/write English, 3) attendance to minimum five intervention sessions, and 4) ability to engage in a daily mild exercise program. Clinical examination evidence of knee osteoarthritis was determined by the treating clinician, taking into consideration the NICE guidelines for diagnosis.[4] The guidelines recommend a diagnosis of OA if the patient is 45 years or older, has usage-related joint pain and either no morning knee stiffness or stiffness of 30 minutes or less. Individual medical records with any of the following criteria were excluded: 1) have been told by a physician that they should not engage in physical exercise, 2) previous knee arthroscopy or joint replacement, or 3) history of any condition that affects the individual's ability to exercise.

Intervention

All participants received a multimodal and self-management training program as part of their chiropractic treatment. The program consisted of one-on-one treatment sessions with one of the authors. Each session was approximately 15 minutes in duration at a frequency of one time per week for six weeks. The intervention was tailored and progressed for each individual, with an emphasis placed on self-management education. The components of the program were as follows:

Education

Patients received education based on current management guidelines[4-6], and were adapted from the educational component of the Good Life with osteoArthritis in Denmark (GLA:D®) program, which are published elsewhere.[15] In brief, the main educational themes consisted of disease etiology, pain science, natural history, prognosis and management options for knee OA. Patients were informed that exercise and weight management comprise the main management program for all individuals with knee OA[6], while manual therapy may confer additional benefit for some patients. They also received instruction on how to self-manage symptoms and maintain daily routines. Educational concepts were discussed and reinforced on an ongoing basis at each treatment session.

Exercise

Patients received a structured home exercise program adapted from the neuromuscular training program described by Ageberg *et al*.[16] The purpose of the neuromuscular training program was to improve sensorimotor control of the lower limb. Specific exercises included sit-to-stand, forward and sideways lunges, quadriceps and hamstring strengthening, and hip abduction exercises. All exercises were individually tailored for each patient and progressed over the course of the intervention.

Manual Therapy

All patients received the same manual therapy protocol aimed at decreasing pain and increasing range of motion at the knee. Joint and soft tissue mobilization and manual muscle stretching was performed for all patients.[17] At each session, manual therapy was directed to both the tibiofemoral and patellofemoral joints, as well as surrounding musculature. The specific manual therapy techniques used were determined based on identified functional impairments and tolerances for the specific patient. For example, if a patient was deemed to have reduced knee flexion, a flexion mobilization of the tibiofemoral joint was performed or if a patient reported increased pain in the

quadriceps muscle group, an increased focus was placed on delivering soft tissue therapy to this muscle. This method of delivering manual therapy has been used in previous studies of degenerative conditions with good results.[18,19]

Outcomes

Patient demographics including: age, sex, and location of knee OA (left, right, or both) were extracted. Data from the Numeric Pain Rating Scale (NPRS) and Knee injury and Osteoarthritis Outcome Scale Physical Function Short Form (KOOS-PS) were collected at baseline and upon completion of the intervention at six weeks. These measures are included in the international standard set of outcome measures for individuals with knee OA.[20] Adverse events were tracked by asking the patient at each treatment session and recorded in the electronic medical record.

The NPRS is an 11-point numeric scale of pain intensity in individuals with chronic pain, where 0 represents "no pain" and 10 represents "worst pain".[21] Psychometric testing has shown the NPRS to be a valid and reliable measure of pain intensity[21] and the minimal clinically important difference (MCID) is 2 points[22].

The KOOS-PS is a 7-item measure used to quantify an individual's difficulties with physical activity due to their knee problems.[23] All items are scored on a 5-point Likert scale.[24] Total scores are then transformed to an interval score of 0 (no difficulty) to 100 (extreme difficulty) using a conversion chart.[23] Psychometric testing has shown the KOOS-PS to be a valid and reliable measure of knee-related function in groups with knee OA and its use has been endorsed by OARSI and Outcome Measures in Rheumatology Clinical Trials.[25] The MCID for the KOOS-PS is 12 points.[26]

Table 1.
Participant demographic characteristics.
Note. OA = Osteoarthritis

Participant	Age	Sex	Location of Knee OA
A	50	Male	Right
B	59	Female	Both
C	42	Female	Left

Data Analysis

Descriptive statistics for each patient were collected. Changes in NPRS and KOOS-PS scores from baseline to post-intervention were calculated for each patient. Comparison to established MCID was performed. No minimum sample size requirements are required as this was a descriptive report of three patients' response to intervention. All participant data was anonymized and securely stored on the St. Michael's Hospital investigator's hospital computer secure server.

Results

Table 1 presents descriptive data for each participant. The average age of the participants was 50.3 years. Two participants had unilateral knee OA, while one individual had bilateral knee OA. The mean NPRS score prior to intervention was 6, while the mean KOOS-PS score prior to intervention was 57.6.

Table 2 shows the changes in NPRS and KOOS-PS scores from pre to post-intervention for each participant. Participant A reported a 1- and 37.4-point improvement in NPRS and KOOS-PS scores, respectively. Participant B showed no change in NPRS scores and a 7.0-point improvement in KOOS-PS scores. Participant C had a

Table 2.
*Change in outcome measures. Note. NPRS = Numeric Pain Rating Scale, KOOS-PS = Knee injury and Osteoarthritis Outcome Scale Physical Function Short Form, * = clinically important change*

Participant	$NPRS_{pre}$	$NPRS_{post}$	$NPRS_{change}$	$KOOS\text{-}PS_{pre}$	$KOOS\text{-}PS_{post}$	$KOOS\text{-}PS_{change}$
A	6	5	-1	77.7	40.3	-37.4*
B	5	5	0	44.0	37.0	-7.0
C	7	1	-6*	51.2	10.5	-40.7*

6- and 40.7-point improvement in NPRS and KOOS-PS scores, respectively. Only participant C met the threshold for a clinically significant change in pain, while participants A and C had clinically significant improvements in function. No adverse events were reported by any of the participants.

Discussion

This case series reviewed the clinical outcomes of three patients with knee OA undergoing a multimodal intervention consisting of self-management education, exercise, and manual therapy. One of the three participants showed clinically significant improvement in pain, while two of the three participants showed clinically significant improvement in function.

Participant B showed an improvement in function scores that did not meet clinical significance and did not show any improvement in pain scores. It is possible that this intervention was not effective, especially considering the small sample size. However, this participant showed mild pain levels and functional impairment prior to treatment, which may have limited their room for improvement. It should also be considered that some individuals may not respond to conservative treatment. Heterogeneous treatment responses to exercise for knee OA have been shown, including individuals with no improvement over a 12-week intervention.[27] It is possible the same applies for multimodal interventions including exercise and manual therapy, or that a 6-week intervention is not long enough for all individuals to experience benefit.

A strength of this intervention is the inclusion of manual therapy into an evidence-based treatment framework including the core treatments for knee OA; self-management education and therapeutic exercise.[4-6] Case reports of manual therapy in conjunction with exercise for knee OA have been published.[28,29] However, these reports describe interventions that do not meet recommendations from current guidelines. For example, neither report delivers patient education, a key component of knee OA management. Furthermore, one report includes the use of electrical stimulation and ultrasound[29] and the other includes platelet-rich plasma injections[28], both of which are not recommended for the management of knee OA. The strength of this case series is the inclusion of manual therapy as part of an evidence-based and guideline-recommended intervention framework for the management of knee OA.

The role of manual therapy as a stand-alone intervention for patients with OA is currently unclear. Knee joint mobilization may modulate central nociceptive pathways, decreasing the pain experienced by the individual[30] and evidence exists supporting the use of manual therapy for patients with hip OA[9,31]. However, systematic reviews on manual therapy for knee OA differ in their findings. One review found inconclusive evidence for the use of manual therapy for patients with knee OA, as only three small randomized controlled trials were available[9], while a more recent review concluded manual therapy offers short-term benefits compared to exercise alone[12].

The combination of manual therapy and exercise may offer better outcomes for patients, but a systematic review demonstrated significant improvements in pain outcomes only.[32] Multiple trials have evaluated combined exercise and manual therapy.[33-37] A wide range of specific exercise and manual therapy protocols have been used in these studies, but all trials report some positive effects for patients in pain and function. The findings of these trials further support our results, as there is a clear trend of improvement when compared to controls. However, a limitation of these trials is non-adherence to current management guidelines, as they lack self-management education.

Following review of the literature, two trials that have evaluated manual therapy in conjunction with education and exercise were identified.[38,39] The first trial included education, exercise, manual therapy and taping for patients with patellofemoral OA.[38] When compared to education alone, the multimodal treatment had significantly greater improvements in the number of patients reporting being much improved and in pain. However, unlike this study, patients with tibiofemoral OA were excluded, likely limiting the utility of this intervention for the general public. The second trial found no difference comparing education, exercise, and manual therapy to a Tai Chi intervention.[39] Unfortunately, it is unclear if all participants received manual therapy or if it was at the discretion of the clinician. It is possible a more standardized manual therapy approach could confer additional benefit to patients.

Individually, both structured education and therapeutic exercise have been shown to effectively manage knee OA symptoms. Cochrane reviews of self-management education[7] and exercise[8,40] have found these inter-

ventions provide clinically meaningful outcomes with a low likelihood of causing harm[7]. Weight management is a key component of self-management education and a core treatment for knee OA in patients who are overweight or obese.[4] Weight reduction through both exercise or dietary interventions has shown that a 5% reduction in body weight over a 20-week period can significantly improve disability.[41] While no patients in this study were deemed to require weight loss education or intervention, weight-loss interventions should be considered for other patients with knee OA as indicated.

Structured education and exercise programs for individuals with hip or knee OA are available, including the GLA:D® program.[15] In Denmark, GLA:D® has shown improvements in pain, function, quality of life, and medication use in 9825 patients.[15] As such, GLA:D® is an effective alternative to current standards of care for knee OA.[42] GLA:D® Canada began in 2015 and preliminary results suggest as good or better results than in Denmark.[43] It is not surprising our intervention showed positive results, as the data available from GLA:D® also supports the use of combined education and exercise. The inclusion of manual therapy in our multimodal intervention may result in greater short-term symptomatic relief, as this has been previously demonstrated.[34,36] It would be of value in the future to evaluate the effectiveness of including manual therapy with the GLA:D® program.

Limitations

The specific treatment protocol used in this intervention, while supported by available evidence, was implemented and adapted at the discretion of the treating clinician. It is possible a more optimal combination of education, manual therapy, and exercise exists. For example, only verbal education was provided and no written education was given. There are also various manual therapy techniques and exercise protocols that may be considered and this intervention can be used as a starting point when designing other multimodal interventions.

It is possible that co-interventions may have contributed to the results observed, as participants were permitted to seek other treatments. For example, the use of medications may have influenced the results. However, no change in prescribed medication was noted in any of the medical records, nor did any participant personally report changes in medication use or use of other interventions. Lastly, the results of this case series must also be interpreted with caution due to the small sample size and lack of control group.

Future Research

This case series should be viewed as hypothesis generating and the results should be used to inform further high-quality research designs. It may be possible that this multimodal approach can improve disability in patients with knee OA. A prospective cohort study with more participants should be performed and if positive results are found, a randomized controlled trial may be conducted, including evaluation for possible adverse events. These types of studies, at minimum, should be performed prior to the adoption of this intervention as a standardized approach in practice. We recommend that manual therapy be tested in conjunction with education and exercise, as this is in-line with current guideline recommendations for the management of knee OA.

Conclusion

A 6-week multimodal, non-surgical intervention including education, exercise, and manual therapy appears to improve function for two of the three individuals with knee OA and improve pain for one of the three patients reviewed in this case series. No adverse events were reported by any of the participants. The design of more robust studies to further examine the effectiveness of this multimodal intervention is warranted.

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The effect of posture on neck proprioception and head/neck stabilization in asymptomatic participants

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Objective: *We sought to determine the effect of different body postures on neck proprioception and head/neck stabilization.*

Methods: *Twelve healthy college students completed a head repositioning task and a 'head still' task while wearing a headpiece (helmet) with laser fixed on top during standing, kneeling, sitting, and sitting with stabilization. Video data of the laser dot coordinates on a projection screen were obtained to examine the accuracy of the two tasks.*

Results: *There was a significant effect of both posture and vision for both vertical and horizontal head movements during the head still task. Standing and kneeling generated more variable head movement than sitting with or without stabilization. Posture did not significantly affect head repositioning accuracy.*

Conclusion: *For healthy young adults, clinicians*

Objectif : *Nous avons cherché à déterminer l'effet de différentes postures corporelles sur la proprioception du cou et la stabilisation tête/cou.*

Méthodes : *Douze étudiants en bonne santé de niveau collégial ont accompli une tâche de repositionnement de la tête et une tâche d'immobilisation de la tête tout en portant un casque avec un laser fixé sur le dessus pendant qu'ils étaient debout, à genoux, assis et assis en état stable. Des données vidéo des coordonnées des points laser sur un écran de projection ont été obtenues pour examiner la précision des deux tâches.*

Résultats : *On a observé un effet significatif de la posture et de la vision pour les mouvements verticaux et horizontaux de la tête pendant l'immobilisation de la tête. Le fait de se tenir debout et de s'agenouiller a généré un mouvement de la tête plus variable que le fait de s'asseoir en étant ou non stable. La posture n'a pas eu d'effet significatif sur la précision du repositionnement de la tête.*

Conclusion : *Pour les jeunes adultes en santé, les*

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and researchers need to be concerned with postural influences on tasks that involve head/cervical spine stabilization, but not head repositioning accuracy.

(JCCA. 2019;63(2):100-110)

KEY WORDS: posture, cervical spine proprioception, head repositioning, motor control, postural balance

cliniciens et les chercheurs doivent se préoccuper des influences posturales pour les tâches qui font appel à une stabilisation de la tête et de la colonne cervicale, mais non de la précision du repositionnement de la tête.

(JACC. 2019;63(2):100-110)

MOTS CLÉS : posture, proprioception de la colonne cervicale, repositionnement de la tête, contrôle moteur, équilibre postural.

Introduction

Proprioception encompasses the sensation of joint movement (kinesthesia) and joint position (joint position sense).[1-5] Perception of the orientation of our head in space as well as on the trunk demands not only the contribution of vestibular and visual cues but also proprioceptive information from the neck[1] and likely from other body regions.[6, 7] A few reflexes contribute to orientation of the head and trunk including the cervico-ocular (COR), vestibulo-ocular (VOR), vestibulospinal (VSR), vestibulo-collic (VCR) and cervicocollic (CCR).[2, 8] The COR stabilizes the eye in response to trunk-to-head movements, the VOR stabilizes gaze during head motion, the VSR produces compensatory body movements to stabilize the body in space, the VCR stabilizes the head relative to space and the CCR stabilizes the head relative to the trunk.[2, 8] The receptors that are responsible for proprioception are found in muscle-tendon units (e.g., muscle spindles, golgi tendon organs), joints (e.g., Ruffini endings, Pacinian endings) and skin (e.g., hair follicles, Ruffini endings).[9] Proprioception is essential for proper joint function in sports and activities of daily living or work-related tasks.[1, 2, 10]

Tests of proprioception have differentiated between the 2 main proprioceptive functions—detection of static position and detection of motion.[4] Detection of motion can be assessed by the threshold of motion detection (e.g., amount/speed of motion required for detection to occur) and the direction of motion (e.g., flexion or extension), which is considered a discrimination task.[4] On the other hand, tests of neck joint position sense to date have focused on two methods involving the accuracy of: a) position-matching tasks involving relocation of the head to a set point or angle or; b) relocation to the natural (neutral) head posture. The reliability of these tests are good to excellent.[11] Accuracy is derived by calculating the difference between the reference target (e.g. natural head posture) and the reproduced position or angle of the head. This accuracy is frequently reported as the absolute value of the constant error computed along the global (hypotenuse), the horizontal (x), and the vertical components (y) of the reproduced position relative to the natural head position. Some factors shown to reduce the accuracy of neck proprioception include fatigue, and pathology such as whiplash and neck pain.[2, 4, 12, 13]

One factor that has not been well studied is the contribution of different body postures to the accuracy of head-neck position sense. A recent study looking at the effect of different induced head-neck-jaw postures on head-neck position sense among healthy subjects did not find any significant difference between postural configurations.[14] Specifically, this study evaluated cervicocephalic kinesthetic sense while standing, habitual sitting, habitual sitting with clenched jaw and habitual sitting with forward head posture during right rotation, left rotation, flexion and extension.[14] Likewise Teng *et al*.[15] considered the most frequently adopted posture during daytime (sitting or standing) as self-reported data in three groups of individuals, a control group of 20 asymptomatic young adults and two groups of middle-aged adults (20 subjects in each group) with or without a history of mild neck pain. While Teng *et al*.[15] collected self-reported differences in daytime posture between these groups, their study was not designed to prospectively test the differences between groups as a function of posture. However, a study investigating the effect of different sitting postures on

cervicocephalic kinesthetic sensibility found that sitting with arms supported decreased head-neck repositioning error.[16] In this study[16], cervicocephalic kinesthetic sensibility was measured in healthy young adults while in a habitual slouched sitting position with arms hanging by the side, a habitual slouched sitting position with arms unloaded (supported) and during upright sitting position with arms hanging by the side during maximum and 30 degree right, left rotations, flexion, and extension. The authors hypothesized that the sitting posture with supported arms, provided a direct mechanical relaxation of muscles allowing the proprioception receptors of the neck to perform in an optimal manner.[16]

Proprioceptive information from cervical muscles is known to play a major role in the control of posture and gait in humans.[11] When neuromuscular function at the neck is impaired either by pathology[17], trauma[12], or by experimental manipulations in healthy subjects (such as neck fatigue[18]), balance and movement control have been shown to decrease.[11] Similarly, individuals with non-traumatic neck pain perform with longer movement times then healthy control participants during a rapid head movement aiming task when task conditions are adequately challenging.[19] In addition, the gain values of the COR were significantly increased in a whiplash associated disorders population.[20] Accordingly, accurate assessments of neck function and proprioceptive abilities appear to be of great importance. The need to conduct the current study stems from the fact that there is no consensus concerning the best method of assessing neck proprioception in the literature.[1] A systematic investigation of the contribution of how motion or position at various joints contributes to the sense of head position and/or neck proprioception has not been performed.

The current study sought to determine the role of body postures on two tasks – a head repositioning task as well as a head still task. We are not aware of any previous study that has looked at the effect of body postures on these two tasks. Head repositioning appears to be a common outcome measure in the literature for proprioception.[2, 11] Likewise, the ability to maintain our head still is also important since postural control functions in a manner that facilitates other higher order (suprapostural) tasks[21] such as looking and reading[22] which require stabilization of the head. These studies, while showing that postural sway is modulated in a task specific manner also show that postural adaptation involves more than basic reduction or increase of motion; it involves multi-segmental coordination of body segments to achieve a particular goal.

We hypothesized that both joint position error during the head repositioning task as well as variability of head position during the head still task would be reduced when segments other than the neck can contribute to the perception of the orientation of our head in space. Participants performed these tasks in different postures including standing, kneeling, sitting, and sitting with the trunk stabilized under eyes open and eyes closed conditions. We further hypothesized that the more unconstrained/unsupported body segments that participated in accomplishing these two tasks, the less the head position error or variability would be. Our hypotheses assumed that the central processing of proprioceptive inputs that arise from numerous muscles and joints contribute to both the overall awareness and control of body posture.[6, 7] The representation of body geometry is based on proprioceptive inputs (primarily from muscle) that convey information about the position of a given segment with respect to the others. Accordingly, all movements of our multi-joint body are calibrated within the proprioceptive field of a postural space.[7] This has led to the notion that there exists a form of "proprioceptive chain" which functionally links the eye muscles to the foot muscles.[6] Therefore, our study assessed how the proprioceptive chain affects head and neck position sense.

This study has practical significance in that it will help inform clinicians and researchers as to the magnitude to which different body postures contribute to head repositioning accuracy and head stabilization. Secondly, it will provide evidence as to which posture(s) need to be included during neck proprioception testing. Lastly, it may provide a framework from which to assess the effect (if any) of various conditions (e.g. hip arthritis) on neck proprioception.

Methods

Participants

Twelve participants (4 males, 8 females) ranging in age from 20-31 (22.2 ± 2.9, Mean±SD) years participated in this study. Participants were recruited from undergraduate and graduate classes in the Department of Kinesiology and Health at Miami University. The study protocol,

all forms used and the informed consent documents were approved by the Human Subjects Institutional Review Board at Miami University. All participants read and voluntarily signed a written informed consent document and completed a health history questionnaire. Exclusion criteria included a history of neck pain, neurological or vestibular impairment, injury or operation to the cervical spine, injury or illness of ankle(s), knee(s), hip(s), back within the last six months, inability to stand, kneel, or sit, and vision that was not corrected to normal.

Design and procedure

Each participant came to the biomechanics laboratory once for a 60-minute session. Participants wore athletic shorts and a t-shirt during the test to minimize the effect of extra clothing on proprioception. Each participant completed two tasks: head repositioning task and the head still task. These tasks are described in detail below. Participants wore an adjustable bicycle helmet and blindfold (sleeping style mask) during the eyes closed conditions of both the head repositioning task and the head still task.

We used a within-subjects study design to determine the influence of posture on each of the tasks and to control for the potential influence of individual differences. Given the within-subjects design, the sample size was estimated based on similar studies involving clinical and asymptomatic populations.[18, 23-26] Both tasks incorporated four postural conditions (standing, kneeling, sitting, sitting with stabilization). Each task, and each postural condition were counterbalanced. To do so, we created four separate orders of conditions (pseudorandom order) between participants to minimize the potential of order and carryover effects. The order of trials (e.g., eyes open first then eyes closed) within a task condition was the same for every participant.

Head repositioning task

For this task, participants were fitted with an adjustable bicycle helmet with laser pointer fixed on top, while wearing a blindfold. The helmet was placed on the head such that the laser pointer was facing directly forward. The laser was fixed to the top of the helmet. Participants confirmed comfort before participating. A high-speed video camera (capturing at 30 frames per second) with SIMI motion software (Unterschleissheim, Germany) was used to track and determine the coordinates of the laser pointer that was calibrated to a white large projection screen located 2.6 meters in front of the participant (Figure 1). SIMI camera based motion software has demonstrated very good agreement with a sensor-based gait analysis system[27] and has been used in a variety of motion analysis[28-30] applications.

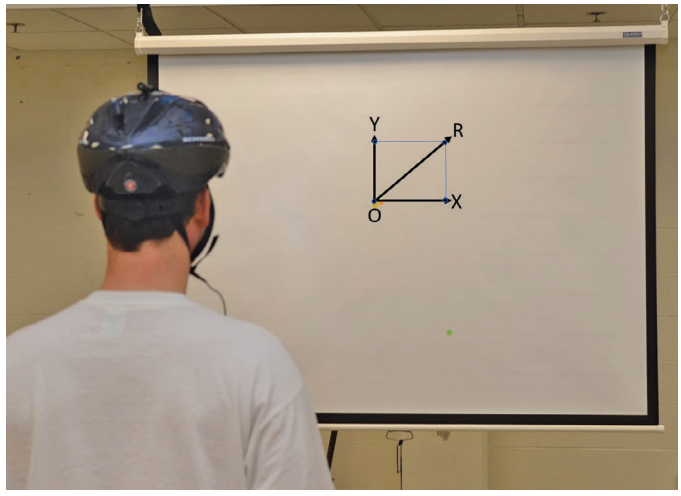

Figure 1.
Experimental setup. Participants wore an adjustable bicycle helmet with a laser pointer on top for both the head repositioning task and the head still task. Head repositioning accuracy (HRA) data involved the measurement of laser dot coordinates. Point O represents the center of the target (coordinates 0,0). Point R represents the position that the laser dot stopped when the head was repositioned after a near-maximal movement. The distance O–R represents the global error of positioning converted into degrees (R) and serves as the outcome of HRA in this study. The horizontal projection (O–X) and the vertical projection (O–Y) indicate the horizontal and vertical components of the global error. Each component was assigned a positive or negative sign according to its position below or above zero on the corresponding axis.

The head repositioning task described below was performed in each of these postures using a counterbalanced order: standing, kneeling, sitting, sitting with stabilization. The support surface for each posture was the hard floor of the laboratory. Standing involved normal upright standing with feet shoulder width apart and arms by their sides. Kneeling involved shoulder width apart knee pos-

ture and an upright trunk. Sitting was on a high back chair with their arms hanging by their sides but their back not touching the backrest. Sitting with stabilization was performed on the same high back chair but their back was touching the backrest. In addition, a Velcro© strap was placed around the subject's trunk and arms (at mid-arm level) and securely fastened to the seatback so as to stabilize the subject and only allow head and neck movements.

Blindfolded subjects with eyes closed, wearing the laser mounted helmet were placed 2.6 meters from the projection screen which was adjustable in height to accommodate all postures. Participants were instructed to: a) memorize their neutral head position, 'put your head into what you think is a straight ahead position' and hold for two seconds; b) perform a near maximal head rotation to the left (or right) for approximately two seconds; and c) to relocate the head to the initial neutral reference head position with maximum precision but without speed instruction and to hold the position for 2 seconds (to allow experimenters to register the position on the screen[31]). Five trials were performed with head repositioning after a right head rotation and five trials after a left rotation under each of the postural conditions. Other authors have found that the greatest test-retest reliability for joint position error testing was obtained with five or more trials.[26] No feedback was given to the subjects regarding their relocation performance. Video capture of the laser defined neutral head and relocated head positions (x, y) on the projection screen was performed. Error distance in centimeters (cm) was derived from the difference between neutral and relocated head positions. This error in centimeters was converted to head repositioning error in degrees (Ø).

Head still task

The same postures and equipment used for the head repositioning task were also used for the head still task. While located 2.6 meters in front of the projection screen, participants were instructed to maintain the laser dot on a colored target circle that was 1.91 cm in diameter for 30 seconds. Two trials were performed in each of the four postures, one trial with eyes open and no mask, the other with eyes closed wearing the mask. No feedback was given to the subjects regarding their performance during eyes open condition. Feedback was given to the subject during eyes closed condition only until the subject achieved placement of the laser dot in the target circle. Once this was achieved, no further feedback was provided. The laser dot positions (x, y) during each trial were determined using video analysis. The variability in position (standard deviation) of the laser dot from the center of the target was determined as the measure of accuracy during this task and represented a proxy for head sway.

Data analysis

For head repositioning accuracy, the laser dot coordinates in x, y dimensions were determined for both the initial starting head neutral position and for the return head position following head rotation right or left. Each x and y dimension of the coordinate pair was given positive or negative values according to its position relative to the calibrated projection screen axes. Using the two coordinate pairs of values, the distance (hypotenuse) between the laser dots of the initial starting position and the relocated position was calculated using the formula of Pythagoras.[25] Negative signs were removed by calculating the root mean square values. We then converted the hypotenuse data into an angle in degrees (Ø) representing repositioning accuracy. To do so, we divided the hypotenuse distance (described above) by the distance of the screen to participant (2.6 m) in order to calculate the arctangent. This provided the angle of the person's head relative to the error position in radians. We then converted radians to degrees. This hypotenuse distance O–R (Figure 1) represents the global error of positioning converted into degrees and serves as the outcome of HRA in this study. The global error of positioning was recorded in centimeters from the target and was converted into degrees by the following formula: Angle θ (degrees) = 57.3 * $\tan^{-1}$ [global error component (cm)/distance to screen (260 cm)], where 57.3 is the conversion factor from radians to degrees. For every subject, the mean angle (degrees) of the five repetitions in each direction of head rotation for each postural condition served as the values that were entered into the statistical analysis. One subject's data was inadvertently not recorded for the knee and seated conditions. To eliminate cells with no data in the analysis, the mean of the other 11 participants' data served as the data point for each of these conditions.

For the head still task, thirty second trials were transformed to 20 second trials by removing the first 5 seconds and the last five seconds of data collection. With a data

collection rate of 30 frames per second this yielded 600 data points of analysis per trial. Out of the 96 head movement variability trials (8 trials per person) in this experiment, three trials (each by different participants) were not recorded or incompletely recorded by the video camera. To eliminate cells with no data in the analysis, the mean of the other 11 participants' data served as the data point for each of these three error trials. The standard deviation of the mediolateral (ML) (x) and vertical (y) laser dot positions generated by head movements for each 20 second trial was calculated and formed the basic unit of head sway analysis.[22] The mean standard deviations for ML and vertical laser dot positions were computed for each experimental condition.

Statistical analyses for head repositioning task accuracy were performed using separate one-way, within-subjects ANOVA's after right and left head rotation respectively. For the head still task, the statistical analyses for head movement variability in mediolateral (ML) (x) and vertical (y) axes were conducted with separate 2x4 factor, repeated measures ANOVA's with the two factors being visual condition (eyes closed, eyes open) and posture (kneeling, seated, standing, seated stabilized). Post hoc tests were performed using Bonferroni pairwise comparisons. All analyses were conducted using IBM SPSS Statistics for Windows, Version 21.0. Statistical significance was set at an alpha value of 0.05.

Results

Head repositioning task

Separate one-way, within-subjects ANOVA's were run for repositioning after right and left head rotation respectively. Mauchly's test of sphericity indicated that the assumption of sphericity was met for repositioning with left head rotation, $\chi^2(5) = 3.581$, $p = 0.613$ as well as right head rotation, $\chi^2(5) = 6.135$, $p = 0.295$. With left head rotation, there was no significant effect of posture on head repositioning accuracy, $F(3,33) = 1.024$, $p = 0.395$. Similarly, after right head rotation, there was no significant effect of posture on head repositioning accuracy, $F(3,33) = 1.943$, $p = 0.142$. See Table 1 for descriptive statistics of head repositioning accuracy (error) following left and right head rotation.

Head still task

ML (x) Head Movements

Mauchly's test of sphericity indicated that the assumption of sphericity was met for x-axis variability in terms of the main effect of posture, $\chi^2(5) = 5.818$, $p = 0.326$ as well as the interaction between posture and visual condition, $\chi^2(5) = 2.714$, $p = 0.863$. There was a significant main effect of posture on ML position (x) variability defined as the mean standard deviation, $F(3,33)= 4.63$, $p =0.008$,

Table 1.

Descriptive statistics for head repositioning accuracy in degrees after left and right head rotation. MHRA = Mean head repositioning accuracy (error) in degrees based upon the global error of positioning, distance O–R (Figure 1).

	Left Head Rotation		Right Head Rotation	
Posture	**MHRA (∅)**	**Standard Deviation**	**MHRA (∅)**	**Standard Deviation**
Knees	2.602	0.752	3.200	1.267
Seated	3.138	1.686	3.100	0.786
Seated Stabilized	2.994	1.205	2.702	0.692
Standing	3.291	1.132	3.312	1.125

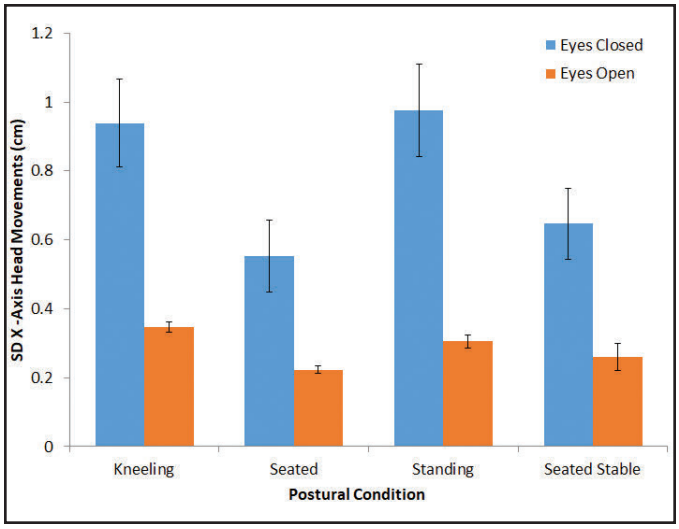

Figure 2.
Mean standard deviation (±SE) of head movements in the x axis for head still task.

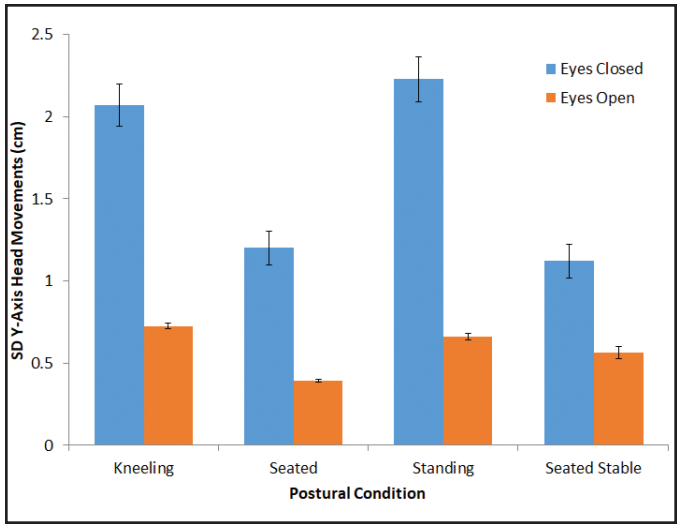

Figure 3.
Mean standard deviation (±SE) of head movements in the y axis for head still task.

$\eta_p^2 = 0.296$ (Figure 2). In general, kneeling and standing conditions generated more ML head movement than seated and supported conditions, however Bonferroni post hoc analyses failed to show any significant differences in pairwise posture comparisons. There also was a significant main effect of vision on ML (x) variability, $F(1,11)= 97.63$, $p < 0.001$, $\eta_p^2 = 0.899$. ML head movement variability was significantly greater in the eyes closed condition (M=0.778 cm, SE=.056 cm) than in the eyes open condition (M=0.284 cm, SE=0.0095 cm). There was no significant interaction between posture and eye condition for ML head movement variability.

Vertical (y) Head Movements

Mauchly's test of sphericity indicated that the assumption of sphericity was met for y-axis variability in terms of the main effect of posture, $\chi^2(5) = 2.265$, $p = 0.812$, but it was not met for the interaction term between posture and visual condition, $\chi^2(5) = 12.910$, $p = 0.025$ so we used the Greenhouse-Geisser correction for the interaction. There was a significant main effect of posture on vertical position (y) variability as defined as mean standard deviation, $F(3,33)= 8.688$, $p < 0.001$, $\eta_p^2 = 0.441$. Post hoc analyses of pairwise comparisons showed kneeling exhibited significantly greater y position variability than seated (p=.030) and supported (p=.012) conditions respectively. In addition, standing exhibited significantly greater y position variability than seated (p=0.050) and supported (p=.014) conditions respectively. There was also a significant main effect of vision on vertical (y) variability, $F(1,11)= 55.13$, $p = 0.000$, $\eta_p^2 = 0.834$. Vertical head movement variability was significantly greater in the eyes closed condition (M=1.65 cm, SE=.15 cm) than in the eyes open condition (M=0.587 cm, SE=0.045 cm). There was a significant interaction between posture and eye condition for vertical (y) head movement variability, $F(2.134, 23.469)= 4.883$, $p = 0.015$, $\eta_p^2 = 0.307$. Post hoc pairwise comparisons for the interaction revealed that for all four postures, the eyes closed condition led to significantly greater y variability than for eyes open ($p<.05$). Figure 3 indicates the greatest differences between eyes open and eyes closed conditions appeared for kneeling and standing.

Discussion

The main findings of this study were that body posture did not influence head repositioning accuracy (either with head rotation to the right or to the left) but did influence head stabilization (head still) performance. There was a significant effect of both body posture and vision on both horizontal (x) and vertical (y) head movements during the head stabilization task with the two seated conditions yielding the least amount of head movement.

The connections between vestibular, visual and proprioceptive afferent information are responsible for postural stability and accuracy of orientation of the head as well as visual balance.[32, 33] Knowledge of higher-order proprioceptive circuitry lags far behind other sensory systems, such as vision, audition, and olfaction.[34] A possible reason for this discrepancy is that proprioceptive sensing is distributed throughout the body, and therefore lacks a single central organ such as an eye or nose.[34] In this light, Roll and Roll[35] suggested that muscle-spindle inputs might form a continuous "proprioceptive chain" from the feet to the eyes. We sought to manipulate this chain by altering posture and thus, the mechanical and proprioceptive degrees of freedom during two different tasks, head repositioning and trying to keep the head still.

We hypothesized that joint position error during the head repositioning task would be reduced when segments other than the neck could contribute to the perception of the orientation of the head in space. We failed to find evidence to support this hypothesis. Rather, it appears that the participants were equally successful at repositioning their heads in space regardless of the number of joints allowed to contribute towards their performance. We take this to mean that for our healthy participants' head repositioning task to be effective, only the cervical spine need be operational. Adding further degrees of segmental freedom is not required, nor helpful in the task of head repositioning accuracy. Our results call into question the hypothesis of a proprioceptive chain – at least with respect to this task and in a healthy population.

Muscle spindle density is very high in the deep muscles of the human neck compared to elsewhere.[36, 37] This high cervical muscle spindle density along with the limited lever action of these muscles suggest that the deep neck muscles allow not only great precision of movement but also adequate proprioceptive information needed both for control of head position and movements and for eye/head movement coordination.[37] For comparison, the deep neck (suboccipital) muscles have almost five times higher the spindle content of the splenius capitis, three times that of the semispinalis capitis, 30 times that of the gluteus maximus and medius, and 75 times that of the gastrocnemius.[38] These histological findings and our behavioral findings suggest there may not have been additional benefit of proprioceptive input from any region other than the neck towards accomplishment of the repositioning task. It may also be possible that healthy participants do not tap into these additional proprioceptive resources.

Clinically, any deterioration of the afferent information received by the senses might lead to inadequate spatial representation, postural alterations and functional impairment of daily activities. Impaired cervical spine proprioception has been demonstrated following fatigue, and pathology such as whiplash and neck pain.[32] In addition, these factors along with neck muscle vibration have also been shown to alter postural stability.[39] Previous evidence has documented that the head repositioning test is a simple and effective method to analyze cervical proprioceptive deficiencies or alterations.[32] In this manner, Dugailly *et al*.[32] studied 36 healthy subjects and 35 chronic neck pain patients (age 30–55 years). Subjects performed the head repositioning test at two different speeds and at two different distances. For each condition, six consecutive trials were sampled. Dugailly *et al*.[32] reported horizontal, vertical and global errors of positioning whereas our current study reports global errors of positioning. Normative values by Dugailly *et al*.[32] of the head repositioning error of 3.3° and 5.4° were identified for asymptomatic and symptomatic subjects (neck pain), respectively. Our subjects fall well within the normative values of the head repositioning error of 3.3° for asymptomatic people. Based upon our results, clinicians can be confident that performing head repositioning accuracy tasks in a variety of postures will not change their results for asymptomatic young subjects. Since many symptomatic subjects have decreased head repositioning accuracy, we suggest repeating our present study with these populations (e.g., whiplash, neck pain) to determine whether they might benefit from greater degrees of postural and proprioceptive degrees of freedom. Posture may be a factor that could bring about improvement in symptomatic patients' cervical spine proprioception in addition to mental training[40], neck muscle vibration[39], exercise and rehabilitation[41], and manual therapy including cervical spine manipulation[31, 41]. Interestingly, spinal manipulation may lead to normalization of afferent input and restoration of appropriate sensorimotor integration and spinal function which has shown improvement in both spinal and limb proprioception.[42]

Regarding the head still task, posture significantly affected performance of the task, but not in the way we hypothesized. We predicted that error during the head still

task would be minimized when joints other than the neck can contribute to the perception of the orientation of our head in space. Rather, the seated position generated the least movement variability as measured by standard deviation compared to the other postures. Standing postures generated the most variability in both x and y directions during the eyes closed condition, while kneeling yielded the most variable movement with eyes open, although there was no significant difference observed between standing and kneeling postures. It is also interesting to note that there was no significant difference between sitting with support (being strapped to the chair) and sitting without support. Perhaps a reason why posture significantly affected the head still task is that trying to fixate on a dot has been found to be more cognitively demanding than other tasks, both in relaxed and steady stances.[43] Such a stationary-gaze task is constraining and may lead to higher cognitive workload and higher attention. It is noteworthy that this contrasts to research on an external focus of attention (e.g., focus on the intended movement effect) versus internal focus of attention (e.g., focus on body movements). An external focus of attention would predict better movement effectiveness, efficiency and automaticity (e.g., less cognitive load) than an internal focus.[44] The stationary task also led to significantly larger interindividual postural sway variability.[43] The larger postural sway variability is supported in theory by the idea of "hypercontrol" during stance which could create postural inefficiency.[21]

In general, our results from the head still task seem to indicate that more degrees of freedom led to increased movement variability, not less as predicted and this effect was magnified in the absence of vision. It is entirely possible that factors other than proprioception contributed to the magnitude and variability of sway during this task. Such factors might include the mechanical nature of sway. Given the inverted pendulum nature of human stance, torques generated at the ankles and knees could lead to larger amplitude deviations of the head accounting for the greater sway variability compared to the seated postures. During seated postures, head sway might be minimized mechanically simply because there is less movement of distal (inferior) structures.

There are several limitations and/or ways to improve this study. Firstly, we used five trials for each direction of the head repositioning task. Authors have found stable estimates of performance were obtained when data from six or more trials was included although, the greatest test-retest reliability was obtained with five or more trials for joint position error.[26] It is possible that more trials could have improved performance estimates, although a practice and/or (attentional or physical) fatigue effect might be at play with further repeated trials. It is also a possibility that even with 5 trials for each direction and for each position that we tested that subjects may have had some degree of muscle fatigue. Second, the number of postures in this study could have been expanded, for example to include standing but restricting ankle movement such as with a boot – this would have the effect of allowing knee and hip joint contribution to stance. Another consideration for future studies would be to measure actual head motion as opposed to laser dot movement. Although this might prove less feasible given current technological constraints, it would mitigate any contribution of laser dot measurement error as a proxy for head sway. Finally, an a priori sample size estimate could have reduced the risk of an underpowered result.

Conclusion

For healthy young adults, clinicians and researchers need to be concerned with postural influences on tasks that involve head/cervical spine stabilization. Contrary to our hypothesis, we provide evidence that during the head still task, more joint degrees of freedom led to increased movement variability of the head. Clinicians can be confident that performing head repositioning accuracy tasks in a variety of postures will likely not change their results for healthy, young asymptomatic subjects. We suggest repeating our present study with both neck pain and whiplash populations to determine whether their head repositioning accuracy might benefit from greater degrees of postural and proprioceptive degrees of freedom.

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Taping protocol for two presentations of pregnancy-related back pain: a case series

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Background: *Back pain is common during pregnancy and can have an adverse impact on the quality of life for some, yet treatment options for this population are limited. We document a chiropractic treatment that involves using kinesiology tape (tape) to help alleviate pregnancy-related back pain in two patients.*

Case Presentation and Management: *Two pregnant women reported to two different chiropractic offices with varying presentations of pregnancy-related back pain. A trial of chiropractic care was rendered in both chiropractic offices, which included the application of tape.*

Outcome and Discussion: *In both case presentations, the addition of tape in the lumbosacral and/or abdominal regions, decreased pain intensity from 9-10/10 to 4/10 or less on the Numeric Rating Scale (NRS). Including a taping protocol to a plan of management in women with pregnancy-related LBP or*

Contexte : *Les maux de dos sont fréquents pendant la grossesse et peuvent avoir un impact négatif sur la qualité de vie de certaines femmes, mais les options de traitement pour cette population sont limitées. Nous documentons un traitement chiropratique qui implique l'utilisation de ruban de kinésiologie (ruban) pour aider à soulager les douleurs dorsales liées à la grossesse chez deux patientes.*

Présentation et gestion de cas *: Deux femmes enceintes se sont présentées à deux cabinets de chiropractie différents présentant des maux de dos différents liés à la grossesse. Un essai de soins chiropratiques comprenant l'application de ruban a été effectué dans les deux cabinets de chiropractie.*

Résultats et discussion : *Dans les deux présentations de cas, l'ajout de ruban dans les régions lombo-sacrées et/ou abdominales a diminué l'intensité de la douleur qui est passée de 9-10/10 à 4/10 ou moins sur l'échelle d'évaluation numérique (EEN).* Le fait d'*inclure un protocole d'utilisation de ruban à un plan de prise en*

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The involved patients provided consent for case publication.

PGP may be a safe and effective option to alleviate pain in this population.

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KEY WORDS: pregnancy, back pain, chiropractic, kinesiology tape, case series

charge chez les femmes présentant une lombalgie ou des douleurs pelviennes liées à la grossesse peut constituer une option sûre et efficace pour soulager la douleur chez cette population.

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MOTS CLÉS : grossesse, maux de dos, chiropratique, ruban de kinésiologie, série de cas.

Introduction

The literature has reported that up to 90% of pregnant women experience some sort of back pain.[1-5] This large prevalence can be accounted for by the fact that back pain experienced during pregnancy is not just one entity. It can be further separated into (1) pregnancy-related low back pain (LBP), from the bottom of the ribs to the inferior gluteal folds[6], characterized as dull and resembling back pain that occurs in the non-pregnant state[7]; (2) pelvic girdle pain (PGP), from the top of the iliac crests to the bottom of the gluteal folds and/or pain in the symphysis pubis, often described as stabbing, shooting, dull, burning or an occasional "catching" sensation in the leg while walking[7]. The pain can occur in either one or both sacroiliac joints (SIJs), in conjunction with/or separate symphysis pubis pain, the pain may radiate into the thigh[7]; or (3) a combination of both pains (combined pain)[8-10]. Clinical features of pregnancy-related LBP and PGP can start as early as the first trimester, but more often, it starts around the 18th week of pregnancy, with a peak intensity between the 24th and 36th week. It often resolves within a few weeks to a few months following delivery.[7]

Previous literature has suggested the importance of distinguishing between pregnancy-related LBP and/or PGP for proper management and prognosis of the two conditions.[7, 11-13] Useful methods of differentiation include the site of pain, its character and severity, provoking factors, resultant disability, and pain provocation tests.[7, 12] A pregnant woman experiencing LBP will likely present with pain during forward lumbar flexion, pain and/or tenderness with palpation of the lumbar erector spinae muscles, and restriction in the lumbar spine. A woman presenting with PGP will likely experience pain with pain provocation tests such as posterior pelvic pain provocation (P4), FABER (flexion, abduction, and external rotation), and palpation of the dorsal sacral ligament. Specifically, to diagnose symphysis pubis pain, direct palpation of the symphysis pubis, modified Trendelenburg's test and active straight leg raise will be painful.[7, 11, 12]

Pregnant women seek out pain relief methods such as pain medication, exercise, education, pelvic support belts, and chiropractic treatments, however, there is limited evidence with regards to their efficacy.[14] In an effort to relieve pain from musculoskeletal injuries, an emerging form of treatment among manual therapists, such as chiropractors, is the use of kinesiology tape, or "tape" as we refer to it in these cases. Taping has been suggested as a treatment for those with musculoskeletal injuries as it has a number of proposed benefits. These benefits include: supporting the affected area, relaxing the muscles and reducing pain sensation[10,15,16]; is designed to replicate the physical properties and characteristics of human skin[15,17]; and it has been suggested in the literature that it helps in the normalization of muscular function, increase lymphatic and vascular flow, reduce pain through neurological suppression, and possibly contribute to corrections of joint misalignments[15,16,18]. Although the full action of tape has yet to be elucidated[16], the effectiveness reported in various clinical conditions has shown both positive[16, 18-21], and negative results[22-24] in the non-pregnant population.

There are currently a few studies regarding taping and pregnancy-related back pain which have demonstrated an improvement in pain and disability in these women.[10,25, 26] In the pregnant population, Kuciel *et al*.[25] and Kalinowski *et al*.[26] applied tape to participants experiencing pregnancy-related PGP and LBP, respectively. Both research groups found a significant reduction in pain two to five days after tape application.[25, 26] Although outcomes in

the pregnancy studies show favourable results, the methodology of both studies exhibit weaknesses. One of the strengths of the Kuciel *et al*. study is that they performed a physical examination on participants to determine if their subjects were experiencing PGP[25]; however, they did not utilize a control group for comparison. Additionally, the study employed a general taping protocol, extending tape from below the posterior superior iliac spine into the scapular region, with an additional horizontal strip over the sacroiliac region.[25] Whereas, Kalinowski *et al*. used a more specific taping protocol, no physical examination was conducted to determine the type of back pain the pregnant women were experiencing.[26] The purpose of this case series is two-fold: (1) to identify the type of pregnancy-related back pain and (2) to demonstrate that an individualized taping protocol applied in women presenting with different pregnancy-related back pains may help to reduce pain levels over the course of their pregnancy.

Case Presentation #1

A 37-year-old female, six-weeks pregnant, presented to her chiropractor with complaints of PGP, localized to the left SIJ, and pregnancy-related nausea. One treatment of manual therapy was performed at this time, including soft tissue therapy (STT) and spinal manipulation (SMT) to the left SIJ. At 16 weeks gestation, the patient reported that she began experiencing round ligament pain and discomfort with associated symphysis pubis dysfunction. However, the patient did not return to the chiropractor until 20 weeks gestation because she was considered high-risk and was undergoing genetic testing.

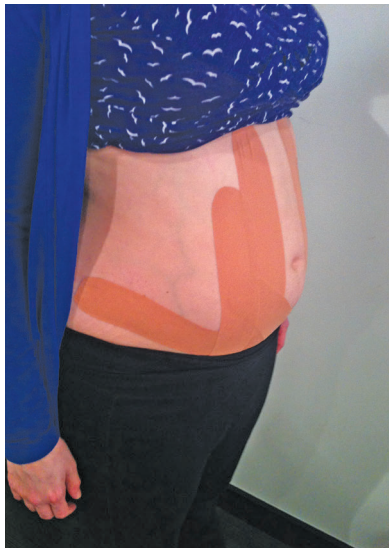
20-27 Weeks Gestation

At 20 weeks gestation, the patient returned to the chiropractor with exacerbated left SIJ and right round ligament pain. At this time, she reported that she was not sleeping well, was unable to perform her activities of daily living (ADLs) (i.e. walking for more than five minutes), and her pain was 9/10 on the Numeric Rating Scale (NRS). The chiropractor reassessed the patient. Physical examination revealed lumbar range of motion was limited in all directions, with bilateral rotation and right Kemp's test recreating the round ligament pain, and flexion and extension causing the left SIJ pain. Bilateral straight leg raise, left FABER, left SIJ compression, left P4, left Yeoman's and left Ely's (modified for pregnancy) tests all aggravated the left SIJ pain. Right FABER and right Yeoman's test elicited the right round ligament pain. Palpation revealed tenderness bilaterally in the lumbar erector spinae, quadratus lumborum, and piriformis (worse on left), left dorsal sacral ligament and right round ligament. Additionally, joint palpation of L3-5 and the left SIJ were painful. The patient was diagnosed with combined pain. Plan of management included STT to the aforementioned musculature, SMT to the involved lumbar segments using Diversified technique and the left SIJ using a table drop piece, taping of the abdomen, birth preparation exercises and advice to hold/support abdomen during transitions. Immediately following the treatment of STT and SMT, the patient reported her pain to be unchanged on the NRS (9/10).

Figure 1. *Front view of patient's abdominal taping (rectus abdominus and external abdominal oblique).*

Taping protocol: At this point the chiropractor applied tape to the abdomen using four strips: two along rectus abdominus and two along external abdominal oblique. The first two strips were started at the superior aspect of the symphysis pubis with no-tape tension for one-inch. Tape-off tension was applied as the tape coursed over the rectus abdominus muscle up to the xiphoid process, with no-tape tension for the last one-inch. The remaining two strips were started on the medial aspect of the symphysis pubis (overlapping rectus abdominus tape medially) coursing along external obliques and anterior superior iliac spine (ASIS) with tape-off tension; no-tape tension was applied to the starting and ending one-inch of tape, acting as anchors (Figure 1). Home care included Kegel

(a)



(b)

Figure 2.
(a) Front and (b) side view of patient's reinforced abdominal taping (rectus abdominus reinforced, external abdominal oblique, symphysis pubis 'X').

Figure 3.
Front view of patient's abdominal and symphysis pubis taping (rectus abdominus, external abdominal oblique, symphysis pubis 'X').

exercises and abdominal bracing. The patient was also receiving care from a naturopath, registered massage therapist and a midwife.

One week later, at 21 weeks, the patient returned to her chiropractor feeling much better; she rated her pain at a 4/10 on the NRS. The patient reported that her combined pain and round ligament pain improved, as well as her sleep. She was able to walk for 10-15-minute intervals with minimal to no pain. The chiropractor performed the same manual therapy intervention as her previous appointment and re-applied tape.

At 23 weeks, the patient reported overall improvement in symptoms, but continued to experience mild combined pain and round ligament pain, as well as interrupted sleep. The chiropractor treated her with the same manual treatments and taping as outlined above. Three days later, the patient returned to the chiropractor stating that she was able to go for a long walk the day before, and that she was experiencing significant relief. However, the patient reported that without the tape, pain returned to 9/10. The tape was re-applied in the same manner outlined above; however, four additional strips were applied for further support. Two strips were used to reinforce the rectus abdominus strips and two creating an "X" over the symphysis pubis (Figure 2). The patient's regular manual therapy was provided at this visit. For convenience, and to ensure continued relief for the patient, the chiropractor taught the patient's husband the taping protocol for home application.

28 Weeks Gestation to Delivery

At 28 weeks, the patient reported the tape continued to provide relief with the round ligament pain as well as combined pain. Unfortunately, the patient started to experience symphysis pubis pain and difficulty walking. At all chiropractic appointments from 28 weeks until delivery, manual therapy was performed, exercises were progressed, and the tape was applied by her husband at home. At 37 weeks, the chiropractor added isometric contraction of the hip adductors to adjust and relieve pain in the symphysis pubis and applied additional tape over the symphysis pubis (Figure 3). At 38 weeks, the patient experienced Braxton Hicks contractions almost immediately following the treatment. At this point, the chiroprac-

tor discontinued SMT because the patient expressed concern. For the remainder of the visits, and with the patient's consent, the chiropractor performed instrument assisted adjustments and STT. Treatment continued until just after 40 weeks. At this time, the patient had a successful labour and delivery and continued to seek chiropractic care postpartum. There were no adverse events reported with any of the treatment provided including the manipulation, STT or the taping intervention. Signed consent for release of patient information and photographs was obtained from the patient.

Case Presentation #2

A 32-year old patient presented to the clinic at 35 weeks gestation with back pain of approximately two-month duration. When asked to point to the area of pain, the patient indicated that her pain was bilaterally at the SIJs. She also reported pain along her anterior thigh and posterior hamstring, not extending distal to the knee. She reported the pain intensity was a 10/10 on an NRS. She denied any numbness or tingling into the lower limb and no change in bowel or bladder control. Although her pregnancy was healthy and uneventful, she was having difficulty with ADLs including, turning over in bed and fast cadence gait as she reported experiencing a "zapping feeling down her leg". The patient was active during her pregnancy, owning a Pilates studio where she continued to teach until 37 weeks gestation.

Physical examination revealed that lumbar range of motion was full and pain-free. A P4 test elicited the chief complaint, with the right-side more painful than left. Palpation of the SIJs and dorsal sacral ligaments were painful bilaterally, as was palpation of the right piriformis. Diastasis rectus testing was negative, as was greater trochanteric compression. A lower extremity neurologic examination, including deep tendon reflexes, sensory (light touch) and motor testing and plantar reflex, were all within normal limits. A diagnosis of pregnancy-related PGP was rendered for this patient. Plan of management included STT of the piriformis, dorsal sacral ligaments, gluteus minimus and medius, taping of the SIJs, and birth preparation exercises.

Taping protocol: Tape application consisted of three strips, creating an H-pattern. The first two strips were parallel to each other and started on the lumbar erector spinae running longitudinally until past the SIJs. Tape was applied for one-inch with no-tape tension. Tape-off tension was applied as the tape was applied along the muscle belly of the lumbar erector spinae to below the SIJs, with no-tape tension for the last one-inch. The third strip was applied over the SIJs, starting at the sacral tubercle. Tape-off tension was applied medial to lateral with no-tape tension at the one-inch ends (Figure 4).

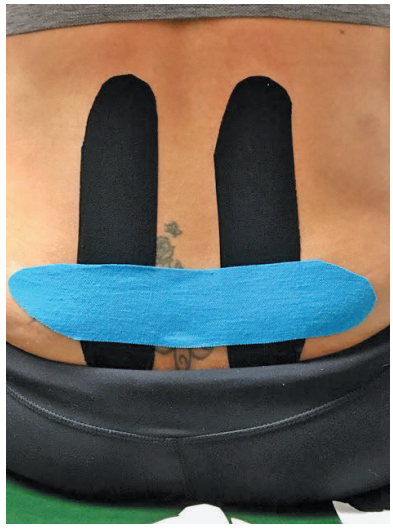


Figure 4.
Posterior view of patient's low back and pelvis (lumbar erector spinae and SIJs creating H-pattern).

Following the first treatment, the patient emailed the chiropractor reporting her pain was non-existent (0/10), she was very pleased with her treatment response. Until she gave birth at 39.5 weeks to a healthy baby, the patient received three more treatments once a week, with the same protocol and same relief. Although the patient has attempted returning for postpartum chiropractic care, she has been unsuccessful with a newborn. There were no adverse events reported with the any of the chiropractic care including the STT and taping intervention. Signed consent for release of patient information and photographs was obtained from the patient.

Discussion

It is well known that pregnancy is often associated with the onset or exacerbation of LBP, PGP or a combination of both of these pains.[6] Despite this, there is limited evidence to the etiology, pathophysiology and treatment strategies for this population.[6,27] As such, healthcare providers often dismiss complaints by the expectant mother because these are considered normal ailments of pregnancy and often resolve once the baby has arrived.[27] Therefore, the im-

portance of differentiating pregnancy-related LBP and PGP for proper diagnosis and management is crucial.[7, 11-13] Both chiropractors in the current case series conducted a thorough history to obtain details of the chief complaint and the ADLs impacted by the patient's back pain. Furthermore, a physical examination using two or more pain provocation tests, was performed in both cases to identify the type of back pain each patient was experiencing. Once a proper diagnosis was identified, taping protocols specific to the patient's symptomatology was rendered.

It has been postulated that the positive results associated with taping may be a result of direct and indirect means.[25] Directly, the tape affects the skin and associated receptors (Ruffinis, pain and deep sensory receptors) and indirectly, through supporting lumbosacral structures (such as thoracolumbar fascia). These mechanisms may help reduce tension in overloaded muscles, thereby increasing stability, reducing paraspinal stress, and stimulating connective tissues, resulting in pain relief.[10,25] Following the application of tape, patients in the current study reported a decrease in pain and qualitative improvement in function and disability. Furthermore, they had less fear of movement with ADLs, which has been noted in previous literature.[19] In the current case series, both patients complained that certain movements were extremely difficult, including changing positions during sleep and an inability to walk quickly and/or longer than five minutes. One patient felt relief immediately, and the other felt relief within one week following the application of tape. Pain scales were originally 9-10/10 and reduced to 4/10 or less. Previous studies have demonstrated that the application of tape increases patients' sense of confidence and safety. It has been suggested that the tape provides a more secure and stable feeling, as well as a proprioceptive reminder to maintain optimal posture.[28] This increased confidence and stability may allow the patient to actively perform exercises, contributing to the efficacy of their treatment.[5]

Previous pregnancy taping studies have found similar results to the current case series. Kaplan *et al*. examined the effectiveness of a five-day intervention that included paracetamol and tape versus paracetamol alone on pain and disability.[10] They determined the addition of tape had more superior effect on the outcomes than paracetamol alone. However, they did not employ a sham group in their study; therefore, a placebo effect cannot be ruled out.[10] Sabbour and Omar randomly assigned pregnant women who were experiencing LBP to receive pelvic tilt exercises or pelvic tilt exercises plus the application of tape (two applications, approximately one week apart).[5] They demonstrated a significant decrease in pain and functional disability scores in the group who received exercise and tape.[5] Kuciel and colleagues also found a reduction in pain and improved disability scores on day five and day ten following a five-day taping protocol.[25] In a crossover study, Kalinowski *et al*. applied tape to women experiencing low back pain and determined that there was a decrease in pain intensity and disability when women had the taping protocol applied, as opposed to the placebo taping protocol.[26] It should be noted that although these can be considered positive results, Kuciel *et al*.[25] and Kalinowski *et al*.[26] did not use any type of control group to compare outcomes.

One difference between the current case series and what is reported in the literature, is that our study followed patients on a long-term basis with multiple tape applications versus a one-time application with shorter follow-up periods.[25, 26] In the present case series, both patients noted their pain returned immediately following the removal of tape and decreased with their next application. In addition, our case series identified different types of pregnancy-related back pain prior to applying an individualized taping protocol based on their symptomology. Kuciel *et al*. [25] utilized the European Guidelines for Treatment and Diagnosis[11] to identify PGP in their subjects, and then employed a generalized taping protocol - tape applied from the bottom of the scapula to the bottom of the sacroiliac joints. Similar to the present case series, Kalinowski *et al*.[26] and Sabbour and Omar[5] applied a specific taping protocol; however, unlike the current case series, they did not identify the type of LBP their patients were experiencing. In the current case series, the patient in the first case experienced combined pain with symptoms most pronounced anteriorly, including round ligament and symphysis pubis pain. Whereas, the patient in the second case presented with PGP, specifically involving bilateral SIJs. Therefore, with differing clinical presentations, examination findings and pregnancy-related back pain, the treating chiropractors applied taping protocols to best support the affected areas; Case One, anteriorly along the patient's abdomen and Case Two, posteriorly along the patient's SIJs. Although the results of this case

series are limited, positive outcomes on the quality of life of the affected pregnant patient were achieved.

Tape is another treatment modality available to chiropractors that can be used as a relief mechanism for pregnant patients. It is a simple technique that can be applied to the patient and can be taught to their partner to apply as a form of pain relief. It is noninvasive, inexpensive, and is well tolerated by the patient, with very few adverse effects.[10,25] This case series demonstrates the effects of individualized taping protocols on pregnancy-related back pains. As tape is easy to apply and relatively inexpensive, we believe it may be an effective option for pain management during pregnancy.

Limitations and future studies

This case series has several limitations. First, one of the patients had a difficult pregnancy with multiple complications. Due to the complicated nature of her pregnancy, she was seeking care from many different healthcare providers and therefore, her pain relief cannot be solely attributed to taping and chiropractic care. Second, both patients presented with different symptomatology and therefore, received slightly varying therapies from their respective chiropractor, including soft tissue therapy and manual adjustments. Consequently, the benefits seen in this case may not be exclusively related to taping. Furthermore, the methodology differed with respect to taping pregnant patients; both chiropractors used different types of tape and neither chiropractor had any formal training with respect to taping pregnant patients. Despite these limitations, there were still positive clinical results. Third, only qualitative measures were obtained in these case reports, including NRS and perceived quality of life. Future studies would benefit from the inclusion of validated outcome measures to assess disability (i.e. Oswestry Disability Index, Roland Morris Disability Questionnaire) or health-related quality of life (i.e. SF-36). Finally, we did not use a sham taping application; therefore, the placebo effect of the taping treatment cannot be discounted.

Our case report highlights the important need for future research. Future studies should focus on the application of an individualized taping protocol that is appropriate for the varying types of pregnancy-related back pain.

Summary

There is limited research on the effectiveness of taping on LBP and PGP during pregnancy and as far as we know, this is the first case series to follow two pregnant women diagnosed and treated with different pain patterns and taping protocols from presentation in clinic until delivery. Upon each application of tape, the patients experienced significant and immediate pain relief as well as increased quality of life. More research is required to differentiate the types of back pain pregnant patients experience (LBP, PGP, combined pain), and furthermore, if varying types of taping protocols are indicated.

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An approach to interprofessional management of complex patients: a case report

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Introduction: *Complex patients are frequently high-users of health care resources. Case management has been demonstrated to be an effective and efficient approach for this demographic.*

Case Presentation: *A 36-year old, medically complex male patient was referred to an interprofessional primary care team to optimize health status. Team involvement included a case manager, nurse practitioner, pharmacist, social worker, team assistant and chiropractor. Interventions involved medication*

Introduction : *Les patients ayant des besoins complexes sont souvent ceux qui utilisent le plus les ressources en soins de santé. La gestion de cas s'est avérée être une approche efficace et efficiente pour ce groupe de personnes.*

Présentation de cas : *Un patient de 36 ans présentant des problèmes de santé complexes a été dirigé vers une équipe interprofessionnelle de soins primaires afin d'optimiser son état de santé. L'équipe comprenait un gestionnaire de cas, un infirmier praticien, un pharmacien, un travailleur social, un assistant d'équipe et un chiropraticien. Les interventions portaient sur la gestion des médicaments, l'abandon du tabac, les*

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management, smoking cessation, mindfulness skills and musculoskeletal treatment.

Summary: *Complex patients are increasingly managed by teams. To continue, these teams will have to demonstrate positive outcomes and cost-effectiveness. Chiropractors have skills that can enhance team-based patient care.*

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KEY WORDS: complex patients, case management, primary care, chiropractic, interprofessional

compétences liées à la pleine conscience et le traitement musculosquelettique.

Résumé : *Les patients ayant des besoins complexes sont de plus en plus pris en charge par des équipes. Pour continuer à exercer, ces équipes devront démontrer des résultats positifs et un bon rapport coût-efficacité. Les chiropraticiens ont des compétences qui peuvent améliorer les soins aux patients dispensés en équipe.*

(JACC. 2019;63(2):119-125)

MOTS CLÉS : patients ayant des besoins complexes, gestion de cas, soins primaires, chiropratique, interprofessionnel.

Introduction

In Ontario, Canada the top five percent of healthcare system users consume two-thirds of healthcare resources.[1] Starting in 2012, the Ontario Ministry of Health and Long-Term Care established Health Links to more effectively provide care to this challenging population.[1] The KW4 Health Link serves a portion of Waterloo Wellington Region.[2] A participating program in this initiative is the KW4 Community Ward Team (CWT).[2] Members of this team include Local Health Integration Network (LHIN) care coordinators, nurse practitioners, a pharmacist, a social worker, an outreach worker, a team assistant, a team manager and a chiropractor.[2] A consultant physician is available to the team as needed.[2] The CWT supports the patient's primary care provider for a period of time until the patient is stabilized and has supports in place.[1] In some cases, the CWT is a patient's only access to primary care and they maintain a longer-term relationship with the patient.[2] The following case illustrates how the CWT manages a complex patient.

Case presentation

History

This patient was a 36-year old male referred to the CWT by a community-based addictions worker. After CWT LHIN care coordinator/case manager triaging, it was determined the patient fit the CWT referral criteria as he had four or more chronic health conditions.[2] His conditions that qualified him included addictions, hepatic encephalopathy, liver cirrhosis and polypharmacy. The referral source's goal for this patient was more support for physical health concerns. The patient's goals were to abstain from alcohol, improve health and get help with their shoulder pain.

The initial visit was made by the CWT nurse practitioner (NP) and social worker. Past medical history included hepatic failure secondary to alcoholism, hepatic encephalopathy, seizures, portal hypertension, depression, anxiety, chronic pain, shoulder bursitis, multiple sports concussions, smoking, falls (8 in 12 months) and polypharmacy. A systems review showed frequent headaches, dizziness, tinnitus, some exertional shortness of breath, decreased appetite, reported vomiting twice a day and shoulder pain affecting function. Based on this analysis, immediate referrals were to the CWT NP for overall case management and to the CWT pharmacist to do a thorough medication review. As the case progressed, the CWT social worker provided counselling and the CWT chiropractor addressed the patient's pain goals and frequent falls.

Intervention and outcome

The CWT NP's role involves having an overall understanding of the patient's case. This includes visiting patients where they reside, triaging immediate concerns, prioritizing the problem list, reviewing medications, reviewing available bloodwork and relevant imaging and

consulting/referring as necessary. This patient had a meaningful connection with their family physician so the NP's role related to acute episodic or significant chronic disease management was less than in cases of orphan patients or where a family physician meaningful connection had not been established.

Occasionally there are medication discrepancies found between the family physician's medication list, pharmacy's list and what the patient is actually taking. Medication concerns noted in this case were that the patient was nonadherent to certain medications due to side effects and an important medication related to liver dysfunction and encephalopathy was not on the family physician's medication list. Rifaxamin is well documented in the treatment of acute hepatic encephalopathy, is shown to reduce symptoms associated with hepatic encephalopathy (for example confusion) and reduces liver failure hospitalizations.[3] Often, the pharmacy is contacted first regarding a medication concern however in this case the family physician was asked about the status of the Rifaxamin. This was being prescribed by the gastroenterology specialist and therefore was not on the medication list provided by the family physician's office. In that same letter, the family physician was notified that a referral for concussion rehabilitation as well as inquiry into the appropriateness of an ABI (Acquired Brain Injury) program, was underway. Since multiple medication concerns were found, the CWT pharmacist was consulted and a full review was performed.

The CWT clinical pharmacist performed an in-home medication review.[4] A medication review is a complete assessment of all medications a patient is taking, including prescriptions, over-the-counter (OTC), herbal or natural products.[4] The purpose of the review is to improve a patient's health outcomes by ensuring the patient understands their medication regimen.[4] Often times, the role of the clinical pharmacist would be more in-depth and involve medication management. Medication management is a more thorough process where medications are optimized to ensure that they are safe, effective and appropriate.[4] This involves pharmacist collaboration with the patient's physician or NP (or any other health care provider) to assess a patient's medications and health conditions, identify drug therapy problems, create a care plan and follow-up with any recommendations made.

An in-home medication review provides more detailed information. It allows the pharmacist an opportunity to declutter and remove expired drugs, determine if the patient is compliant and if they have a system that is working for them.[4] The clinical medication review has been shown to have positive results for all outcomes and is favoured over other adherence reviews which show no significant effect on hospitalization or mortality. Evidence suggests that advanced clinical medication reviews for hypertension and dyslipidemia are most effective in improving health outcomes.[4] The CWT pharmacist and NP discussed the patient's medications. In this case, the patient was not using his lactulose as prescribed for hepatic encephalopathy and they stated that their physician knew. A letter was faxed to the physician to confirm this finding along with a complete medication list.

It was also identified that this patient was interested in quitting smoking. The CWT pharmacist and social worker collaborated on this objective. Pharmacists in Ontario are able to help with smoking cessation by determining the patient's stage of change, assessing the patient's condition and history of smoking, providing education and prescribing smoking cessation therapy when indicated. The transtheorectical model of behavior change or "stages of change" was used.[5] There are six stages where the patient could reside including precontemplation, contemplation, preparation, action, maintenance and termination.[5] You identify which stage the patient is at and try to progress them through to the next higher stage.[5] This patient was in the contemplative stage prior to pharmacist involvement, and successfully moved to the preparation and then action stages thereafter. He was prescribed nicotine replacement therapy, which was covered by the STOP (Smoking Treatment for Ontario Patients) program. However, after a short trial of the patches and nicotine inhalers, he realized that he did not like them and continued with "the reduce to quit" method.

As the case evolved, the CWT social worker introduced the therapeutic approach of mindfulness to help the patient accept and adapt to the uncontrollable circumstances of a chronic illness. Mindfulness can be defined as "paying attention in a particular way: on purpose, in the present moment and non-judgementally".[6] It is important that they learn to live with peace and purpose despite the limitations imposed by their condition, essentially making peace with the life they now have. Mindfulness tools

were introduced to the patient to help ground them in the present and to combat anxiety about the future.

The concept of Acceptance was integrated to recognize the perfection of each moment, as each moment is caused by all that preceded it, and could not, therefore, be otherwise more perfect than it is.[7,8] Embracing Acceptance on a journey with chronic illness is critical. The Buddhist teacher, Shinzen Young, developed this formula to explain suffering. He said "Suffering = Resistance x Pain, Acceptance is the opposite of Resistance".[9] The more you resist and struggle with your illness, the greater the suffering you experience.

Working alongside team members, the CWT chiropractor's contribution to a case may include mobility and falls assessments of a patient's function and their home environment, attending and advocating for a patient at specialist appointments and at times manual therapy and prescribing specific exercises. In this particular case a chiropractic history and examination occurred. Diagnoses included a right shoulder strain, a lumbar spine strain with distal radiation and a thoracic spine strain. Manual therapy and specific exercises were administered. Techniques avoiding pressure on the liver were utilized as the gastroenterologist identified a risk with any sudden pressure on the diseased liver which could lead to its injury. The outcomes of chiropractic treatment are as follows: the right shoulder strain went from a pain score of 6-9/10 to near complete relief, the lumbar spine strain with distal radiation went from an 8/10 to mild pain and the thoracic spine strain went from a 4-10/10 to 50% improved. The patient was pleased with the results. As this was a male patient, after a few visits the chiropractor attended the patient independently. This would not occur with a female patient for risk management reasons. By the time the CWT chiropractor assessed the patient, the falls had stopped without CWT intervention and were not a concern.[10] This was likely due to a multitude of factors. Previous to team involvement the patient had been hospitalized and had become deconditioned. The patient credited starting to exercise at a gym with stopping the falls.

Throughout this case, the CWT team assistant (TA) fielded calls from the patient, notified the required professionals for their services as needed and facilitated the booking of ongoing visits. The TA provided the necessary co-ordination and made sure nothing slipped through the cracks. The CWT electronic medical record allowed for documentation and secure communication within the team. The CWT also used Clinical Connect which is a secure, web-based portal that provides clinicians with real-time access to their patients' electronic medical information from all acute care hospitals, LHIN Home & Community Care Services and Regional Cancer Programs in South West Ontario, plus various provincial data repositories.

Discussion

Chiropractors are increasingly participating in team-based care.[10] This may involve managing a complex patient who is defined as having multiple comorbidities, being at high risk for poor outcomes and being high cost to the healthcare system.[11] The CWT classifies complex patients as having four or more chronic health conditions (with a focus on mental health and addictions, frail elderly or prematurely frail), polypharmacy (five or more medications) and negatively affected by the social determinants of health.[2] The social determinants of health includes factors such as early childhood development, education, employment, income, social support, housing and gender.[12] The CWT patients require many aspects of the healthcare system, are unwell and have barriers to care including limitations in the social determinants of health (Figure 1).

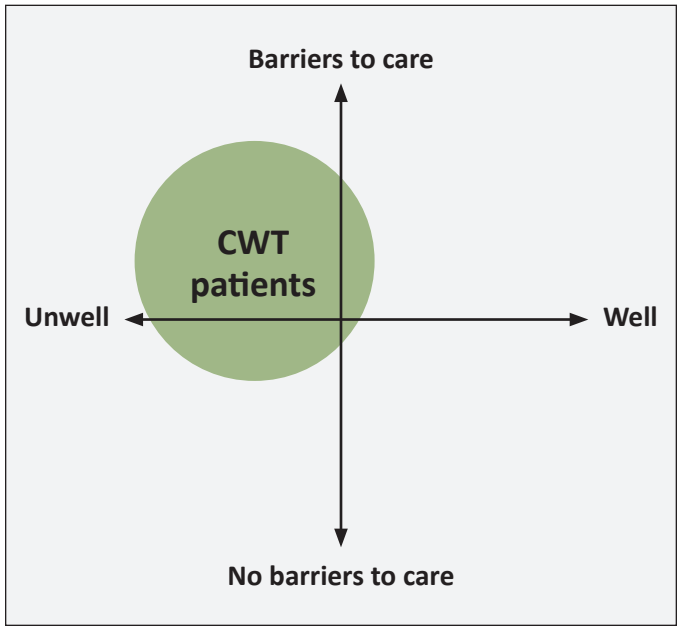

Figure 1.
The Community Ward Team (CWT) patient population.

Table 1.
Acute Care Utilization 3, 6, and 12 months Pre- and Post-Community Ward Team (CWT) Involvement.[2]
ED (Emergency Department), LOS (Length of Stay in hospital), ALC (Alternate Level of Care in hospital), ACSC (Ambulatory Care Sensitive Condition in hospital). Data collected from 2014 and 2015.

3 months pre-post CWT (n=83)	**6 months pre-post CWT (n=79)**	**12 months pre-post CWT (n=78)**
Average age: 65.2	Average age: 64.3	Average age: 64.2
• ED visits decreased by 38.3% • Total inpatient visits decreased slightly by 20.7% • Total inpatient LOS decreased by 35.6% • Average inpatient LOS decreased by 18.8% • Total ALC LOS decreased by 64.6% • Total inpatient readmission remained relatively constant	• ED visits decreased by 31.9% • Total inpatient visits remained relatively unchanged • Total inpatient LOS decreased by 30.3% • Average inpatient LOS decreased by 28.9% • Total ALC LOS decreased by 20.8% • Total inpatient readmissions decreased slightly by 22.7%	• ED visits decreased by 33.3% • Total inpatient visits, total inpatient LOS, and average inpatient LOS remained relatively similar • Total inpatient visits for ACSC increased by 260% • Total ALC LOS increased by 36.6% • Total inpatient readmissions decreased by 33.3%

Anyone within the team's sub-region can refer to the CWT including patients themselves.[2] Where the referral criteria are met, patients are triaged by need and the presence, or not, of other healthcare supports. The CWT is not for crisis management; but rather to support patients and more effectively and efficiently manage them to reduce hospital Emergency Department (ED) utilization and hospital admissions.[13-19] This team has demonstrated a decrease in health care system usage by its rostered patients in a number of key areas (Table 1).[2] There is also literature demonstrating this approach is ineffective in reducing ED utilization.[20-22] Increasingly, the CWT is also working with patients who previously were non-users of healthcare.

The TA first makes contact with the patient and asks a series of questions to determine if the patient's home is a safe environment for team members. This entails inquiring about pets, weapons and bed bugs. As the CWT members meet the patient in their home, much information can be learned from this including whether the patient is at risk of physical danger, has healthy food or hoards. The CWT's initial assessment involves two team members spending around 90 minutes on two occasions with the patient and working through the intake form. This covers multiple domains to holistically assess the health status of the patient. This document identifies where the patient's health is most vulnerable and improvement is potentially possible. Topics assessed include the major organ systems, medications, mental health, mobility, current care team members and Do Not Resuscitate (DNR) status. Medical Assistance In Dying (MAID) on occasion is requested by the patient, indicating how desperate they are.[2] The entire team convenes at weekly case review and this is where patient information is discussed and, as a group, decisions are made as to which team members will be involved going forward. Frequently the team looks for the "low hanging fruit" where a little help can make a big difference. Common approaches formulated during case review are "meeting the patient where they are", "leaning in" and "holding the narrative for the patient" (their story).

The CWT assists the patient in navigation of the healthcare and social support systems to optimize outcomes.[23] The CWT uses a case management approach: a collaborative process of assessment, planning, advocacy and facilitation for services and options to meet an individual's health needs through available resources; and communication to promote quality cost-effective outcomes.[19] In the past a patient's management was dictated by the system. This often fails and so the CWT attempts to partner with patients to support their efforts to achieve their goals and best life. This may be different than the system's desires.

Table 2.
Trauma-informed care involves 5 principles.[27]

1.	Bear witness to the patient's experience of trauma (commonly adverse childhood experiences).
2.	Help patients feel they are in a safe space and recognize their need for physical and emotional safety.
3.	Include patients in the healing process.
4.	Believe in the patient's strength and resilience.
5.	Incorporate processes that are sensitive to a patient's culture, ethnicity, and personal and social identity.

The CWT patients' goals may be as diverse as stable housing and food supply to finding better pain management.[23]

The CWT works with the patient to support their goals and hopes while trying to steer them towards the best practice management of their conditions. This type of relationship-based care/practice shifts the health care professional from being task-oriented to becoming more aware of caring practices.[24] Instead of focusing on the everyday tasks of patient care, the professional can now focus on what is important to the patient and include the patient and family in a plan of care.[24] The patient is asked to set a goal, which can be quite simple.[24] The professional gets to know the patient as a person versus a diagnosis.[24] Best practice is striving to implement evidence-based programs and strategies to enhance health and lower health problems with the greatest preventable burden.[25] By helping the patient reach their goals while striving for best practice, the CWT aims for a win-win situation.

Frequently with this demographic there is a mental health component to management including trauma.[26] The CWT attempts to practice trauma-informed care as required (Table 2).

The CWT's patient demographic can be challenging to work with. Compassion-fatigue, moral injury or vicarious trauma is more common for health professionals serving this population.[28] This compromises the health care professionals' ability to care for these patients due to avoidance of patient suffering, intrusive thoughts or dreams of distressing symptoms.[28]

Summary

Health care initiatives are under greater pressure than ever to prove their effectiveness and efficiency. For such programs to continue to exist, they will need to be measured for success by a number of different metrics. The CWT's purpose includes generating data and knowledge for future planning. Being able to generate consistent data is an important secondary objective of this program. The chiropractor has since started collecting data on patient outcomes using the BQ (Bournemouth Questionnaire) Pre-Treatment (Baseline) and Post-Treatment (Discharge).[29]

At the time of publication, the patient remained stable however it was determined the patient was not a liver transplant candidate. The patient remained on the CWT case load and monitoring continued. Participating on an interprofessional team is becoming more common for chiropractors. It has been a tremendous source of professional satisfaction for the principal author where every day with the team there is a learning moment. We highly recommend it for those who have this opportunity present itself.

Footnote:
As of April 1st, 2019 the KW4 Community Ward Team no longer has embedded LHIN Care Coordinators but is an entirely The Centre for Family Medicine Family Health Team – based team.

Take Away Points

- Case management is an effective approach for complex patients
- It can improve outcomes and more efficiently administer resources
- Team-based care is becoming more common
- Chiropractors are well positioned to contribute to such teams

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Suspected trigeminal nerve neuropathy causing persistent idiopathic facial pain: a report of four cases

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Persistent idiopathic facial pain is often a disabling condition for patients. Due to a lack of agreed upon diagnostic criteria and varied symptomatology, the diagnosis of persistent idiopathic facial pain is elusive and remains one of exclusion. It is typically described as a unilateral, deep, poorly localized pain in the territory of the trigeminal nerve, however there are a number of case reports that describe bilateral symptoms. Unlike trigeminal neuralgia, the condition encompasses a wider distribution that does not conform or relate to a specific dermatome. In addition, the pain is typically continuous, with no periods of remission and there are no signs or symptoms suggestive of autonomic involvement. Reports documenting the response to various conservative treatments for persistent idiopathic facial pain have been widely variable likely due to the heterogeneity of the condition. Four cases of persistent idiopathic facial pain

La douleur faciale idiopathique persistante est souvent une condition invalidante pour les patients. En raison de l'absence de critères diagnostiques convenus et d'une symptomatologie variée, le diagnostic de douleur faciale idiopathique persistante est difficile à établir et demeure un diagnostic d'exclusion. On décrit généralement la douleur comme étant unilatérale, profonde et mal localisée dans la région du nerf trijumeau, mais il existe un certain nombre de rapports de cas qui décrivent des symptômes bilatéraux. Contrairement à la névralgie faciale, l'affection englobe une distribution plus large qui n'est pas conforme ou liée à un dermatome précis. De plus, la douleur est généralement continue, sans période de rémission et il n'y a aucun signe ou symptôme suggérant une atteinte du système nerveux autonome. Les rapports documentant la réponse à divers traitements conservateurs utilisés pour la douleur faciale idiopathique persistante ont été très variables, probablement en raison de l'hétérogénéité de l'affection. Quatre cas de douleur faciale idiopathique persistante due à un soupçon de neuropathie du nerf trijumeau et

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due to suspected trigeminal nerve neuropathy and their management are presented. A specific form of targeted, manual, instrument-assisted, intra-oral vibration therapy appeared to provide relief in the four cases described.

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KEY WORDS: trigeminal nerve$_1$, facial pain, chiropractic, vibration therapy, atypical facial pain

leur prise en charge sont présentés. Dans les quatre cas décrits, une forme spécifique de thérapie par vibration ciblée, intraorale, manuelle, appuyée par des instruments, a semblé apporter un soulagement.

(JACC. 2019;63(2):126-138)

MOTS CLÉS : nerf trijumeau$_1$, douleur faciale, chiropratique, thérapie par vibration, douleur faciale atypique.

Introduction

Lifetime prevalence of non-specific facial pain is estimated at 26%.[1] The condition presents as a diagnostic challenge for even the most experienced clinicians due to its multifaceted presentation and etiology.

Persistent idiopathic facial pain (PIFP) is typically described as deep, poorly localized pain in the territory of the trigeminal nerve. It is most often unilateral, however approximately one-third of patients develop bilateral symptoms.[2] PIFP often spreads to a wider area, not conforming to a specific dermatome. In addition, the pain is typically continuous with no periods of remission and there are no signs or symptoms of autonomic involvement.[2] Recent evidence supports the notion that PIFP may itself be a neuropathic pain condition.[2, 3-7] Friedman *et al*.[8] demonstrated consistent maxillary alveolar tenderness (MAT) in the distribution of the maxillary nerve on the involved side of the face in patients diagnosed with atypical facial pain. The authors state that this tenderness is often incorrectly attributed to lateral pterygoid hypertonicity or spasm, despite the inaccessibility of this muscle to digital palpation.[8] They found intra-oral tenderness in the maxillary molar periapical area in over 90% of asymptomatic migraine patients, with laterality and degree of tenderness closely related to headache symptoms.[9] Significant ipsilateral tenderness and increased local temperature was found in the maxillary molar periapical area on palpation bilaterally during unilateral migraine, tension type headaches, and facial pain.[10]

Neuropathic pain secondary to trigeminal nerve injury has also been poorly defined in the literature.[11] Benoliel *et al*.[11] has proposed a modified diagnostic criteria for peripheral painful traumatic trigeminal neuropathy (PPTTN) with PPTTN defined as "spontaneous or touch evoked pain predominantly affecting the receptive field of one or more divisions of the trigeminal nerve. The duration ranges widely from episodic (minutes to days) and may also be constant. The pain tends to spread with time and is mostly unilateral without crossing the midline."[11 (pg 50)] They also suggest that the pain develops within three months of an identifiable trauma and lasts for more than 3 months.[11]

The purpose of this article is to familiarize the clinician with the clinical presentation, diagnosis, etiology and management of PIFP secondary to trigeminal nerve neuropathy as currently understood. Four cases are presented to illustrate the features of the disorder.

Case presentations

Case 1

A 28-year old female employed as a full-time medical receptionist presented to a chiropractic clinic with a 1.5-year history of bilateral facial pain located over the temporal and temporomandibular joint regions. The onset was attributed having a "nerve struck" while undergoing extensive dental work for caries. Subsequently, a root canal and a dental crown were required. The pain over the temporomandibular region was described as an intermittent dull ache. The temporal headache pain was described as a constant squeezing and pressure sensation occurring weekly. Both painful areas were rated at seven out of 10 on the numerical rating scale (NRS). The major aggravating factor was clenching. The pain was worse in the morning and after work. The patient denied any lancinating, burning or paroxysmal sensations in the face and denied

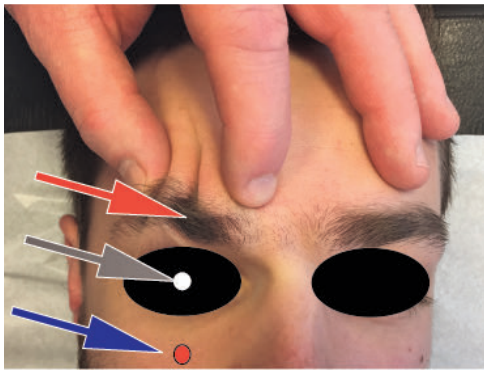

Figure 1.
Location of the infraorbital and supraorbital foramen. The infraorbital foramen is the exit point of the infraorbital nerve from the skull (red dot indicated by the blue arrow). By pinching the skin over the eyebrow directly over the pupillary line (with midline gaze), the supraorbital foramen (or notch) can be easily palpated and subsequently the supraorbital nerve (indicated by the red arrow). The location of the pupil (white dot) is indicated by the grey arrow.

Figure 2.
Proposed trigeminal nerve tension test. Palpating the supraorbital nerve just superior to the supraorbital foramen (or notch) (red arrow) the patient is asked to open their mouth. An increase in tension and/or pain would indicate a positive test.

any associated nausea, vomiting, photophobia, phonophobia and auras. At baseline, her Jaw Functional Limitation Scale score was zero, indicating no limitations. The Jaw Functional Limitation Scale has demonstrated good reliability and construct validity in assessing limitations in mastication, jaw mobility and verbal and emotional expression.[12]

On examination, the patient weighed 54 kg and stood 163 cm tall. Active and passive ranges of motion of the cervical spine were full and pain free. Orthopaedic testing of the cervical spine, inclusive of Spurling's and Jackson's, was unremarkable. The cervical paraspinal musculature was diffusely tender, however no pain referred to the areas of the chief complaint. A rightward C-shaped shift of the mandible was visualized during jaw opening but no obvious clicking was noted. A palpable clunk was noted bilaterally upon closing the mouth. No locking was reported or visualized. Incisal opening, using a Therabite ROM scale[13], measured 53mm, with left lateral mandibular deviation measuring 15mm and right lateral mandibular deviation measuring 10mm. There was a restriction noted during posterior-anterior joint play of the right temporomandibular joint (TMJ). There was moderate tenderness over the temporalis, masseter and medial pterygoid muscles bilaterally. Palpation over the infraorbital and supraorbital foramen, the exit point from the skull of the infra (See Figure 1) and supraorbital (See Figure 1) nerves, respectively, elicited tenderness, which increased on mouth opening (See Figure 2) recreating a lesser version of her temporal headache pain. Intra-oral palpation along the superior mucobuccal fold in the area of the superior alveolar nerve and inferior mucobuccal fold at the exit and distribution of the mental nerve, the terminal branch of the inferior alveolar nerve, was locally tender bilaterally reproducing her presenting symptoms. Examination of other cranial nerves and peripheral nervous system was within normal limits.

An MRI, ordered by her family medical doctor, of both temporomandibular joints (TM) demonstrated "a normal left TMJ. The right TMJ meniscus was anteriorly subluxed in the resting/neutral position but reduced on opening. There was no abnormality of configuration or signal change within the condylar head or glenoid surface of the TMJ."

Figure 3.
Locating the supratrochlear foramen (red arrow). By pinching the skin over the eyebrow directly over the inner canthus, the supratrochlear foramen can be easily palpated and subsequently the supratrochlear nerve.

A diagnosis of persistent idiopathic facial pain secondary to trigeminal nerve neuropathy, tension type headaches and temporomandibular disorder (TMD) was made. The plan of management consisted of mobilizations to the TMJ bilaterally, soft-tissue therapy to the affected cervical and orofacial musculature and intra-oral vibration-assisted soft-tissue therapy directed to the affected muscles of mastication and the superior and inferior mucobuccal fold in the area of the superior alveolar and mental nerves (see management section for further description of treatment procedure).

After 10 treatments over a 12-week period, the patient reported significant improvement in their headache and jaw pain. She reported experiencing less than one headache per month at 5-month follow-up. She reported a complete resolution of the pain located over the temporomandibular region. Follow-up at 5 months revealed that her jaw remained symptom free, but she continued to have occasional mild headache pain, as previously described.

Case 2

A 24-year old female employed as a part-time cashier and full-time student presented to a chiropractic clinic with a seven-year history of bilateral orofacial pain and a three-year history of recurrent neck pain. The patient reported the onset of pain overlying the TMJ began when she was 17-years old after her orthodontic braces were removed. The pain persisted and was made worse during a standard dental procedure. The pain was rated at a constant four out of 10 throughout the day. The pain was described as a dull ache with consistent referral of pain to the temporal, frontal and supraorbital regions of the head. Eating large pieces of food, requiring her to open her mouth wide, aggravated the pain. In addition, chewing gum and the use of a store bought mouth guard also exacerbated her symptoms. The patient received treatment intermittently directed to the TMJ for her jaw and headache symptoms from a chiropractor over a period of several years. These treatments consisted of cervical spinal manipulative therapy, soft-tissue therapy to the cervical and facial musculature and intra-oral vibration-assisted therapy directed at the muscles of mastication. She reported the treatments provided temporary relief of the headache pain but had improved opening and closing of her jaw considerably.

The patient felt her neck pain was due to poor postures while attending school. The pain was located bilaterally at the base of the neck extending to the postero-lateral shoulders bilaterally. The pain was described as a dull ache and rated at an average of 4 out 10. The pain was made worse by prolonged studying at a desk. The patient denied any upper extremity pain referral or paraesthesia.

On examination, the patient weighed 62 kg and stood 155 cm tall. Active and passive ranges of motion of the cervical spine were full and pain free. Spurling's and Jackson's tests were unremarkable. The cervical posterior paraspinal musculature was tender, however there was no pain referred to the areas of the TM complaint. A right-sided deviation on opening of the mouth was visualized. No jaw locking was seen on examination or reported by the patient. Incisal opening, using a Therabite ROM scale, measured 37mm. Palpation intraorally along the superior mucobuccal fold in the area of the superior alveolar nerve and inferior mucobuccal fold at the exit and distribution of the mental nerve was locally tender as were the muscles of mastication bilaterally. There was tenderness over the supraorbital (Figure 1) and supratrochlear (Figure 3) foramina, bilaterally and increased with mouth opening (See Figure 2). At baseline her Jaw Functional Limitation

Scale score was 17. Cranial nerves and peripheral nervous system examination was otherwise within normal limits.

A diagnosis of persistent idiopathic facial pain secondary to trigeminal nerve neuropathy and TMD was made on the basis of the clinical assessment. The plan of management consisted of mobilizations to the TMJ bilaterally, soft-tissue therapy to the affected cervical and orofacial musculature and intra-oral vibration-assisted soft-tissue therapy directed to the affected muscles of mastication and the superior and inferior mucobuccal fold in the area of the superior alveolar and mental nerves (refer to management section for further description of treatment procedure).

The patient reported a complete resolution of her orofacial pain after seven treatments over a seven-week period. Follow-up at three-months revealed that she had remained symptom free.

Case 3

A 25-year old female full-time student presented to a chiropractic clinic with a five-year history of bilateral jaw pain and headaches. The jaw pain and headaches reportedly began shortly after prolonged dental work. The headaches were described as a daily dull ache located over the temporal regions. The patient denied any nausea, vomiting, photophobia, phonophobia, or auras. Prolonged studying, sitting and/or driving aggravated the headache pain. Self-directed, extra-oral massage of the TMJ muscles reportedly decreased the intensity of the headaches. The jaw pain was described as dull and achy and rated three to four out of 10 on an NRS. Teeth grinding and not wearing her nighttime mouth guard, prescribed by her dentist, increased the TMJ pain. The patient described a non-painful clicking on opening and closing the jaw. The patient denied any lancinating or paroxysmal pain. At baseline her Jaw Functional Limitation Scale score was 25. The patient reported receiving ongoing treatments at intermittent frequency directed to the TMJ for her jaw and headache symptoms from a chiropractor for a period of several years. Her treatments consisted of cervical and facial soft-tissue therapy and intra-oral vibration-assisted therapy directed at the muscles of mastication. She reported the treatments provided only temporary relief of the headache pain but had improved her jaw function considerably.

On examination, the patient weighed 70 kg and stood 155 cm tall. Active and passive ranges of motion of the cervical spine were full and pain free. Spurling's and Jackson's test were unremarkable. The posterior cervical paraspinal musculature was tender, however there was no referred pain to the areas of the TM complaint. Opening and closing the mouth revealed a non-painful audible clicking in the bilateral TMJ. Incisal opening, using the Therabite ROM scale, measured 38mm, with left lateral mandibular deviation measuring 10mm and right lateral mandibular deviation measuring 11mm. Maximal opening caused pain along the left TMJ. Restriction during anterior-posterior joint play of bilateral TMJ was noted. Palpation intraorally of the muscles of mastication and along the superior mucobuccal fold in the area of the superior alveolar nerve elicited tenderess bilaterally. Palpation over the supraorbital foramen (or notch) (Figure 1), was tender bilaterally, increasing with mouth opening (See Figure 2). Examination of the cranial nerves and peripheral nervous system was within normal limits.

A diagnosis of persistent idiopathic facial pain secondary to trigeminal nerve neuropathy and TMD was made. The plan of management consisted of mobilizations to the TMJ bilaterally, soft-tissue therapy to the affected cervical and orofacial musculature and intra-oral vibration-assisted soft-tissue therapy directed to the affected muscles of mastication and the superior and inferior mucobuccal fold in the area of the superior alveolar and mental nerves.

The patient reported a significant improvement in her headache and jaw pain. After 20 treatments over a nine-week period the patient reported having less than one headache per month and intermittent, two out of 10 jaw pain intensity. Follow-up at eight-months revealed that the improvement had been maintained. The patient is currently receiving ongoing symptomatic care for her intermittent low-grade jaw pain and headaches.

Case 4

A 63-year old female retired florist presented to a chiropractic office with a three-month history of left jaw pain. No headaches were reported. The jaw pain reportedly began shortly after a dental crown procedure. The patient described the pain as a constant dull ache; however, she noted that up to three times per week she experienced sharp, shooting pain along the left mandible from the angle of the mandible toward the mental foramen. The jaw pain was rated at a daily four to five out of 10 on an NRS with episodes of sharp, shooting pain reaching eight out of 10.

Aggravating factors for the jaw pain included eating large mouthfuls of hard foods, such as apples, and talking and moving her tongue in side-to-side movements. At baseline her Jaw Functional Limitation Scale score was zero.

On examination, the patient weighed 79.4 kg and stood 152.4 cm tall. Active and passive ranges of motion of the cervical spine were within normal limits. Spurling's and Jackson's were unremarkable. The cervical paraspinal musculature was tender, however there was no pain referred to the areas of the TM complaint. No audible click or deviation was noted during maximal jaw opening. Incisal opening, utilizing the three finger test, was full (three fingers) and pain free. Palpation intraorally along the superior mucobuccal fold in the area of the superior alveolar nerve and inferior mucobuccal fold at the exit and distribution of the mental nerve revealed local tenderness on the left side more so than the right. Palpation of the inferior mucobuccal fold in the area of the exit and distribution of the mental nerve reproduced the chief complaint. Palpation over the supraorbital (See Figure 1) and supratrochlear (See Figure 3) foramina was unremarkable bilaterally. Palpation of the temporalis muscle was nontender, however, palpation intra and extra-orally of all other accessible muscles of mastication were focally tender. Examination of the cranial nerves and peripheral nervous system was within normal limits.

A diagnosis of persistent idiopathic facial pain secondary to trigeminal nerve neuropathy and trigeminal neuralgia was made. The plan of management consisted of facial acupuncture, low level laser therapy (LLLT) to the TM joint, soft-tissue therapy to the affected cervical and orofacial musculature, and intra-oral vibration-assisted soft-tissue therapy directed to the affected muscles of mastication and the superior and inferior mucobuccal fold in the area of the superior alveolar and mental nerves bilaterally. The patient had had success with both LLLT and acupuncture for previous cervical and upper thoracic musculoskeletal complaints so it was included in the plan of management.

The patient reported a significant improvement in her jaw pain after her first treatment. After six treatments over a six-week period the patient reported having a minimal amount of pain and very few episodes of shooting pain. The patient also received a custom-made mouthguard from her dentist three weeks into treatment, which she felt also helped her jaw pain. Follow-up at three-months revealed no pain since her last visit. The patient continued to receive ongoing supportive care as above.

Discussion

The applicability of understanding this complex anatomy for clinicians who utilize intra-oral digital palpation as part of their assessments cannot be overstated. Under normal circumstances, tissues overlying peripheral nerves are painless to gentle palpation, however, when neural tissue is inflamed even gentle palpation can cause focal and referred pain.[14, 15]

Given the complexity of trigeminal nerve neuroanatomy and its' numerous connections, dysfunction and/or injury can result in a myriad of symptoms, some of which will be discussed below. (See Appendix 1 for a more thorough discussion of the anatomy of the trigeminal nerve).

Pathophysiology of nerve injury

Peripheral neuropathy is a blanket term used to describe disorders affecting the function of one or more peripheral nerves due to numerous causes including trauma, disease or dysfunction.[16] Within the nerve sheath, the blood nerve barrier is vital to ensure homeostasis. Breakdown of the barrier can occur with a nerve injury, including compression and tension injuries.[17] Secondary to breakdown of the barrier, is a loss of homeostasis within the endoneural environment.[17] This results in the accumulation of various proteins as well as infiltration by lymphocytes, fibroblasts, and macrophages as a reaction to previously protected antigens contained within the perineurial space.[18, 19] There is thus the potential for a mini-compartment syndrome within the fascicles due to the build up of fluid.[18] The tortuous arrangement of the fascicles in the peripheral nerves embedded in its fibrous matrix is predisposed to the mechanical propagation of this neurogenically induced local compartment syndrome.[18] The neurogenically induced inflammation provides a cytokine-based mechanism for a cycle of inflammation without frank demyelination. This helps to explain the inconclusive and often normal findings found on electrophysiological studies or other imaging modalities in disorders where patients report paraesthesia, such as in carpal tunnel syndrome, thoracic outlet syndrome and atypical facial pain.[18]

Relatively recent diagnostic ultrasound work has demonstrated that affected nerves show a significantly increased cross-sectional area compared to the asymptomatic

side.[20] Kara *et al*.[20] showed an increase in the sciatic nerve in those patients with low back pain and unilateral sciatica. Cartwright *et al*.[19] reported similar findings in the median nerve in unilateral carpal tunnel syndrome, as did Cho *et al*.[21] in unilateral occipital neuralgia with the greater occipital nerve. It would stand to reason that an inflamed nerve would be predisposed to a greater number of entrapment sites along its route due to an increase in cross-sectional area and thus a greater proximity to surrounding soft tissue.

Support for the hypothesis that an inflamed nerve is more predisposed to compression has come from the results of decompression procedures.[22] Direct observation of the scalene triangle during such procedures has demonstrated fibrous bands and persistent adhesions that can be visualized to limit normal movement of the brachial plexus.[22] Small perineural adhesions are said to be responsible for the continued symptom formation and, as noted, are difficult to visualize with current imaging techniques. Resection of the perineural adhesions normalizes blood flow and results in the resolution of distal vasospasms as confirmed thermographically.[22] Ellis[17] reported that more than 100 surgical decompressive cases utilizing intraoperative thermography documenting the presence of both directly mechanical (fibrous bands and fibrotically transformed muscles) and neurogenic (small perineural adhesions) mechanisms for neural constriction, inflammation and distal vasospasm. Evidence of similar pathophysiology has been documented for ulnar and median nerve entrapments. Rosenbaum *et al*., concluded that the presence of endoneural fibrosis was the most damaging due to the direct effect on nerves and blood vessels.[23]

Upton and McComas[24] introduced the concept of the double crush phenomenon in the early 1970's. They stated that a proximal nerve injury might predispose distal sites to compression and that the opposite, reverse double crush, was also a possibility. Upton and McComas[24] noted that compression along the nerve alters axoplasmic flow, which subsequently leads to pathology and symptomatology. These changes have been demonstrated in animal models.[24] Although most authors agree it is more of a "multiple crush" presentation[25], this is an important concept clinically as the failure to recognize and treat the multiple points of injury will likely result in an unsuccessful outcome.

Sauer *et al*.[26] showed that the nervi nervorum provide innervation to the nerve sheath in peripheral nerves and are nociceptive in nature. The nervi nervorum may be the cause of both nerve trunk pain and dysesthetic pain which may be caused by persistent inflammation in the nerve.[27] Bove states, "symptoms can appear as local tenderness of the nerve (nerve trunk pain), or a 'doorbell' type of response, where palpation leads to dysesthetic symptoms in a more distal region."[27] Cranial nerves are known to be richly innervated by nervi nervorum.[28]

The neurogenic inflammation, causing an irritation within the nerve sheath, leads to the cascade of swelling, cellular recruitment and spread of perineural and endoneural fibrosis, and provides a possible explanation for the enigmatic clinical presentation of persistent idiopathic facial pain. This mechanism helps explain the finding of pain on palpation of the various branches of the trigeminal nerve in the presented cases and also the lack of sensory findings (numbness or tingling) or motor deficits (weakness or atrophy).

Diagnosis

Atypical facial pain or more commonly referred to as, persistent idiopathic facial pain (PIFP) is typically described as deep, poorly localized pain in the territory of the trigeminal nerve. It is most often unilateral, however approximately one-third of patients develop bilateral symptoms.[2] Unlike trigeminal neuralgia, PIFP often spreads to a wider area, not conforming to a specific dermatome. In addition, the pain is typically continuous with no periods of remission and there are no signs or symptoms of autonomic involvement. Apart from sensitivity to palpation and tension along the course of the nerve, as demonstrated in our four cases, the condition shows no other abnormal findings in clinical examination or additional investigations and as a result, the diagnosis of PIFP is one of exclusion.[2]

Currently there is a lack of diagnostic criteria and validated diagnostic procedures. However, the International Headache Society (IHS) describes PIFP as persistent facial and/or oral pain, with varying presentations but recurring daily for more than two hours per day over more than three months, in the absence of clinical neurological deficits.[28] In addition, IHS suggests the following diagnostic criteria:

- facial and/or oral pain that is recurring daily for greater than two hours per day for greater than three months;
- the pain is characterized as being poorly local-

ized, not following the distribution of a peripheral nerve;
- the character of pain is either dull, aching, or nagging.
- In addition, the clinical neurological examination is normal and the pain cannot be attributed to any known or understood pathological process.[28]

Recent evidence supports the notion that PIFP may itself be a neuropathic pain condition.[2,3-7] Friedman *et al*.[8] demonstrated consistent maxillary alveolar tenderness (MAT) in the distribution of the maxillary nerve on the involved side of the face in patients diagnosed with atypical facial pain. The authors state that this tenderness is often incorrectly attributed to lateral pterygoid hypertonicity or spasm, despite being inaccessible to digital palpation.[8] Preliminary data of intra-oral tenderness in the maxillary molar periapical area was noted in over 90% of asymptomatic migraine patients, with laterality and degree of tenderness closely related to headache symptoms.[9] On palpation bilaterally during unilateral migraine, tension type headaches, and facial pain, the maxillary molar periapical area displayed significant ipsilateral tenderness and increased local temperature.[10] As previously stated, local neural inflammation can result in symptom provocation to normally non-noxious palpation.[15] In this example, tissues surrounding the jaw supplied by terminal branches of the maxillary nerve are sensitized secondary to neural inflammation.

In order to improve the accuracy of detecting subclinical cranial neuropathies, sensitive neurophysiological recording and quantitative sensory testing (QST) have been recommended. Jääskeläinen *et al*.[5] examined whether electrophysiological testing of trigemino-facial system could aid in the diagnosis of atypical facial pain. They found that atypical facial pain patients could be divided into three groups based on their electrophysiological findings:

1. major trigeminal neuropathy, representing the patients that had both clinical and electrophysiological signs of trigeminal neuropathy despite normal brain magnetic resonance imaging (MRI);
2. minor trigeminal neuropathy, where the patients displayed increased excitability during electrophysiological testing signifying trigeminal nerve dysfunction despite normal MRI; and
3. idiopathic, where despite the clinical symptoms there were no abnormalities with electrophysiological or QST testing.[5]

Forssell *et al*.[2] sought to elucidate the pathophysiology of atypical facial pain and test the idea that the disorder represents a clinically undiagnosed neuropathic pain condition through the use of clinical, neurophysiologic and thermal QST. They examined atypical facial pain patients and patients with diagnosed trigeminal neuropathic pain. The majority (75%) of atypical facial pain patients showed abnormalities in electrophysiological or thermal QST recordings or both. It would appear that PIFP is a heterogeneous entity, which is better understood as a disorder that forms a spectrum of neuropathic involvement due to the amount of inflammation present.

Due to the relative lack of a gold standard in diagnosing trigeminal neuropathic pain, the suspected diagnosis of PIFP secondary to trigeminal neuropathy may best be facilitated with palpation and proposed nerve tensioning (i.e. mouth opening). Ellis[17] states that the inflammation, if not identified and treated appropriately, can spread both perineurally (within the nerve) and endoneurally causing activation of the CNS. A study done by Younger *et al*.[29] showed changes in the gray matter in several areas of the brain in patients with TMD versus controls on MRI. The authors reported these changes were likely associated with dysregulation of the trigeminal and limbic systems as well as somatotopic reorganization in the putamen, thalamus and somatosensory cortex[29]. The mechanism outlined by Ellis can then cause the subsequent inflammation of other adjacent/connected nerves, and can lead to mirror symptoms in the opposite limb.[17] With respect to the four cases outlined above, this could be the mechanism involved in the tenderness of several branches of the trigeminal nerve accessible to palpation including the supratrochlear, supraorbital, infraorbital, the muscular branches to the muscles of mastication and the superior alveolar and mental nerves that may occur bilaterally even with a unilateral complaint.

Therefore, tenderness of the nerves branches would suggest inflammation along the route of the trigeminal nerve with an element of central sensitization. It can be difficult to be confident that the nerve is being palpated especially when other structures in the area could be the source of pain. Increasing pain with palpation of the nerve under tension especially with no other soft tissue structure crossing the tensioned structure would reinforce that the

palpated tissue is in fact the nerve. In cases 1, 2 and 3 there was tenderness over the supraorbital nerve branch that increased under the tension position of mouth opening. This position would theoretically create tension in the trigeminal nerve due to a stretch of the inferior alveolar branch, in particular the mental nerve, with opening. To our knowledge, this proposed trigeminal nerve tension test has not previously been described in the literature.

Management

Secondary to the concept of increased tenderness and temperature being signs of inflammation, Friedman *et al*.[9] examined the efficacy of intra-oral chilling for the relief of headache pain. A randomized controlled study design was used to evaluate the effectiveness of maxillary intra-oral chilling (MIC) in 35 symptomatic episodic migraine patients compared to oral sumatriptan and sham (tongue) chilling. Pain and nausea was recorded at baseline, one, two, four and 24 hours after the treatment. Both MIC (p= 0.001) and sumatriptan (p= 0.024) significantly reduced the pain scores compared to control group. No significant differences between MIC and sumatriptan were noted.[9] Of interest, the sumatriptan caused mild side effects, including dizziness, paraesthesia, and somnolence, in seven of 12 subjects. Only one in 12 subjects for the MIC group reported a minor side effect, which included dizziness and post-treatment gingival tenderness.

Friedman *et al*.[30] also investigated the effectiveness and tolerability of topical nonsteroidal anti-inflammatory drugs (NSAID) to treat headache patients. Topical medication was applied to the periapical area of the maxillary molars on the symptomatic side(s) in 20 chronic headache patients. Patients recorded the frequency, severity, duration, and type of headache for 60 days. After 30 days the patients were instructed on how to apply the medication and told to apply the medication once daily for 30 days. The patient reported headache burden was examined, which was defined as the average headache intensity (0 to 10 scale), multiplied by its duration in hours. The authors noted that the average headache burden score decreased from 454.8 (baseline 30 days) to 86.5 (during the 30 day treatment phase).[30] The reported side effects were minor and included one incident of gastrointestinal intolerance, one incident of burning gingiva, and one incident of cheek irritation. They concluded that NSAID applied to the periapical area of the maxillary molars on the symptomatic side(s) was significant (p<0.001) in reducing headaches with minimal side effects.[30]

Figure 4.
Depiction of palpation and treatment of the left mental nerve using targeted vibration therapy. Acupuncture points are targeting (from left to right to midline) Gallbladder 1, the right supraorbital foramen (exit of the supraorbital nerve), Urinary Bladder 2 – right supratrochlear foramen (exit of supratrochlear nerve), and Extra 3 (Yintang).

In addition to the conservative treatments outlined above, a targeted manual instrumented assisted intra-oral vibration therapy has demonstrated effectiveness (See Figures 4 and 5)[31]. Targeted vibration therapy is hypothesised to aid in the breakdown of scar tissue and mobilize the surrounding soft-tissues[32], increase local blood flow[33] and reportedly, to decrease pain.[34, 35] Kerschan-Schindl *et al*.[33] demonstrated a significant increase in blood volume and mean blood flow after three-minutes of vibration exercise in the quadriceps and gastrocnemius muscles. The suggested mechanism of action of vibration therapy is to reduce blood viscosity and improve blood flow velocity.[33] Intra-oral vibration therapy theoretically disrupts the proposed dynamic ischemia occurring within the trigeminal nerve in patients with PIFP via mobilizing the nerve itself and surrounding soft-tissues, as well as promoting improved local circulation.

It is important to note, however, that a lack of a clear pathophysiological basis makes it difficult to establish a validated/evidence-based treatment protocol. Given the

Figure 5.
Depiction of palpation of the left superior mucobuccal fold in the area adjacent to the left superior alveolar nerve.

psychosocial comorbidities that accompany chronic pain conditions and the lack of randomized controlled trials for the putative diagnosis, a pragmatic multidisciplinary approach is recommended. Since the application of vibratory stimulation has been shown to increase local blood flow[33] and decrease pain[34, 35], it may be reasonable to assume that targeted vibration therapy may be an effective intervention in the treatment of the suggested dynamic ischemia in the terminal branches of the trigeminal nerve.

Summary

Four cases of PIFP due to suspected trigeminal nerve neuropathy and their conservative management are described. Studies have postulated that in patients with PIFP there is local pathology (inflammation) of the distal branches of the trigeminal nerve, which results in the range of symptoms described. Empirically, the use of specific manual release techniques (soft tissue therapy), and the use of a hand held apparatus that transmits vibrations might be useful. At this time, PIFP thought to be due to trigeminal nerve neuropathy remains a diagnosis of exclusion. Awareness of other sinister conditions that may mimic PIFP such as, giant cell arteritis, septic arthritis/infection of the TMJ, tumors and pseudo-tumours of the TMJ as well as common etiological conditions with accepted standard treatments is imperative for the practitioner managing persistent facial pain. If suspected, appropriate referral and investigations should be pursued prior to the commencement of the treatments proposed above or if the clinical picture fails to resolve in a reasonable amount of time.

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Appendix 1.
Anatomy review of the trigeminal nerve

The trigeminal nerve is the second largest cranial nerve in the body.[36,37] The nerve is enveloped by a connective tissue called the mesoneurium, which is critical to allow gliding during movement.[36,37] The mesoneurium is continuous with the external epineurium, which defines and surrounds the nerve trunk. The nerve trunk is composed of myelinated and unmyelinated nerve fibres bound together within fascicles, which are surrounded by the perineurium.[36] In between the fascicles is the internal epineurium. Finally, the connective tissue within the matrix of the fascicle is termed the endoneurium.[36]

The trigeminal nerve supplies both general somatic afferent and special visceral efferent to derivatives of the first pharyngeal arch.[36,37] It is comprised of four nuclei, three sensory and one motor.

The composition of the sensory root is predominantly pseudounipolar neurons that make up the trigeminal ganglion. The trigeminal ganglion, housed within a dural recess lateral to the cavernous sinus, receives afferent and efferent information from the peripheral ganglionic neurons forming the three nerve divisions: ophthalmic (CN V1), maxillary (CN V2), and mandibular (CN V3).[36, 38]

CN V1 exits the cranium via the superior orbital fissure and provides the afferent supply to the skin, mucosa membranes and conjunctiva of the front of the head and nose as well as the anterior and supratentorial dura mater.[36, 38] The first branch from the ophthalmic division is the small recurrent meningeal branch, which supplies the sensory innervation to the dura mater. The ophthalmic division divides into three branches the names of which indicate the structures innervated: lacrimal nerve, frontal nerve and nasociliary nerve.[38] The frontal nerve further branches into the supratrochlear and supraorbital nerves that exit the skull from their similarly named foramina. Fallucco *et al*. report that only 26.7% of the cadavers in their study had a supraorbital foramen while 83.3% had a notch.[39] In 10% of the cadavers there was both a foramen and a notch.[39] In 86.0% of their cases with a notch, the neurovascular bundle passing through was encased by fibrous bands suggesting a potential compression site.[39] The supratrochlear nerve supplies sensation to the skin and soft tissue of the glabella, lower medial portions of the forehead, upper eyelid and conjunctiva.[40] The supraorbital nerve further branches into a superficial and deep branch. The deep branch runs deep to the frontalis muscle in a superior direction supplying sensation to the frontoparietal scalp. The superficial branch further divides with some divisions either penetrating or passing over the frontalis muscle as it travels superiorly to supply sensation to the forehead and anterior scalp.[41]

CN V2 exits the cranium via the foramen rotundum and enters the pterygopalatine fossa and divides into the zygomatic nerve, ganglionic branches to the pterygopalatine ganglion and infraorbital nerve.[36, 38] CN V2 supplies sensation to the skin of the face over the maxilla, including the upper lip, maxillary teeth, mucosa of the nose, maxillary sinuses, and palate, as well as the dura matter of the anterior part of the middle cranial fossa.[36, 38]

The infraorbital nerve passes through the inferior orbital fissure to enter the infraorbital canal and exits onto the face via the infraorbital foramen (See Figure 1).[42] Its fine terminal branches supply the skin between the lower eyelid and upper lip. The anterior superior alveolar branches, middle superior alveolar branches and posterior superior alveolar branches are the other terminal branches and form the superior dental plexus, which supplies sensory innervation to the maxillary teeth.[38] These three branches are often the targets for nerve blocks during dental procedures.[42] Since there are numerous overlying structures in this area, including medial and lateral pterygoid muscles, a structurally accurate diagnosis cannot be made through palpation alone. However, sensitivity in this area suggests local inflammation, which may be inclusive of the posterior superior alveolar nerve.[43, 44]

Unlike the other two branches, CN V3 has both general sensory and branchial motor components. The mixed afferent-efferent mandibular division exits the cranium through the foramen ovale and enters the infratemporal fossa on the external aspect of the base of the skull.[36, 38] It gives off a meningeal branch, which re-enters the middle cranial fossa to supply sensory innervation to the dura.[36, 38] The mandibular division has four main sensory branches: auriculotemporal nerve, lingual nerve, inferior alveolar nerve, and buccal nerve. The branches of the auriculotemporal nerve supply the temporomandibular joint, the temporal skin, external auditory canal, and the tym-

panic membrane. The lingual nerve supplies the sensory fibers to the anterior two-thirds of the tongue. The afferent fibers of the inferior alveolar nerve pass through the mandibular foramen into the mandibular canal and branches into the inferior dental plexus, which supply the mandibular teeth. The mental nerve, a terminal branch of the inferior alveolar nerve, supplies the skin of the chin, lower lip and body of the mandible. The buccal nerve pierces the buccinators muscle and supplies sensation to the mucous membrane of the cheek.[36, 38]

The efferent fibers of the inferior alveolar nerve supply the mylohyoid and the anterior belly of the digastric muscles. The pure motor branches of the CN V3 leave the main trunk distal to the origin of the meningeal branch and supply the masseter muscle (masseteric nerve), temporalis muscle (deep temporal nerve), pterygoid muscle (pterygoid nerve), tensor tympani muscle (nerve of the tensor tympani muscle) and tensor veli palatini muscle (nerve of the tensor veli palatini muscle).[36, 38]