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3 ABSTRACT

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4 Neck pain is a common problem in the general population with high risk of ongoing 5 complaints or relapses. Range of motion (ROM) assessment is scientifically established in 6 the clinical process of diagnosis, prognosis and outcome evaluation in neck pain. 7 Anatomically, the cervical spine (CS) has been considered in two regions, the upper and 8 lower CS. Disorders like cervicogenic headache have been clinically associated with dysfunctions of the upper CS (UCS), yet ROM tests and measurements are typically 9 10 conducted on the whole CS. A cross-sectional study assessing 19 subjects with non-specific 11 neck pain was undertaken to examine UCS extension-flexion ROM in relation to self-reported 12 disability and pain (via the Neck Disability Index (NDI)). Two measurement devices 13 (goniometer and electromagnetic tracking) were employed and compared. Correlations between ROM and the NDI were stronger for the UCS compared to the CS, with the 14 strongest correlation between UCS flexion and the NDI-headache (r = -0.62). Correlations 15 16 between UCS and CS ROM were fair to moderate, with the strongest correlation between UCS flexion and CS extension ROM (r = -0.49). UCS flexion restriction is related to 17 headache frequency and intensity. Consistency and agreement between both measurement 18 systems and for all tests was high. The results demonstrate that separate UCS ROM 19

assessments for extension and flexion are useful in patients with neck pain.

Extension and flexion in the upper

² cervical spine in neck pain patients

3 Keywords:

4 Upper cervical spine, range of motion, neck pain, headache, disability.

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INTRODUCTION

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8	Neck pain is common in the general population with a 12-month prevalence between 10 -
9	20% (Hoy et al., 2010). Non-specific (or idiopathic) neck pain predominates (McLean et al.,
10	2010). People in high income countries, particularly women, office, or computer workers are
11	most affected (Hoy et al., 2010). Previous neck pain is a strong risk factor for ongoing
12	complaint or relapse (Hush et al., 2011).
13	Cervical spine (CS) range of motion (ROM) is inversely associated with neck pain (Dall'Alba
14	et al., 2001), and popularly used for diagnosis, evaluation (de Koning et al., 2008) and
15	treatment (Jull et al., 2008a). CS ROM has been shown to predict recovery in Whiplash-
16	Associated Disorders (WAD) (Dall'Alba et al., 2001)) and non-specific neck pain (Olson et al.,
17	2000). Conversely, a recent, large cohort study found no difference in CS ROM between
18	young subjects with chronic neck pain, and healthy volunteers (Kauther et al., 2012).
19	The cervical spine is divided into upper (occiput to C2/3) and lower (C3/4 to C7) regions,
20	which differ considerably mechanically (Bogduk and Mercer, 2000). Extension-flexion (E-F)
21	in the upper cervical spine (UCS) involves a head-on-neck motion strategy that reflects the
22	unique shape and structure of the occiput and first two cervical vertebrae (Bogduk and
23	Mercer, 2000). Pathoanatomically, cervicogenic headache has been attributed to the UCS as
24	the site of the trigeminocervical nucleus where trigeminal nerve afferents merge with the
25	upper three cervical nerves (Bogduk, 1994, Jull, 1994). Clinically, ROM of the UCS can be
26	assessed and related to headache and neck pain (Ogince et al., 2007).
27	Radiographic investigations suggest that UCS E-F may not be effectively detected during
28	ROM tests of the CS (Ordway et al., 1997, Ordway et al., 1999). In particular, Ordway et al.
29	caution that cervical curvature may relax at end of range (EOR). An important example for
30	the UCS is the chin moving forward at EOR flexion, which induces UCS extension. Maximal
31	E-F of the UCS is better assessed by examining retraction and protraction, respectively

32 (Ordway et al., 1999, Takasaki et al., 2011). However, whether limited retraction or protraction relates to mobility restriction in either the UCS and/or lower CS has not been well 33 34 identified (Hanten et al., 2000, Severinsson et al., 2012). 35 Relationships between UCS dysfunction and headache are known (Amiri et al., 2007, Jull et 36 al., 2007, Gadotti et al., 2008, International Headache Society, 2013). However, limited 37 evidence exists for the relationship between UCS E-F, and neck pain (Rudolfsson et al., 38 2012), or headache (Zito et al., 2006). The supine Flexion-Rotation test examines UCS 39 rotation in a position of full CS flexion (Hall and Robinson, 2004) and is frequently impaired in 40 neck pain and cervicogenic headache (Hall and Robinson, 2004, Smith et al., 2008). The 41 craniocervical flexion test is often positive in patients with neck pain, and with headache (Jull 42 et al., 2007, Jull et al., 2008b). Performed as an exercise, this movement has shown efficacy 43 in treating both clinical presentations (Jull et al., 2002, Falla et al., 2008, Falla et al., 2011). 44 However, association between subjectively reported neck pain or headache, and objectively measured ROM of the UCS, has not been investigated yet. The aim of the present study 45 therefore was to assess the ROM in the UCS and the whole CS in patients, and to 46 47 investigate a correlation between ROM and the patients' pain and disability.

48 METHODS

<u>Design</u>

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50 Cross-sectional study.

<u>Subjects</u>

Subjects with non-specific neck pain were recruited through online advertising at the local university campus. Subjects were included according to the following criteria: working-age patients suffering from sub-acute or chronic non-specific head and neck pain, with disability due to their neck pain (at least five points on the Neck Disability Index; NDI), for four weeks (or longer) prior to data collection.

- ACCEPTED MANUSCRIPT 57 Subjects with comorbidities known to influence the UCS were excluded. Exclusions included: Current or previous head and neck pain due to specific disorders, such as WAD, cervical 58 59 radiculopathy, migraine, tension or cluster-type headache; Systemic inflammatory disease 60 (like rheumatoid arthritis); Osteoporosis; Central nervous system diseases (like Parkinson's); 61 Ear infection with dizziness or tinnitus; Medication interfering with perception; Diabetes; Tumours; and pregnancy. 62 63 Prior to measurements, all included patients signed informed consent. The study was 64 approved by the regional ethics committee. 65 **Measurement Systems** The CROM™¹ is a cervical range of motion device with proven clinical utility in measuring E-66 F of the UCS (Dhimitri et al., 1998), and validity for use in the CS (Tousignant et al., 2000). 67 Inter-tester reliability for the CROM is reported for the UCS to be ICC>=0.89 (Dhimitri et al., 68 1998). The Polhemus G4 (originally called the 3-Space, Colchester, Vermont, USA) is an 69 70 electromagnetic 3D-tracking device used to quantify UCS (Amiri et al., 2003) and CS ROM 71 (Ordway et al., 1997, Tousignant et al., 2000, de Koning et al., 2008). Within and between-72 day reliability for the 3-Space has been reported to be ICC>=0.97 (Amiri et al., 2003). Using 73 a common protocol, we employed both instruments concurrently to assess extension-flexion 74 motion in the UCS, which broadened our study's relevance to both the clinical and
 - CS movements were recorded using the CROM[™], and the G4. Measurement systems set-up is illustrated in Figure 1. Patients wore the CROM[™] without its horizontal magnetic compass for measuring rotation. The G4 sensor was attached to the CROM[™] above the nose and plugged into the G4 System Electronics Unit (Hub). The G4 system source was placed 120cm above the ground and distanced 80cm from the patient's stool.

laboratory-based settings, and enabled comparison between the devices.

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¹ Performance attainment associates: http://www.spineproducts.com

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Procedure

84 All subjects completed the NDI questionnaire within one week before measurement. The NDI 85 is widely accepted for use with neck pain patients (Vernon and Mior, 1991, MacDermid et al., 86 2009, Swanenburg et al., 2013). Scores range from 0 - 50 points, expressed in percent. 87 According to Vernon and Mior a score below 10% represents no disability, 10 - 28% mild 88 disability, 30 - 48% moderate disability, 50 - 68% severe disability and >68% complete 89 disability. 90 ROM tests were performed in the seated position as a modification to the standing method described by Dhimitri et al. 1998, and to accommodate subjects with a hyper-kyphotic 91 92 thoracic spine. During CS E-F tests, subjects were asked to sit upright, with both hands 93 relaxed on their lap. To achieve a neutral head and neck position, they were instructed and manually guided by a tester, to position their forehead vertically. Subjects were asked to 94 95 move as far as possible into extension, and flexion, without changing their upper body 96 position. 97 For UCS E-F, subjects were asked to sit upright, rest their hands on their lap, and keep their 98 head in an upright position by leaning their back and occiput against a wall, while maintaining 99 their forehead in a vertical position (Figure 1). Subjects were coached to keep their thoracic 100 spine and shoulder blades in contact with the wall during testing. In assessing UCS 101 extension, subjects were instructed to, "Move the chin upwards while gliding with the occiput 102 downward" (Figure 2) and, "Move the chin downwards, like nodding, and the occiput 103 upwards" for UCS flexion (Figure 3). 104 Two warm-up trials were completed with verbal and manual coaching from our main tester. 105 Thereafter, three independent repetitions of a full cycle (E-F) were performed by the subject, 106 starting with extension. The test order (UCS or CS motion first) was randomised.

107	<u>Tester</u>
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A single experienced tester (MJE) monitored subjects' movement, read the CROM™ instrument and recorded the neutral, maximal extension, and maximal flexion values in degrees. A second tester (SS) operated and monitored the G4.

Data Analysis

112 As the CROM ™ measurement scale cannot be adjusted to zero, maximal E-F values were 113 computed by subtracting the values of the neutral start position, e.g. maximum flexion value 114 - start position value = flexion ROM. Maximal E-F values derived by the G4 during the 115 different tasks were computed using the VRRS Cervix Software (Kymeia Group, Padova, 116 Italy) and used later for statistical analysis. Extension was expressed as negative values, 117 flexion as positive. 118 Statistical analysis was performed using the software package R (R Development Core 119 Team, 2008). Mean of three repetitions was calculated for each movement and used for 120 comparisons between the UCS versus the CS, and UCS or CS versus the NDI. The total 121 NDI, NDI-pain item (#1), and NDI-headache item (#5) were separately compared to ROM. 122 Correlations between measurements for the UCS and CS versus the NDI were analysed 123 using Pearson's product moment correlations. Correlations, irrespective of the direction, of < 124 0.25 indicate little or no correlation, 0.25 - 0.5 fair, >0.5 - 0.75 moderate to good, and values > 0.75 denoted strong correlation (Portney and Watkins, 2000, Friendly, 2002). 125 126 Comparisons between the two measurement systems were calculated using Intraclass-127 Correlation Coefficients of Consistency (ICC, C, 1), and Agreement (ICC, A, 1) for each 128 repetition (Shrout and Fleiss, 1979, de Vet et al., 2006). Values above 0.8 are considered 129 good to high (Portney and Watkins, 2000). Agreement between both measurement systems 130 was additionally analysed using 95% limits of agreement (Bland and Altman, 1986).

131	RESULTS

132 An initial cohort of 116 subjects (21 men) registered for the study. Of these, 70 persons were 133 available for screening by telephone. A further 51 were excluded by not fulfilling inclusion 134 criteria. Nineteen subjects (four men) were included (mean age 29.2yrs SD: 10.3; neck pain 135 duration median 3yrs (interquartile range 1.25-5.5yrs). Further sample characteristics are 136 presented in Table 1. 137 The CROM™ and G4 measured almost identical ROM values, recording UCS E of -33°± 138 8.4 (mean ± sd) (CROM) and -32° ± 8.5° (G4), and 13 ±4.5 (both devices) for UCS F. All 139 ROM data are presented in Table 1. Based on the strong similarities between measurements 140 derived from both devices, data for CROM™ will be presented further. Comparison data 141 between the devices (Appendix A), and correlation between G4 ROM and NDI (Appendix B) 142 are included as supplementary files. 143 A Correlation-Matrix between the CROM™ assessments and the NDI is presented in Figure 144 4. UCS E-F had a fair correlation to NDI-total score. The strongest (fair to moderate/good) 145 association to UCS range of motion was shown between UCS flexion, and NDI-headache. A 146 decreased ROM in UCS F is associated with an increased score for NDI headache (CROM r=-0.62) (Figure 4). Comparing the UCS and CS E-F ROM showed fair relationships. CS E-F 147 148 showed little correlation with NDI-total score, and fair correlation with NDI-headache.

149 **DISCUSSION**

We employed both a clinic-friendly goniometric device (CROM), and an electromagnetic device (G4) typically used in the laboratory setting, to examine upper and total cervical extension-flexion ROM in 19 subjects with neck pain.

Our results showed that in subjects with non-specific neck pain, UCS E-F ROM has little to fair relationship with ROM of the whole CS. The strongest correlation (fair) occurred between

UCS F, and CS E (r=-0.49, Figure 4). Rudolfsson et al. showed reduced UCS E and lower

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156	CS F in subjects with chronic neck pain, highlighting intra-regional differences in motion of
157	the cervical spine (Rudolfsson et al. 2012). These results may confirm a biomechanical
158	difference between the upper and lower cervical spines, and support a need for separate
159	assessment of UCS E-F ROM in neck pain to enable improved treatment specificity.
160	UCS F is positively related to deep flexor motor control in asymptomatic subjects (Falla et al.,
161	2003). Impaired deep flexor motor control in turn, is associated with increased headache in
162	cervicogenic headache patients (Jull et al., 2002, Jull et al., 2008b). Investigations using the
163	flexion-rotation test to target rotational ROM in the UCS have reported strong associations
164	with headache (Hall and Robinson, 2004, Ogince et al., 2007, Smith et al., 2008). We believe
165	our study is the first to reveal that decreased UCS ROM has a fair (for extension) and
166	moderate/good (for flexion) relationship with an increased NDI headache score. Zito et al.
167	reported reductions of UCS E-F ROM in cervicogenic headache patients compared to
168	asymptomatic controls and migraine subjects. Converse to our results, they found stronger
169	discriminatory validity of CS E-F ROM (Zito et al., 2006). Our findings support the
170	pathoanatomical model for cervicogenic pain as proposed by Bogduk and Jull (1994), and
171	suggest benefit in objectively testing UCS E-F in patients with secondary headache of
172	cervical origin (International Headache Society, 2013) in order to determine regional
173	specificity for treatment direction .
174	Associations between the total NDI score were in general stronger towards Upper cervical
175	spine ROM compared to cervical spine ROM (Figure 4). No study has previously examined
176	associations between UCS E-F ROM and the NDI. Cramer et al. reported fair correlations
177	with ROM of the whole cervical spine in a large cohort of acute to chronic neck pain subjects
178	(Cramer et al., 2014). Kwak et al. reported little to no correlation for CS flexion, and fair
179	correlations for CS extension in small sample of mildly disabled elderly (Kwak et al., 2005).
180	Our results of cervical spine extension-flexion are in line with these studies. Further
181	investigation that specifically targets the upper cervical spine range of motion in relation to
182	neck pain and/or headache appears warranted in confirming our findings.

We measured mean values of 13°UCS F and 33°UCS E (Table 1). Studies using similar measurement protocols to examine asymptomatic controls showed less UCS extension (Dhimitri et al., 1998, Amiri et al., 2003), and less (Dhimitri et al., 1998) or similar UCS flexion (Amiri et al., 2003). Our UCS E values might be greater compared to those by Dhimitri et al. and Amiri et al. due to procedural inequities where these investigators manually blocked lower cervical motion, while we limited thoracic spine movement. In clinical reality, it might be difficult to isolate absolute upper cervical spine motion in the absence of contributions from the lower CS. Studies using videofluoroscopy showed that cervical segments aren't moving consecutively to end of range but instead show varying contributions during a movement cycle (Wu et al., 2007, Wu et al., 2010). During manual blocking, later occurring movements of the UCS might remain undetected. Our procedure in contrast might overestimate EOR movement by not limiting ongoing motion down the CS. Range variability reported between these studies may reflect normative heterogeneity in selected samples from various origins. Increased UCS E in neck pain subjects seems unlikely to occur, as Rudolfson et al. measured "reduced" average values of 40°UCS E in a chronic female neck pain sample compared to the control group (Rudolfsson et al., 2012). Their results are not directly comparable to ours primarily because they used a different testing procedure in free sitting without restricting ROM (Rudolfsson et al., 2012). Our UCS F results may be ranked at the lower limit of reported reference values of 15-25° (White and Panjabi, 1990, Ordway et al., 1999, Bogduk and Mercer, 2000). It is probable that our testing procedure at the wall, limits secondary movements like retraction that typically contribute to UCS flexion. Future studies should investigate the validity of this and other measurement protocols that use different blocking motion-limiting methods to isolate the upper cervical spine.

Limitations

Our subjects showed in general only mild disability (Table 1) (Vernon and Mior, 1991,

MacDermid et al., 2009) which may limit its generalizability towards more disabled subjects.

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Criterion validity has not been examined in our study. Our results demonstrate the comparability and exchangeability of the CROM, and G4, in subjects with non-specific neck pain, and in measuring CS and UCS extension-flexion. Perhaps the safest skeletal-surface imaging to act as a 'criterion' to validate our methods would be MRI in an upright posture, which should be considered for further investigations.

Correlations do not allow causal relationships between UCS E-F-ROM, and disability or headache. Our sample size was too small for detailed data analysis of additional interacting variables.

Future studies should use case-control designs to examine the capability of UCS range of motion to discriminate between healthy subjects and symptomatic patients. Longitudinal studies should examine the responsiveness of UCS range of motion towards treatment

interventions.

CONCLUSION

Upper cervical flexion shows moderate, and extension fair, correlation with headache frequency and intensity. Higher levels of headache are associated with less UCS flexion. Relationships between cervical spine extension-flexion, and neck pain or disability, are weaker than those for the upper cervical spine. The need for a separate extension and flexion ROM assessment for the upper cervical spine has been supported. Using a common procedure, the CROM ™ and the Polhemus G4 achieve similar results in measuring upper cervical extension-flexion in patients with neck pain.

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354 355 356	Table 1: Characteristics of included subjects: NDI= Neck disability index, CS= cervical spine, UCS= upper cervical spine, E= extension, F= flexion, CROM™= Cervical Range of Motion device, G4= electromagnetic tracking device. Values are: means (SD; or otherwise indicated).
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358	Figure 1: Set-up with neutral upper cervical position,
359	Figure 2: Upper cervical spine extension (UCS-E)
360	Figure 3: Upper cervical spine flexion (UCS-F)
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362 363 364 365 366	Figure 4: Correlation Matrix of ROM (CROM™) and NDI variables: UCS = upper cervical spine, CS= cervical spine, E=Extension, F=Flexion NDI= Neck Disability index, NDI headache= frequency and intensity of headache, NDI pain= neck pain intensity. Values are Pearson's product moment correlations with 95% Confidence intervals in brackets. Extension expressed in negative values; Flexion in positive (Friendly et al. 2002)
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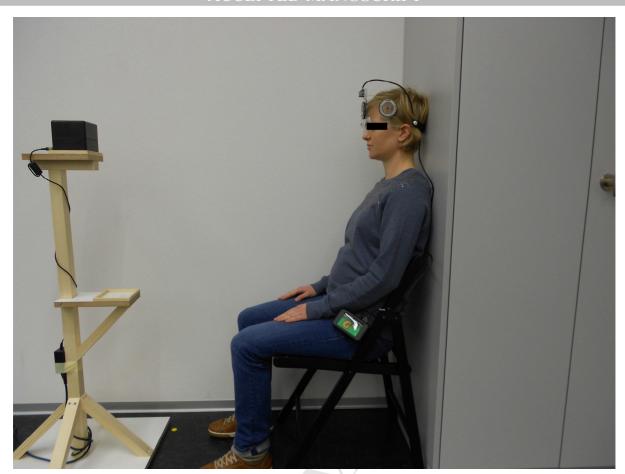
We thank all patients for their participation in the study.

Table 1

Table 1				
Variable	Statistic			
n	19			
Age (years)	29.16 (10.26)			
Duration in years	3y (1.25-5.5y) median (iqr)			
Gender	Female: male: 15:4 (proportion)			
Height (cm)	170 (8)			
Weight (kg)	64 (10)			
NDI%	23 (8)			
NDI pain item (0-5)	1.5 (0.7)			
NDI headache item (0-5)	2.6 (1.4)			
UCS E (CROM™and G4)	-33° (8.4) and -32° (8.5)			
UCS F (CROM™ and G4)	13° (4.5) and 13° (4.5)			
CS E (CROM™ and G4)	-74° (18.2) and -76° (18.6)			
CS F (CROM™ and G4)	60°(10.6) and 60° (9.8)			







Correlation Matrix CROM and NDI

UCS-E

					UCS-F	-0.53 (-0.79,-0.10)
				NDI	-0.31 (-0.67,0.17)	0.44 (-0.02,0.74)
			NDI Headache	0.84 (0.62,0.94)	-0.62 (-0.84,-0.23)	0.47 (0.02,0.76)
		NDI-pain	0.25 (-0.23,0.63)	0.34 (-0.13,0.69)	0.25 (-0.23,0.63)	-0.04 (-0.48,0.42)
	CS-E	0.01 (-0.45,0.46)	0.37 (-0.10,0.71)	0.19 (-0.29,0.60)	-0.49 (-0.77,-0.05)	0.31 (-0.17,0.67)
CS-F	-0.57 (-0.81,-0.16)	-0.01 (-0.46,0.45)	-0.35 (-0.69,0.13)	-0.12 (-0.55,0.35)	0.21 (-0.27,0.61)	-0.33 (-0.68,0.15)

Extension and flexion in the upper cervical spine in neck pain patients

Highlights

- Upper cervical extension-flexion correlates fair to moderate/good to headache intensity/frequency
- The more headache the less upper cervical flexion ROM
- The two measurement instruments (CROM and Polhemus G4) used, achieve similar results

Test Trial		ICC2(C,1)	ICC2(A,1)	Mean Difference	
		95% CI	95% CI	(ULA to LLA)	
UCS Ext.	Trial 1	0.95 (0.85-0.98)	0.95 (0.88-0.98	0.58°(6.01° to -4.85°)	
	Trial 2	0.94 (0.79-0.97)	0.94 (0.83-0.98)	1.10°(6.91° to -4.70°)	
	Trial 3	0.96 (0.90-0.98)	0.96 (0.92-0.98)	1.00°(6.12° to -4.12°)	
UCS Flex	Trial 1	0.75 (0.47-0.95)	0.74 (0.43-0.94)	-1.21° (5.83° to -8.25°)	
	Trial 2	0.82 (0.60-0.94)	0.83 (0.58-0.93)	0.16°(5.42° to -5.10°)	
	Trial 3	0.85 (0.67-0.94)	0.86 (0.70-0.95)	-0.11°(5.82° to -6.03°)	
CS Ext	Trial1	0.99 (0.95-1)	0.99 (0.96-1)	-1.26°(4.35° to -6.87°)	
	Trial 2	0.98 (0.91-0.99)	0.97 (0.93-0.99)	-2.79°(4.41° to -9.99°)	
	Trial 3	0.99 (0.94-0.99)	0.98 (0.95-0.99)	-2.47°(3.43° to -8.38°)	
CS Flex	Trial 1	0.97 (0.88-0.99)	0.97 (0.91-0.99)	0.11°(4.62° to -4.41°)	
	Trial 2	0.97 (0.90-0.98)	0.97 (0.89-0.98)	-0.05°(5.54° to -5.65°)	
	Trial 3	0.98 (0.95-0.99)	0.98 (0.96-0.99)	-0.32°(5.48° to -4.85°)	

Appendix A: Validity of measuring with the CROM™ vs. the Polhemus G4 tracking device: ICC= Intra-class correlation coefficient. C= consistency, A= agreement, 95% CI= 95% Confidence interval, UCS= upper cervical spine, CS= cervical spine, Mean Difference: Polhemus G4 – CROM™, ULA= Upper limit of agreement, LLA= Lower limit of agreement

Correlation Matrix Polhemus G4 and NDI

U	CS	3-1	Е

0.44						
-0.41 (-0.73,0.05)	UCS-F					
0.50 (0.06,0.78)	-0.44 (-0.75,0.02)	NDI				
0.47 (0.01,0.76)	-0.73 (-0.89,-0.41)	0.84 (0.62,0.94)	NDI Headache			
-0.05 (-0.49,0.42)	0.12 (-0.35,0.55)	0.34 (-0.13,0.69)	0.25 (-0.23,0.63)	NDI-pain		
0.19 (-0.29,0.59)	-0.34 (-0.69,0.14)	0.20 (-0.28,0.60)	0.34 (-0.14,0.69)	0.02 (-0.44,0.47)	CS-E	
-0.30 (-0.66,0.18)	0.22 (-0.26,0.61)	-0.18 (-0.58,0.30)	-0.39 (-0.72,0.07)	0.02 (-0.44,0.47)	-0.55 (-0.80,-0.12)	CS-F

Appendix B: Correlation Matrix of ROM (G4) and NDI variables: UCS = upper cervical spine, CS= cervical spine, E=Extension, F=Flexion NDI= Neck Disability index, NDI headache= frequency and intensity of headache, NDI pain= neck pain intensity. Values are Pearson's product moment correlations with 95% Confidence intervals in brackets. Extension expressed in negative values; Flexion in positive (Friendly et al. 2002)