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Prevalence and factors associated with musculoskeletal complaints and disability in individuals with brachial plexus injury: a cross-sectional study

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ABSTRACT

Purpose: (1) To determine the prevalence of musculoskeletal complaints (MSCs) in the non-affected bodily structures in individuals with brachial plexus injury (BPI) and (2) to analyse factors associated with MSCs and disability.

Methods: Survey among individuals with BPI and a control group. Multivariable logistic and linear regression analyses were used to identify factors associated with MSCs or disability.

Results: Forty-nine percent of individuals (34/70) with BPI experienced MSC, which was not significantly different from controls (35%, $n = 40/113$). Complaints were most often located in high back (OR = 3.6) or non-affected limb (OR = 2.2) or neck (OR = 2.1). Greater disability was associated with the presence of MSC in individuals with BPI (OR = 1.1, 95% confidence interval (95% CI) = 1.0; 1.1). Those with no or a low level of education ($B = -10.2$, 95% CI = –19.6; –1.4), a history of nerve surgery ($B = 11.1$, 95% CI = –0.2; 20.9), and moderately affected active range of motion (AROM) of the affected limb ($B = 20.7$, 95% CI = 8.8; 31.0) experienced most disability. Individuals with severely affected AROM showed a wide range of experienced disability.

Conclusions: Clinicians should be aware that almost half of individuals with BPI have MSCs in the non-affected bodily structures, which was associated with increased disability.

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Brachial plexus; musculoskeletal pain; disability; prevalence; upper extremity

➤ IMPLICATIONS FOR REHABILITATION

- Musculoskeletal complaints of the non-affected limb, back and neck are common among individuals with brachial plexus injury, and are associated with more disability.
- Disability was associated with loss of active range of motion (AROM) in the affected limb, although there was a wide variation in experienced disability among individuals with no or a very limited AROM.

Introduction

Brachial plexus injury (BPI) may occur during birth, i.e., brachial plexus birth injury (BPBI) or may be acquired during life as a result of a trauma, cancer, or has a iatrogenic origin. BPI causes disability because of diminished motor and sensory function, but previous studies showed that also age, work status, time since the onset of injury, pain catastrophising, and pain were associated with more disability in individuals with upper extremity impairment [1,2].

Many individuals with BPI experience pain [3–6]. In addition to disability, the presence of pain, neuropathic, or musculoskeletal, impacts the quality of life of individuals with BPI [6,7]. Musculoskeletal complaints (MSCs) are defined as complaints in muscles or joints that are not caused by a trauma or a systemic disease [8]. Risk factors for the development of MSCs are static muscle contractions, forceful and repetitive movements and working in awkward positions for prolonged durations [9,10]. Furthermore, gender, age, marital status, education level, self-reported fitness, and low co-worker support are associated with development of MSCs [11–13]. Besides these physical and social factors, psychological factors like coping style and mental health may also be associated with MSCs following the biopsychosocial model [14]. Approximately, one-third of the general population experiences MSCs of the arm, neck, and shoulder [8]. Individuals with only one upper limb, due to a congenital reduction impairment or acquired amputation, experience twice as many MSCs compared with the general population [15]. Individuals with one fully active limb probably compensate for the loss of activity by increasing the load and number of repetitions on the non-affected limb. They also make compensatory movements with the remnants of the affected limb, non-affected limb and trunk, in order to perform daily and work-related tasks [16–19]. These compensation strategies may explain why individuals with one active

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limb are more susceptible to MSCs of the upper limbs, neck, and back [20,21]. Individuals with BPI generally have diminished activity in their affected upper limb, which could increase their susceptibility to MSCs in non-affected bodily structures as well. About half of individuals with a BPBI have MSCs in the upper limbs, neck, and/or knees [4]. MSCs are observed in individuals with acquired BPI too, but literature substantiating this finding and associated factors is lacking.

Knowledge about the prevalence of and factors associated with MSCs in individuals with BPBI and BPI acquired during life as result of a trauma or other cause may help in the prevention and treatment of MSCs, and thus decrease pain and increase quality of life.

This study aims (1) to assess the point and year prevalence of MSCs in the non-affected bodily structures in individuals with BPI, and to compare it with the prevalence in a control group and (2) to analyse factors associated with MSCs and disability in individuals with BPI.

Methods

Design and participants

For this cross-sectional study, a convenience sample of participants was recruited from three outpatient rehabilitation centres in the northern region of the Netherlands in 2015. At all the centres, a database search for individuals was performed using codes from the International Classification of Disease (ICD-9) for BPI and injury to the brachial plexus due to birth trauma. At one of the centres, an additional free text search for “brachial plexus” and “Erb’s palsy” was carried out in the digital correspondence related to consultations in the centre. A search of this kind was not possible at the other centres due to technical limitations.

The eligibility criteria were having a BPI (defined as injuries proximal to the branches but distal to the nerve roots), being aged 18 years or older, the date of injury being more than one year ago and having a good understanding of written Dutch. Persons with a BPI due to neuralgic amyotrophic were excluded. The control group consisted of acquaintances of the investigators and was largely assembled for a study of transverse upper limb reduction impairment and amputation [14]. However, extra controls were recruited among acquaintances of the researchers to match the age distribution per decade and gender distribution of the patient group. STROBE guidelines were followed to report this cross-sectional study [22].

Survey

A survey used previously [15] and adapted to the current population was sent out by postal mail, it contained 69 questions and assessed five domains:

(1) Participant characteristics, including date of birth, gender, marital status, highest level of education, comorbidity, employment status, working hours and self-perceived work quantity and quality (only for individuals with paid work; answered on an 11-point Likert scale, where 0 = unable to work/delivered work was of very bad quality and 10 = normal quantity or quality).
(2) BPI-related data, including cause of BPI, date of injury, and affected limb. The survey also inquired whether individuals had had surgery on nerves or muscles of the affected limb, and about the locations of sensibility loss and pain in the affected limb. The Dutch language version of Disability of Arm, Shoulder and Hand (DASH-DLV) was included to inquire about functioning. The DASH-DLV contains 30 questions in categories of ability to perform activities and symptoms which were scored on a five-point Likert scale (1 = no difficulties to perform, 5 = unable to perform). Higher scores indicate greater disability. The DASH-DLV is reliable and valid for assessing disability and symptoms [23]. The active range of motion (AROM) of the affected limb was assessed using a self-developed rating scale, which was based on clinical experience. This scale was used to classify ability to perform 10 different movements of the upper limb on a four-point Likert scale (0 = no difficulties in performing movement; 1 = can perform but with some difficulty; 2 = cannot perform complete movement; 3 = cannot move at all). The movements were abduction and external rotation of the shoulder; flexion and extension of the elbow; flexion and extension of the wrist; flexion, extension and abduction of the fingers; and thumb opposition. The movements were illustrated using pictures. The sum score of the 10 items was calculated for statistical analysis (range 0–30). Higher scores reflect less AROM of the affected limb. The BPI-related questions were omitted from the survey for controls.
(3) The point and year prevalence of MSCs: the point prevalence was assessed using the following question: “Did you have regular complaints in the muscles, tendons and/or bones during the previous four weeks which were not caused by an accident, sports injury, infection or joint disease?”. Examples of MSCs were added, like epicondylitis and bursitis. Participants were instructed to answer the question for complaints in other body parts than the affected limb, or in case of bilateral plexus injury for other body parts than both upper limbs. They were given a list of body parts and were asked to tick the boxes of the body parts in which they experienced MSCs. Complaints in the affected limb were not assessed because the question did not distinguish between MSCs and neuropathic pain. In order to assess the year prevalence, the question was repeated for complaints lasting a minimum of four weeks in the preceding year.
(4) MSC-related data (assessed for MSCs in other body parts than the affected limb): if the response to point or year prevalence was positive, the survey inquired about the location, type and duration of complaints, the pain score (answered on a Numeric Rating Scale (NRS) from 0 (no pain) to 10 (worst imaginable pain) and the two-item RAND36 pain subscale [24]) and healthcare consumption in the previous year due to the MSCs. Disability due to complaints was assessed using the Pain Disability Index (PDI). The PDI assesses seven domains of daily living. The questions were answered on an 11-point Likert scale (0 = no disability, 10 = completely disabled). The outcomes were summed up to produce a score ranging from 0 to 70. A higher score indicates greater disability [25,26]. The Dutch language version has good cross-cultural validity for measuring disability [25].
(5) Assessment of possible associated factors for MSCs: upper limb work demands were assessed using the seven-item Upper Extremity Work Demands Scale (UEWD), which assesses the demands of handling objects and movements during work [27]. The UEWD has been validated and provides reliable self-reported estimations of upper extremity work demands [28,29]. Items are rated on a four-point Likert scale (1 = none or rarely, 4 = almost always). The range is 7–28. Higher scores indicate higher work demands. The active, avoidance, and support-seeking subscales (7, 8, and 6 items respectively)

from the Utrecht Coping List (UCL) were administered. The UCL has sufficient internal consistency and construct validity in the general population [30]. The questions have four-point Likert scale answers (1 = never or rarely, 4 = very often), yielding a total range of 7–28 for an active coping style, 8–32 for an avoidant coping style, and 6–24 for a support-seeking coping style. Higher scores indicate the greater presence of a specific coping style. Participants were asked about their perceived general and mental health using the subscales (both consisting of five items) of the RAND36 [24]. The outcomes of the RAND36 subscales, including the pain score mentioned previously, were transformed to a 0–100 scale. Higher scores indicate better health condition.

Data were collected from July 2013 to July 2015. The Medical Ethics Committee of the University Medical Centre Groningen decided that no formal approval was needed (METC file number: METc 2013/199). Every individual signed written informed consent before answering the survey and was sent a 10 euro voucher after returning the survey. All procedures followed were in accordance with the Helsinki Declaration of 1975, as revised in 2000.

Statistical analysis

Returned surveys were checked for duplicates. The data were checked for missing values. Cases with missing values for the point and year prevalence of MSCs were excluded from all the analyses. Missing values for DASH-DLV and RAND36 were handled in line with the manual for the respective questionnaire [24,31]. For the PDI, UEWD and UCL active, avoidant and support-seeking subscales, a maximum of three, one, one, two, and one missing items respectively were allowed and corrected for. Correction consisted in imputing the average of the answered items.

SPSS for Windows (version 23.0.0; SPSS Advanced Statistics, Chicago, IL, USA) was used for data analysis. Continuous data were checked for normality by visually examining QQ plots and interpreting skewness and kurtosis statistics (both = <1 was considered as a normal distribution) [32]. Categorical data were analysed using a Chi-squared test and the odds ratios were calculated. The effect size (w) was calculated by taking the squared root of the Chi-square value divided by the study size [30]. Normally distributed continuous data and homogeneity of variances (checked with Levene's statistic), were analysed using an independent sample t-test. The test value (t) and 95% confidence interval (95% CI) were calculated [32]. If the data were not normally distributed or if variances were inhomogeneous, a Mann–Whitney's U test was used and the test statistic (U) and effect size (r) were calculated. The effect size (r) was calculated by dividing the Z score by the squared root of the study size [32]. A p value of <.05 was considered to be statistically significant.

Factors associated with the presence of MSCs were determined using multivariable logistic regression analysis, using a backward procedure. The variable point prevalence was used, as several dependent variables inquired about a situation in the previous four weeks, e.g., the RAND36 Mental Health Subscale. First, all the variables shown in Table 1 were univariately analysed (Tables 2 and 3) and those with a p value <0.1 were selected for multivariable analysis. Continuous variables were checked for linearity; if no linearity was found, the variable was entered either into four groups based on quartiles or into groups based on biological or clinical relevance. The backward procedure was used, excluding the variable with the highest p value, until all the remaining variables had a p value <0.1 [33]. Factors associated with disability in individuals with BPI were examined using multivariable linear regression analysis. Residuals of the DASH outcome variable score were checked for normal distribution. Continuous variables were checked for linearity and recoded the same way as in the logistic regression if no linearity was found. The aforementioned procedure regarding univariate and multivariable analysis using a

Table 1. Characteristics of the study population.

Characteristic	BPI ($n = 70$)	Controls ($n = 113$)
Demographic data		
Gender, male	46 (66)	72 (64)
Age (years)	53.5 ± 15.5	50.4 ± 15.5
Level of education		
None/low	35 (50)	16 (14)
Medium/high	34 (49)	96 (85)
Marital status		
Single	15 (21)	12 (11)
Living together or married	52 (74)	90 (80)
Divorced or widowed	3 (4)	11 (10)
Employed	36 (51)	78 (69)
Coping styles		
UCL: active	19.5 ± 3.4	20.5 ± 3.6
UCL: avoidance	16.7 ± 3.3	15.5 ± 3.6
UCL: support seeking	11.7 ± 3.4	13.7 ± 3.3
Health related data		
RAND36: general health (median (IQR))	70.0 (52.5, 85.0)	75.0 (60.0, 90.0)
RAND36: mental health (median (IQR))	78.0 (68.0, 87.0)	84.0 (76.0, 88.0)
Presence of comorbidity	22 (31)	22 (20)
Joint diseases	10 (14)	2 (2)
Heart/vascular diseases	5 (7)	10 (9)
Diabetes mellitus	2 (3)	5 (4)
Pulmonary diseases	5 (7)	6 (5)
Other[a]	11 (17)	13 (12)
BPI related data		
Cause of BPI		NA
BPBI	7 (10)	
Trauma	51 (73)	
Tumour	3 (4)	
Radiation	2 (3)	
Other[b]	7 (10)	
Side of BPI		NA
Right	27 (39)	
Left	39 (56)	
Bilateral	4 (6)	
Time since BPI (years) (median (IQR))	9.0 (4.0, 19.0)	NA
Age at BPI (years) (median (IQR))	40.0 (20.5, 54.5)	NA
Sensory loss in affected hand/arm		
No sensory loss	17 (24)	NA
Sensory loss in affected hand	24 (34)	NA
Sensory loss in affected hand and arm	29 (41)	NA
Pain in affected hand/arm		
No pain	33 (47)	NA
Pain in affected hand	21 (30)	NA
Pain in affected hand and arm	16 (23)	NA
Nerve surgery	21 (30)	NA
Muscle surgery	11 (16)	NA
Self-assessed AROM (median (IQR))	10.0 (4.0, 22.8)	NA
DASH score (median (IQR))	29.4 (19.3, 41.5)	NA

AROM: active range of motion; BPI: brachial plexus injury; BPBI: brachial plexus birth injury; DASH: Disability of Arm, Shoulder and Hand; IQR: inter quartile range; NA: not applicable; UCL: Utrecht Coping List. Data are presented as n (%) or mean ± SD, unless otherwise stated.
Missing values in the BPI-group: one for level of education, presence of comorbidity, time since BPI, age at BPI and muscle surgery; two for DASH scores and RAND36 subscale mental health; 5 for RAND36 subscale general health and 6 for self-assessed AROM. Missing values in the control group: one for level of education, RAND36 subscale general health and RAND36 subscale mental health.
[a]Other comorbidities mentioned were allergies, headache, Parkinson disease, and psychological disorders.
[b]Other causes of BPI were related to surgery ($n = 3$), anaesthetics ($n = 1$), accident with knife ($n = 1$), "wrong treatment in hospital" (no further explanations provided) ($n = 1$) and working with a chainsaw (no further explanations provided) ($n = 1$).

Table 2. Univariate analyses between individuals with and without MSCs during the previous four weeks among individuals with BPI.

	BPI with MSCs ($n = 28$)	BPI without MSCs ($n = 42$)	Sig.	Test statistic and effect sizes[a]
Demographic data				
Gender, male	17 (61)	29 (69)	0.472	$\chi^2 = 0.5$, OR = 0.7
Age (years)	57.8 ± 13.3	54.0 ± 16.9	0.263	$t =$ 1.1, 95% CI of t [−3.2, 11.5]
Education level			0.256	$\chi^2 = 1.3$, OR = 1.8, $w = 0.15$
No/low	16 (57)	19 (45)		
Medium/high	11 (39)	23 (55)		
BPI related data				
Time since BPI (years) (median (IQR))	11.0 (5.0, 33.0)	7.5 (3.8, 17.0)	0.064	$U = 416.5$, $r = -0.22$
Cause of BPI				
Trauma	17 (61)	34 (81)		
BPBP	4 (14)	3 (7)		
Other	7 (25)	5 (12)		
Self-assessed AROM (median (IQR))	27.0 (13.0, 30.0)	7.0 (3.0, 10.5)	<0.001*	$U = 160.0$, $r = -0.52$
DASH score (median (IQR))	41.3 (30.6, 55.4)	20.7 (10.1, 30.0)	<0.001*	$U = 202.5$, $r = -0.54$
Pain in the affected arm and/or hand	19 (68)	18 (43)	0.020*	$\chi^2 = 5.4$, OR = 3.3, $w = 0.30$
Loss of sensibility in the affected arm and/or hand	19 (68)	34 (81)	0.409	$\chi^2 = 0.7$, OR = 0.6, $w = 0.11$
Nerve surgery	10 (36)	11 (23)	0.309	$\chi^2 = 1.0$, OR = 1.7. $w = 0.12$
Muscle surgery	3 (11)	8 (19)	0.379	$\chi^2 = 0.8$, OR = 0.5, $w = 0.11$
Health related data				
RAND36: general health (median (IQR))	65.0 (40.0, 76.3)	75.0 (60.0, 90.0)	0.034*	$U = 349.5$, $r = -0.26$
RAND36: mental health (median (IQR))	72.0 (60.0, 88.0)	84.0 (70.0, 86.0)	0.085	$U = 417.0$, $r = -0.21$
Presence of comorbidity	10 (26)	12 (29)	0.461	$\chi^2 = 0.5$, OR = 1.5, $w = 0.09$
Coping styles				
UCL: active	19.1 ± 3.8	19.9 ± 3.2	0.361	$t = 0.9$, 95% CI of t [–0.9, 2.4]
UCL: avoidance	16.7 ± 3.1	16.8 ± 3.5	0.965	$t = -0.04$, 95% CI of t [–1.6, 1.7]
UCL: support seeking	11.0 ± 2.6	12.3 ± 3.8	0.129	$t =$ 1.5, 95% CI of t [0.4, 2.9]
Work related data				
Employed	13 (46)	23 (55)	0.494	$\chi^2 = 0.5$, OR = 0.7, $w = 0.09$
UEWD (median (IQR))	15.0 (13.0, 18.0)	12.0 (10.0, 17.0)	0.282	$U = 42.0$, $r = -0.23$

AROM: active range of motion; BPBP: brachial plexus birth palsy; BPI: brachial plexus injury; CI: confidence interval; DASH: Disability of Arm, Shoulder and Hand; IQR: inter quartile range; MSCs: musculoskeletal complaints; UCL: Utrecht Coping List; UEWD: upper extremity work demands.

Data are presented as n (%) or mean ± SD, unless otherwise stated. Variables were univariately analysed without correcting for possible confounders. Missing values in the MSC-group: one for education level; one for gender and time since BPI; five for self-assessed AROM; two for RAND36 subscale general health and one for RAND36 subscale mental health. Missing values in the non-MSC-group: one for self-assessed AROM; one for muscle surgery; two for RAND36 subscale general health; one for RAND36 subscale mental health, two for DASH score.

*Statistically significant at $p < 0.05$.

[a]For analyses of categorical data, Chi-square statistic (χ^2) odds ratio (OR) and effect size w are presented. All Chi-squared tests had one degree of freedom. For continuous data without a normal distribution, the Mann–Whitney's U statistic and effect size r are presented. For continuous data with a normal distribution, the t statistic and a 95% CI of t are presented.

backward selection procedure was used for these analyses too, except that the "paid work" variable was excluded, as it was considered most likely to be a consequence of disability. A "point prevalence of MSCs" variable was also added.

Results

A total of 190 questionnaires were sent out; 22 were returned "addressee unknown" and 89 questionnaires were not returned for unknown reasons, a one-time reminder was sent. Seventy-nine questionnaires were eventually returned, of which five were found to be unusable. A further four responses were omitted because of too many missing values. These related to three women (aged 75, 77, and 88 years) and one man (aged 57 years). Three had experienced MSCs in the previous year while one failed to answer this question. Seventy individuals (response rate: 39%) with BPI were thus included in the analyses (Table 1), with 113 controls. Comorbidities were frequently present in both individuals with BPI and controls, but the comorbidity "joint diseases" was more often present in individuals with BPI (Table 1). Furthermore, the education level differed between the groups, individuals with BPI were more often not or low educated compared to controls.

Presence of MSCs

Thirty-seven individuals with BPI reported MSCs during the previous year, of which three reported MSCs in the affected limb only, despite instructions to exclude this limb when reporting MSCs. Thus, 34 individuals with BPI were included in analyses regarding presence of MSCs. In individuals with MSC, the comorbidity joint diseases was present in three (9%) individuals with BPI and two (5%) controls.

The year and point prevalence of MSCs tended to be higher among individuals with BPI than in the control group, but differences were not significant (Table 4). When MSCs were present, individuals with BPI experienced more frequent complaints in the neck, high back, and non-affected limb as well as more pain compared with the control group. Furthermore, the healthcare consumption of individuals with BPI was higher if MSCs were present. The type of complaint most frequently reported was pain (reported by 23 (68%) individuals with BPI who had experienced MSCs in the previous year), while stiffness, weakness, and tingling were reported by 17 (50%; of which two individuals also had joint diseases), 14 (41%), and 10 (29%) individuals, respectively (three missing values). These complaints were most often located in the neck and the non-affected limb. In the non-affected limb, the shoulder ($n = 14$) and elbow ($n = 9$) were most often affected. Among individuals with BPBI, the neck ($n = 4$), higher back ($n = 2$), lower back ($n = 3$), and non-affected limb ($n = 1$) were the most affected.

The point prevalence of MSCs did not differ between individuals with a left-sided (13/39, 33%) and right-sided BPI (12/27, 44%) ($\chi^2 = 1.1$, df = 1, $p = 0.587$, $w = 0.13$).

Four individuals with BPI were bilaterally affected; two of them had experienced complaints during the previous year (both had experienced MSCs of the back and responded,

Table 3. Univariate linear regression analyses of factors associated with disability assessed by means of the DASH among individuals with BPI.

				95% CI for *B*	
Individuals with BPI ($n=68$)	*B*	SE	Sig	Lower	Upper
Demographic data					
Gender	4.1	5.0	0.398	−5.7	14.4
Age[b]					
Low (18–40 years)	Reference				
Middle (>40–65 years)	7.8	4.8	0.109	−1.8	17.3
High (>65 years)	−3.4	0.3	0.654	−16.2	12.9
Education level[a,*]	−10.4	4.7	0.037	−19.6	−1.2
Marital status[b]					
Single	Reference				
Living together or married	2.9	3.9	0.455	−4.3	10.5
Divorced or widowed	4.7	10.1	0.715	−11.4	21.1
BPI related data					
Time since onset BPI[b,*]					
Q1 (<4 years)	Reference				
Q2 (4–9 years)	−9.3	7.7	0.228	−23.5	6.9
Q3 (9–19 years)	−11.2	5.4	0.052	−21.8	−0.4
Q4 (19–39 years)	−1.7	6.2	0.799	−14.7	11.2
Cause of BPI[a,*]	9.7	5.4	0.074	−0.6	20.2
Self-assessed AROM[b,*]					
Low score (0–10)	Reference				
Medium score (11–20)	−2.8	8.1	0.776	−19.9	11.3
High score (21–30)	46.6	−1.5	0.093	14.6	76.9
Pain in the affected arm and/or hand*	12.2	4.3	0.011	4.1	20.4
Loss of sensibility in the affected arm and/or hand	−2.6	5.7	0.660	−13.1	8.4
Nerve surgery*	9.2	5.1	0.084	−0.5	19.3
Muscle surgery	3.2	6.4	0.588	−9.5	16.7
Health related data					
Rand 36: general health[a,*]	−12.6	4.3	0.010	−21.2	−4.4
Rand 36: mental health[a,*]	−10.1	4.7	0.047	−19.5	0.1
Presence of comorbidity*	14.2	5.0	0.012	4.6	24.6
Presence of MSC (previous 4 weeks)*	20.9	4.2	0.001	11.9	28.9
Coping styles					
UCL: active[b]					
Q1 (0–17)	Reference				
Q2 (>17.0–20.0)	−6.8	6.4	0.292	−19.8	6.0
Q3 (>20.0–22.0)	−1.8	6.2	0.782	−13.8	9.7
Q4 (>22.0)	−5.4	6.7	0.413	−19.8	7.4
UCL: avoidance[b]					
Q1 (0–14)	Reference				
Q2 (>14.0–16.0)	−3.5	9.3	0.746	−19.4	18.4
Q3 (>16.0–20.0)	−9.1	10.0	0.450	−25.6	14.6
Q4 (>20.0)	12.0	20.6	0.622	−23.2	56.4
UCL: support seeking[b]					
Q1 (0–9.9)	Reference				
Q2 (>9.9–11.5)	−37.0	23.9	0.278	−76.9	1.6
Q3 (>11.5–13.3)	−40.1	24.5	0.274	−79.6	−1.3
Q4 (>13.3)	−37.1	23.0	0.436	− 67.5	−6.7

AROM: active range of motion; *B*: regression coefficient; BPI: brachial plexus injury; CI: confidence interval; DASH: Disability of Arm, Shoulder and Hand; MSCs: musculoskeletal complaints; OR: odds ratio; SE: standard error; UCL: Utrecht Coping List; Q: quartile.
Variables were univariately analysed, with 1000 bootstrap samples, as residuals of the DASH score did not show a normal distribution. Missing values: two for the DASH score, one for education level; one for gender and time since BPI; six for self-assessed AROM; four for RAND36 subscale general health and two for RAND36 subscale mental health.
*$p < 0.1$, variable selected for multivariable analysis.
[a]Variables were analysed dichotomous: level of education entered with the categories "no/low" and "medium/high", cause of BPI entered with the categories "trauma" and "other", general health (measured with the Rand 36 subscale general health) no linearity, therefore, added as a dichotomous variable: lower vs. equal or higher than the national mean (=72.2) [17], mental health (measured with the RAND36 subscale) no linearity, therefore, added as a dichotomous variable: lower vs. equal or higher than the national mean (=76.8) [17].
[b]Variables were categorised because of no linearity: age (no linearity, therefore added in three categories: young, middle, and high age), time since onset of BPI (no linearity; therefore, added as a categorical variable based on quartiles), and self-assessed AROM score (no linearity; therefore, added as a categorical variable in three groups; score range 0–10, 11–20, and 21–30).

despite the instruction not to answer the question for their upper limbs, that they experienced pain of both limbs and the neck as well).

Univariate analyses showed that gender and age did not differ between those with and those without MSCs (Table 2). DASH-scores and self-assessed AROM were significantly lower in individuals without MSCs, indicating less disability and a better AROM of the affected limb in individuals without MSCs. Perceived general health, measured using the RAND36 subscale, was significantly lower in individuals with MSCs than individuals without MSCs ($p=0.03$, $r=0.26$). The presence of MSCs was not related to coping style or employment status.

Table 4. Presence and characteristics of MSCs among individuals with BPI and two-handed controls.

	BPI ($n = 70$)	Controls ($n = 113$)	Sig.	Test statistic and effect sizes[a]
Presence of MSCs during the previous four weeks				
Prevalence of all locations combined	28 (40)	32 (28)	0.102	χ^2=2.7, OR = 2.1, $w = 0.12$
Pain score (NRS)	6.0 (3.0, 7.0)	3.0 (2.0, 4.0)	0.001*	$U = 181.0$, $r = -0.46$
RAND36 pain score	57.1 (44.9, 67.3)	67.3 (67.3, 77.6)	0.006*	$U = 220.5$, $r = -0.37$
PDI score	24.5 (17.0, 38.3)	8.5 (4.8, 15.0)	<0.001*	$U = 86.5$, $r = -0.60$
Presence of MSCs during the previous year				
Prevalence of all locations combined	34 (49)	40 (35)	0.078	χ^2=3.1, OR = 2.2, $w = 0.13$
Prevalence of specific locations				
Neck	20 (29)	18 (15.9)	0.040*	χ^2=4.2, OR = 2.1, $w = 0.15$
High back	14 (20)	6 (5.3)	0.002*	χ^2=9.6, OR = 3.6, $w = 0.23$
Low back	11 (16)	10 (8.8)	0.157	χ^2=2.0, OR = 1.8, $w = 0.10$
Non-affected limb[b]	23 (34)	21 (18.8)	0.019*	χ^2=5.5, OR = 2.2, $w = 0.17$
Total number of body parts with MSCs	3.0 (2.0, 4.0)	2.0 (1.0, 3.0)	0.003*	$U = 169.5$, $r = -0.27$
Duration of complaints[c]			0.184	χ^2=6.2, $w = 0.18$
<3 months	7 (21)	13 (33)		
3–12 months	4 (12)	11 (28)		
>12 months	21 (62)	15 (39)		
Healthcare use due to MSCs	21 (57)	9 (23)	0.002*	χ^2=9.7, OR = 4.5, $w = 0.22$

BPI: brachial plexus injury; MSCs: musculoskeletal complaints; NRS: Numeric Rating Scale; PDI: Pain Disability Index.
Data are presented as n (%) or median (IQR). Missing values in the BPI-group: one for healthcare use; one for pain score (NRS); two for total number of complaints and duration of complaints; two for PDI score. Missing values in the control group: one for duration of complaints and healthcare use; five for RAND36 pain score and pain score (NRS); eight for total number of complaints; 10 for PDI score.
*Statistically significant at $p < 0.05$.
[a]For analyses of categorical data Chi-square statistic (χ^2), all Chi-squared tests had one degree of freedom, odds ratio (OR), and effect size w are presented, while for continuous data the Mann–Whitney U statistic and the effect size r are presented.
[b]Non-affected limb corresponds with the dominant limb of the controls, while the affected limb corresponds with the non-dominant limb. Four individuals with BPI (of which three experienced MSC) were bilaterally affected and two controls were ambidextrous (of which one experienced MSC); these individuals were omitted from analyses.
[c]Of the individuals with complaints in the affected limb, two had complaints in this limb only.

Disability among individuals with BPI

Univariate analyses of factors associated with disability are shown in Table 3. Individuals with paid work experienced less disability (median DASH score of employed individuals 25.9 (IQR 16.0, 35.1), versus 32.5 (21.2, 54.0) for unemployed individuals; two missing values; Mann–Whitney's U-test 414.5; SE 81.4; $p = 0.047$). Among those who were in employment, disability was not significantly related to work quantity or quality (univariate linear regression with 1000 bootstrap samples, as residuals of the DASH score did not show a normal distribution; work quantity: $B = -2.9$, 95% CI = –6.3; 0.4, $p = 0.078$; work quality: $B = -1.7$, 95% CI = –5.2; 2.1, $p = 0.308$; one missing value).

All variables were univariately analyzed, in order to perform the first step of the backward procedure of the multivariable logistic regression analysis (e.g., including all variables which show a p value of <0.1 after univariate analyses) to examine factors associated with MSCs. In the initial model including self-assessed AROM and DASH, the regression coefficients and related standard errors were extremely large. Probably due to insufficient cell filling, they could not be estimated adequately. Changing self-assessed AROM score into a dichotomous variable did not solve this problem and therefore self-assessed AROM was removed from the model. The association between MSCs, DASH, and self-assessed AROM score is shown in Figure 1.

Multivariable logistic regression analysis using a backward procedure yielded presence of MSCs during the previous four weeks was associated with DASH scores (Table 5).

Linear regression analysis showed that disability was associated with self-assessed AROM score, nerve surgery of the affected upper limb and level of education (Table 6). The explained variance of the final model for disability was 57%.

Discussion

This study is the first study specifically examining factors associated with MSCs in the non-affected bodily structures among individuals with BPI. The results may help clinicians to identify individuals with BPI who are at risk for development of MSCs. Almost half of the individuals with BPI experienced MSCs during the previous year. MSCs were most often located in the neck, high back, and non-affected upper limb. Among individuals with BPI, presence of MSCs was associated with disability. Disability was associated with loss of functionality as depicted by self-assessed AROM score, level of education, and a history of nerve surgery.

In contrast to individuals with upper limb reduction impairment or amputation, the prevalence of MSCs was not significantly higher among individuals with BPI compared to matched controls with two normally functioning upper limbs [15]. However, individuals with BPI, like individuals with upper limb reduction impairment or amputation, did experience more pain due to MSCs (RAND-36 pain score individuals with BPI and upper limb reduction impairment or amputation 53.5 ± 18.4; controls 67.3 ± 10.6) and their healthcare consumption related to these complaints was higher (health care use individuals with BPI and individuals with upper limb reduction impairment or amputation 62.6%; controls 27%). Furthermore, they had MSCs at more bodily locations. The burden caused by the presence of MSCs therefore seems to be higher for individuals with a diminished function in one upper limb.

Individuals with BPI experienced significantly more often MSCs in their non-affected upper limb, compared to the dominant limb of two-handed matched controls, and in their high back and neck. The increased susceptibility to MSCs in these body parts is presumably related to loss of activity in the affected limb and the consequential increased load on the non-affected bodily structures because of compensatory strategies [18,19]. This assumption is supported by our finding that individuals with MSCs reported more decrease in AROM of the affected limb, suggesting a greater loss of activity. It was not possible to assess the association between the presence of MSCs and self-assessed AROM in a logistic regression, as the data did not permit this. The study sample

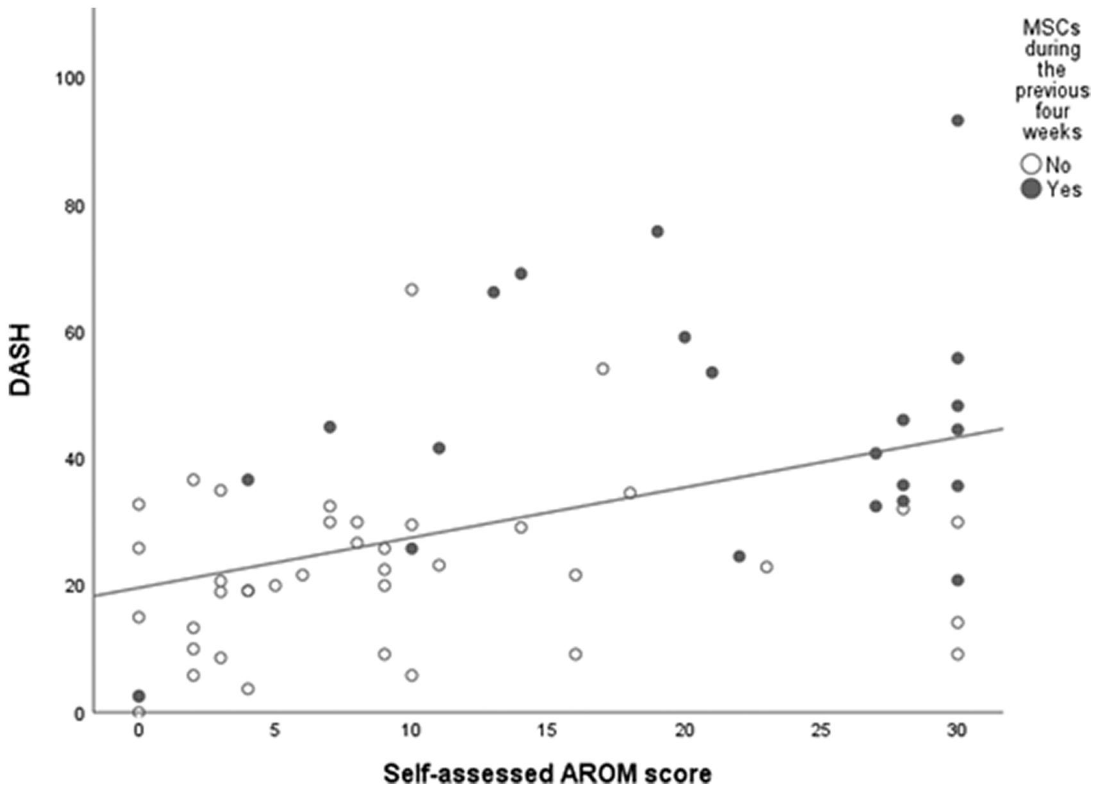

Figure 1. Association between DASH scores, self-assessed AROM score, and presence of MSCs during the previous four weeks among individuals with BPI.
Individuals with lower self-assessed AROM of the affected limb (represented by higher scores) often had MSCs, and generally greater disability (represented by higher DASH scores). Contrarily, individuals with little disability and normal or near to normal self-assessed AROM of the affected limb were often free from MSCs. Missing values: two for DASH score, six for the self-assessed AROM score. Number of individuals in this figure: 62. *Abbreviations*: AROM: active range of motion; BPI: brachial plexus injury; DASH: Disability of Arm, Shoulder and Hand; MSCs: musculoskeletal complaints.

Table 5. Logistic regression with backward selection procedure to statistically predict presence of MSCs during the previous four weeks among individuals with BPI.

					95% CI for OR	
	B	SE	Sig.	OR (Exp(*B*))	Lower	Upper
Time since BPI (per year)	0.04	0.02	0.090	1.0	1.0	1.1
DASH (per point)	0.08	0.02	<0.001*	1.1	1.0	1.1
Constant[a]	–3.4	0.86	<0.001*	0.0		

AROM: active range of motion; *B*: regression coefficient; BPI: brachial plexus injury; CI: confidence interval; DASH: Disability of Arm, Shoulder and Hand; MSCs: musculoskeletal complaints; OR: odds ratio; SE: standard error.
After univariate analyses, the following variables were included in the multivariate logistic analysis: pain in affected arm and/or hand (added as a dichotomous variable: no pain in affected arm and hand vs. pain in affected arm and/or hand), age (no linearity, therefore, added in three categories: young, middle, and high age), mental health (measured with the RAND36 subscale; no linearity, therefore, added as a dichotomous variable: lower vs. equal or higher than the national mean (=76.8) [17]), time since onset of BPI, DASH score and self-assessed AROM (no linearity; therefore, added as a categorical variable with three groups; score range 0–10, 11–20, and 21–30). In the initial model including self-assessed AROM and DASH and the regression coefficients and related standard errors were extremely large. Probably due to insufficient cell filling, they could not be estimated adequately. Changing self-assessed AROM score into a dichotomous variable did not solve this problem and therefore self-assessed AROM was removed from the model. The final model included 67 individuals. A Hosmer–Lemeshow test with 8 degrees of freedom gave a Chi-square of 11.8, with a p value of 0.160.
*Statistically significant at $p < 0.05$.
[a]Constant for MSCs during the previous four weeks is an individual with DASH-score 0 (meaning no disability) and 0 years since occurrence of BPI.

was unfortunately too small to allow for stratification. The relationship between the presence of MSCs and loss of activity has been established previously in a population of individuals with finger or partial hand amputation [34]. In that study, decreased range of motion of the wrist was found to be related to higher risk of MSCs, even though, like in this study, the prevalence of MSCs in the study population did not differ from the prevalence in the general population. Possibly, the loss of activity was not so substantial that individuals with finger or partial hand amputations or with BPI became more susceptible to the development of MSCs. Clinicians are advised to actively inquire about the presence of MSCs, as more than half of the individuals with BPI and MSCs considered it normal to have MSCs, which might lead to underreporting of these complaints.

Loss of activity was also related to disability, represented by the DASH score. Interestingly, only a medium score on self-assessed AROM showed a significant association with disability, while a high score on self-assessed AROM, indicating more severe loss of activity of the affected limb, did not show a significant association with disability. A wide variation in level of disability was found among individuals with a more severe loss of activity of the affected limb. Apparently, a selection of these individuals did not experience much disability, despite their severely affected upper limb, while others did. Possibly those who experience greater impairment, but little disability are better adapted to life with only one fully functioning upper limb, e.g., using more often assisting devices or alternative strategies. Further research should focus on level and type of activity loss and the relation to MSCs and disability.

Multivariable linear regression analysis showed that, besides medium loss of activity, disability was also associated with a history of nerve surgery and no or a low education level. Individuals with no or a low education level often perform more physically demanding work, which may cause them to experience greater disability. Individuals with BPI who had nerve surgery were

Table 6. Linear regression with backward selection procedure to statistically predict disability assessed by means of the DASH among individuals with BPI.

				95% CI for B	
	B	SE	Sig.	Lower	Upper
Constant[a]	25.9	6.9	0.001*	13.8	39.9
Level of education	−10.2	4.6	0.037*	−19.6	−1.4
Pain in affected hand and/or arm	6.5	3.4	0.067	−0.2	12.8
Self-assessed AROM					
Medium score (11–20)	20.7	5.6	0.001*	8.8	31.0
High score (21–30)	9.1	5.7	0.127	−2.3	20.1
Time since onset BPI					
Second quartile (4–9 years)	−3.2	6.5	0.657	−15.8	9.7
Third quartile (9–19 years)	−13.3	6.6	0.056	−26.0	0.1
Fourth quartile (19–65 years)	2.3	7.0	0.756	−11.1	15.8
History of nerve surgery	11.1	5.2	0.041*	−0.2	20.9
RAND36 general health score	−7.6	4.0	0.071	−15.4	0.9

AROM: active range of motion; *B*: regression coefficient; BPI: brachial plexus injury; CI: confidence interval; DASH: Disability of Arm, Shoulder and Hand; SE: standard error.

After univariate analyses, the following variables were added to the initial model: level of education, presence of comorbidity (added as a dichotomous variable), cause of BPI (added as a dichotomous variable with categories "trauma" and "other"), pain in affected arm and/or hand, history of nerve surgery, presence of MSCs during the previous four weeks, age (no linearity, therefore, added in three categories: young, middle, and high age), general health (measured with the RAND36 subscale; no linearity, therefore, added as a dichotomous variable: lower vs. equal or higher than the national mean (=72.2) [17]), mental health (measured with the RAND36 subscale; no linearity, therefore, added as a dichotomous variable: lower vs. equal or higher than the national mean (=76.8) [17]), time since onset of BPI (no linearity; therefore, added as a categorical variable based on quartiles), and self-assessed AROM score (no linearity; therefore, added as a categorical variable in three groups; score range 0–10, 11–20, and 21–30). The final model included 57 individuals. R^2 of the total model was 0.577. Residuals of outcome variable DASH score did not show a normal distribution, neither did a transformation. Therefore, the analysis were performed using bootstrapping. Results are based on 1000 bootstrap samples.

*Statistically significant at $p < 0.05$.

[a]Constant for disability is an individual with the following features: no or low level of education, no pain in the affected arm and hand, a self-assessed AROM score of 0–10 (meaning normal or near to normal AROM of the affected limb), time since the onset of BPI ≤4 years (first quartile), no history of nerve surgery and a RAND36 general health score of 0.

probably more severely affected. Although most individuals with BPI were satisfied with the results of nerve surgery, they still scored high on the DASH, indicating considerable disability [5]. In contrast to previous studies on disability among individuals with BPI or diminished activity of one upper limb, pain and a shorter time since the onset of injury were not associated with greater disability [1,2]. Both factors were included in the final model of the multivariable linear regression analysis, but were not significantly related. This result could be the effect of the relative small sample size, but it could also be the effect of a longer time since the onset of injury of the individuals included in this study. In previous studies, the time since BPI was relatively short (median 14–17 month) so that spontaneous recovery of nerve injury was still relevant, while in our study the time since BPI was much longer (median 11 years) meaning that spontaneous nerve recovery was not likely anymore, the remaining activity of the affected limb might be expected to be more stable and individuals have developed most of their compensatory strategies. It is important to note that 57% of the variance in disability could be explained by the three factors included in the final model, meaning that 43% of the variance in disability is explained by other, yet unknown, factors.

Coping style was not associated with the presence of MSCs nor with disability in individuals with BPI, like in individuals with an upper limb amputation [15]. Coping style was studied in order to study all factors of the biopsychosocial model [14]. Mental health seemed to be lower in individuals with MSCs, but we did not find a statistical significant difference. This is in agreement with individuals with finger or partial hand amputations, but in contrast to individuals with upper limb amputations in whom the presence of MSCs was related to lower mental health [15,34]. The association between mental health and MSCs among individuals with loss of activity of one upper limb therefore remains uncertain and deserves further investigation through a prospective study with a large sample size. Furthermore, a qualitative study regarding the perspectives and ideas of individuals with BPI regarding MSCs, may provide more insight in psychological factors associated with MSCs in individuals with BPI.

Study limitations

In this study, individuals with BPI acquired during life and BPBI were included. Combining both groups is not done regularly, but was possible in our case, since we looked at MSCs in other body parts than the affected upper limb. The 39% response rate is low, but comparable to the 30–60% found in other studies using postal questionnaires [35,36]. It was not possible to investigate the characteristics of the group of non-respondents due to privacy regulations, hence selection bias cannot be ruled out. However, our group of participants seems to reflect other BPI study populations since most of the individuals with BPI were males and sustained their injuries at a young age due to trauma [1–3,37,38].

Individuals with BPI may experience both BPI-related (neuropathic) pain and MSCs in their affected limb. Although we carefully tried to differentiate between the two types of pain in the survey by asking participants not to answer questions on MSCs for their affected upper limb, we cannot rule out the possibility that some individuals reported MSCs whereas they had actually experienced neuropathic pain related to their BPI. It could have been difficult for participants to differentiate between neuropathic pain and MSCs of their neck. Furthermore, nine individuals with MSCs filled out the questions on MSCs also for their affected limb. This has not affected the overall point and year prevalence of MSCs, because all individuals with neck pain and the nine individuals who filled out the questions for their affected upper limb also experienced complaints in other body parts than their neck and affected upper limb. The prevalence of MSCs may have been influenced by not measuring MSCs in the affected upper limb. Also the presence of joint diseases, although only present in three individuals with BPI (9%) and two controls (5%), may have slightly influenced the prevalence of MSCs and the reported type of complaints. Furthermore, a recall bias may have influenced the year prevalence of MSCs.

Our control group consisted of selected individuals, mostly acquaintances of the investigators, and was matched for gender and age. However, the individuals with BPI had a lower level of education. The work carried out by those who have a lower level of education is often more physically demanding than the work carried out by highly educated persons, which could have led to an overestimation of the difference in MSCs between individuals with BPI and controls. Nevertheless, self-reported upper extremity work demands did not differ significantly between individuals with BPI with and without MSCs. Neither did employment status differ.

Existing scoring systems for the capacity of individuals with BPI assess functioning in terms of activity [39,40]. A scoring system for capacity in terms of the physical functioning of the affected limb, such as joint AROM and sensation, is currently lacking. A self-constructed scale was therefore used to assess the AROM of the affected limb. Text and pictures were used to minimise the risk of misinterpretation of the questions. The reliability and validity of this self-constructed scale are unknown, however, and results based on this scale must be interpreted with caution. Development of a reliable and valid scoring system for physical functioning of the affected limb is needed in rehabilitation medicine, because it is important for determining remaining activity, functional possibilities, and risks on MSC.

Conclusions

Individuals with BPI experienced significantly more often MSCs in the non-affected limb, upper back, and neck compared to matched controls, which may be caused by an increased load on these non-affected bodily structures in order to compensate for the loss of activity in one upper limb. Although the overall year prevalence of MSCs was not significantly higher among individuals with BPI compared to able-bodied controls (49 vs. 35%, respectively), the burden of MSCs among individuals with BPI seems to be higher as they perceived MSCs as more painful and MSCs were present at more locations. Presence of MSCs was related to disability and self-assessed AROM of the affected limb. Individuals with no or a low level of education, a history of nerve surgery and moderately affected self-assessed functionality of the affected limb (mildly or severely affected) experienced most disability. Severely affected self-assessed functionality showed a broad variation in disability. Clinicians are advised to actively inquire about MSCs and disability in individuals with BPI.

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