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Recovery priorities of patients with degenerative cervical myelopathy

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SCHOLARONE[™] Manuscripts

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Abstract

Objectives

To establish the recovery priorities of individuals suffering with degenerative cervical myelopathy (DCM).

Design

A cross-sectional, observational study.

Setting

Patients from across the world with a diagnosis of DCM accessed the survey over an 18month period on Myelopathy.org, an international myelopathy charity.

Participants

481 individuals suffering from DCM completed the online survey fully.

Main outcome measures

Functional recovery domains were established through qualitative interviews and a consensus process. Individuals were asked about their disease characteristics, including limb pain (visual analogue scale) and functional disability (patient derived - modified Japanese Orthopaedic Association score). Individuals ranked recovery domains (arm and hand function, walking, upper body/trunk function, sexual function, elimination of pain, sensation and bladder/bowel function) in order of priority. Priorities were analysed as the modal first priority and mean ranking. The influence of demographics on selection was analysed, with significance p<0.05.

Results

Of 659 survey responses obtained, 481 were complete. Overall, pain was the most popular recovery priority (39.9%) of respondents, followed by walking (20.2%), sensation (11.9%) and arm and hand function (11.5%). Sexual function (5.7%), bladder and bowel (3.7%) or trunk function (3.5%) were chosen less frequently. When considering the average ranking of symptoms, whilst pain remained the priority (2.6±2.0), this was closely followed by walking (2.9±1.7) and arm/hand function (3.0 ±1.4). Sensation ranked much lower (4.3±2.1). With respect to disease characteristics, overall pain remained the recovery priority, with the exception of patients with greater walking impairment (p<0.005) who prioritised walking, even amongst patients with lower pain scores.

Conclusions

This is the first study investigating patient priorities in DCM. The patient priorities reported provide an important framework for future research and will help ensure that it is aligned with patient needs.

Strengths

- **Patient-Focussed Research.** The misalignment of researcher and patient objectives has been found to contribute to research wastage therefore recovery domains were agreed using a patient focus group, patients have been involved in all stages and are represented amongst the authors.
- Large survey population, including both patients who have and have not undergone surgical treatment. Not all DCM patients require surgery, but DCM research has largely reported on cohorts of less than 300 surgical DCM patients, therefore the broader sampling in this study is likely more representative.
- Demographic data, including validated outcome tools, allowed subgroup analysis. DCM can cause wide ranging disability, and a representation of this was captured using additional data points, in order to ascertain whether recovery priorities were generalisable.

Limitations

- **Self-selected population.** Patients who do not engage with Myelopathy.org would not have had the opportunity to complete the questionnaire.
- Risk of inclusion of non-patients. Respondents were asked to click through a description of DCM and confirm they had a diagnosis from a medical professional before completing the questionnaire.

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Introduction

Degenerative cervical myelopathy [DCM] has been coined as an umbrella term for degenerative and congenital or acquired conditions of the cervical spine, such as spondylosis or ossification of the posterior longitudinal ligament (OPLL), which lead to symptomatic cord compression.[1] With an estimated prevalence of up to 5% in individuals above 40 years old,[2]·[3] DCM is the most common cause of spinal cord dysfunction worldwide.[1] Given its degenerative aetiology and the rising age of the population, this incidence is expected to rise.[4]

The cervical spinal cord acts as a processor and conduit of information between the brain and the periphery. Its injury can therefore give rise to a range of possible symptoms.[1] These include pain, paraesthesia, weakness, unsteadiness, frequent falls, bladder or bowel dysfunction and impotence in men.[5] At early stages, individual symptoms may occur in isolation, but more typically occur in combination, especially as the disease advances.

At present, decompressive surgery is the only evidence-based treatment for DCM.[6]. Surgical decompression is able to halt the progression of symptoms and offer limited, albeit clinically relevant[7] improvements across a range of domains.[8]·[9] However, due to the limited intrinsic capacity for the spinal cord to repair, most patients do not make a full recovery, and instead suffer lifelong disabilities.[9] As a consequence, unemployment and/or dependency is prevalent amongst individuals with DCM.[4]·[10]·[11] Moreover, a recent study has identified that DCM severely impacts quality of life with recorded SF-36, patient reported outcome scores amongst the lowest of all chronic disease.[12] Improving recovery is therefore a major unmet clinical need in DCM.[13]

Medical research is primarily designed by health care professionals. This bears the risk of not taking into account actual patient needs. The concept of 'research wastage' has emerged to depict healthcare research that does not yield actual or potential clinical benefit. In the 2014 *Lancet* series, Chalmers et al. estimated that as much as 85% of the US\$240 billion expended on health research in 2010 was wasted and an important contributing factor was the misalignment of patient and clinician research objectives.[14]·[15] As a consequence, several research funding bodies now advocate the involvement of patients in the design and conduct of research. This has demonstrable beneficial impact.[16] Patient and public involvement (PPI) plays a particularly important role in the National Institute for Health Research (NIHR).[17] In addition to participation and engagement in the research process, the involvement of patients in identifying relevant research topics and their prioritisation is particularly encouraged. Organisations such as the James Lind Alliance have

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successfully brought together patients, professionals and industry in order to set research priorities, e.g. for spinal cord injury.[18] However, the research priorities for individuals suffering from DCM have not yet been assessed.

A recent systematic review of DCM research demonstrated a heavy focus on surgical technique.[19] [20] However, the research needs of patients with DCM and their priorities remain unknown. Moreover, as part of a core-outcomes initiative (REsearch Objectives and COmmon Date Elements in DCM [RECODE-DCM]) we have identified that outcome domains are not consistently reported in current clinical research.[19] The present project set out to establish the needs and priorities of individuals suffering from DCM. This will help to determine the outcome assessments that should be included in clinical research and to better direct future research.

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Methods

 Reporting adheres to the EQUATOR Network STROBE checklist[21].

Survey development

Patient and Public Involvement

Individuals with DCM and their caregivers were invited to attend the Myelopathy.org Patient and Public Involvement day, hosted at the University of Cambridge and captured by Cambridge TV in their documentary.[22] Myelopathy.org is an international, charitable organisation for individuals affected by or working with DCM. As part of the event, qualitative interviews (N=9) were used to establish relevant functional domains that affected quality of life of individuals with DCM. These were found to resemble domains previously reported by Anderson et al. (2004), who conducted a survey amongst patients with traumatic spinal cord injury asking them to rank seven domains of spinal cord function in order of priority for recovery.[23] Using this as a template but broadening 'upper body/trunk strength and balance' to upper body/trunk function, the following recovery domains were agreed by participants: elimination of pain, arm and hand function, walking, sexual function, upper body/trunk function, sensation and bladder/bowel function. For brevity, in this article they are referred to as arm/hand, walking, sexual function, pain, sensation, trunk and bladder/bowel.

These questions were embedded into an existing electronic survey initiative, developed using Survey Monkey (California, USA) and following the Checklist for Reporting Results of Internet E-Surveys (CHERRIES),[24] investigating patient reporting of DCM. This iteration was piloted by the lead investigators and a selection of individuals with DCM. Ethical approval was granted by the University of Cambridge. Study objectives were outlined on the initial page, including details of the host organization and estimated time required to complete the survey. This acted as the electronic consent, with continuation into the survey as agreement. Respondents were also presented with a description of DCM, including relevant synonyms, and required to confirm they suffered with the condition.

Respondents were asked to rank recovery domains in order of priority and provide details about their DCM. DCM characteristics included age, gender, history of surgery, best daily limb pain score (using a visual analogue scale), duration of symptoms and disease severity as measured using the self-reported, modified Japanese Orthopaedic Association [mJOA].[25] The mJOA is amongst the most commonly utilised assessments of disease severity[19]·[20] and is fully validated.[26] It is a composite score based on upper limb function, lower limb function, upper limb sensation and bladder function. The score is valid for analysis in its entirety or per domain. All questions were mandatory, but respondents were not required to rank every recovery domain, on the basis that some domains may not

be a priority for them. The sequence of questions and order of responses was not altered from respondent to respondent.

Survey administration

The survey was accessed via a landing page on Myelopathy.org, allowing assessment of response rates using Google Analytics (California, USA). Individuals with DCM were recruited over an eighteen-month period. The recruitment process has been described in detail previously[27] but in short, the survey was advertised using Google Adwords (California, USA) and through Myelopathy.org and its social media outlets. The survey was voluntary and internet protocol addresses were used to prevent users submitting multiple responses. A missing data analysis was conducted between complete and incomplete survey responses to consider if particular subgroups were more likely to terminate early. Complete responders were defined as having provided answers for all aforementioned variables.

Analysis

Research priorities are presented using summary statistics, including average ranking and overall proportion of patients per domain. Domains which were not ranked by a respondent were omitted from these scores. For subgroup analysis, variables were dichotomised and thresholds were chosen based on the graphical distribution of responses and sample sizes. Categorical variables were compared using the Chi-Squared test. For continuous variables, the Shapiro-Wilks test was used to assess for parametric distribution of data sets. The Mann-Whitney U test was then used to compare the means of non-parametric distributions whilst a two-tailed T-test used to compare the means of parametric distributions. Pearson's correlations were performed to assess between group differences in characteristics, which could have influenced sub-group analysis. Significance was set at p < 0.05.

Results

Respondents

The survey was uniquely accessed 1463 times, with 659 visitors entering the survey (participation rate of 33%). A total of 481 responses contained complete data (completion rate 73%). A missing data analysis was conducted comparing incomplete and complete responses. Patients who completed the survey in full were more likely to have undergone surgery (p = 0.04), otherwise there was no statistical difference within variables of interest (Supplementary Data 2). Only complete responses were analysed in the present study. Of these responses domains were ranked more than 80% of the time: pain (400, 83%), sensation (428, 89%), walking (396, 82%), arm and hand (393, 82%), sexual (388, 81%), bladder and bowel (399, 83%) and trunk function (407, 85%).

On average respondents were more likely to be female (341, 70%) and suffer with moderate myelopathy (11.9 ±3.0) for between 3 and 10 years (181, 31%). Around half of patients (221, 46%) had undergone surgery. Overall respondent demographics are summarised in Table 1. Considering group differences, patients who had suffered from the disease for longer were more likely to have undergone surgery (p < 0.01) and have worse myelopathy (-0.22, p < 0.005). They were also more likely to suffer more pain (-0.14, p < 0.01). Average pain scores were 3.1 (±2.4) for patients suffering with the disease for less than a year, rising to 4.5 (±3.0) for patients suffering for at least 10 years. There was no relationship between severity of myelopathy and pain scores (-0.04, p = 0.36). Between group differences are summarised in Supplementary Data 3.

Ranking of spinal cord dysfunction domains

Overall, pain was the most popular number one ranked recovery domain, chosen by 39.9% of respondents. This was followed by walking (20.2%), sensation (11.9%) and arm and hand function (11.5%). Sexual function (5.7%), bladder and bowel (3.7%) or trunk function (3.5%) were chosen less frequently. When considering the average ranking of symptoms, whilst pain remained the priority (2.6 ± 2.0), this was closely followed by walking (2.9 ± 1.7) and arm/hand function (3.0 ± 1.4) (Figure 1). Sensation ranked lower (4.3 ± 2.1).

Impact of baseline characteristics on ranking of spinal cord dysfunction domains

Respondents who had undergone surgery were more likely to prioritise walking (p < 0.005) and trunk function (p = 0.03), whereas patients who had not yet undergone surgery were more likely to prioritise upper limb function (p < 0.05) (Figure 2). Patients with poor upper limb or lower limb function were more likely to prioritise arm/hand recovery (p < 0.005) and

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When considering the average rankings pain, arm/hand function and walking remained the top three recovery priorities (Figure 3). However, amongst the subcategories, the order of these priorities differed slightly (Supplementary Data 4). Patients who were male, or who had undergone surgery, or who had greater lower limb or bladder functional disability, prioritised recovery of walking, over pain and arm/hand function; patients with greater upper limb function or sensory disability prioritised recovery of arm/hand function over pain and walking.

When overall mJOA scores were considered to evaluate mild, moderate and severe patients,⁶ no variation was seen in modal or average ranked priorities.

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Discussion

This is the first study to systematically survey functional domains relevant to DCM and to ask patients to rank them in order of importance to their quality of life. The established priorities are likely to reflect symptom prevalence and their impact on day to day life.[23] The analysis of 481 completed answers demonstrated that pain, arm/hand function and walking emerge as the most important spinal cord dysfunction domains. Although based on averaged rankings, there were some subtle differences in ordering of these three domains. With the exception of patients with significant gait impairment, elimination of pain was the recovery priority independent of baseline characteristic.

These findings are surprising: functional disability (specifically recovery of arm/hand and walking function) has been and continues to be a focus for researchers, typically in response to surgery,[8] but more recently with a shift towards enhancing post-surgical recovery.[13]·[27] In contrast, pain is not widely recognised as an important relevant domain. Our recent review of outcome reporting in DCM clinical trials demonstrated that the overwhelming majority of studies (90%) reported outcomes related to function, but only 27% of studies reported outcomes related to pain,[19] despite the fact that pain is a well-recognised feature of DCM,[5] which often improves following surgery.[11] The present findings highlight the fact that systematic research of patient needs is sorely lacking in DCM. A possible explanation for this discrepancy is that surgeons, who play a significant role in the management of DCM and predominate this research field, remain biased towards functional domains because pain is not a recognised indication for surgery in DCM.[6]

The priorities established in the present study differ from those of individuals suffering from spinal cord injury. Although pain is amongst the most prevalent symptoms of traumatic spinal cord injury,[28]·[29] the "elimination of chronic pain" was considered to be a relatively low priority amongst those surveyed in Anderson's study[23] and a similar study by Kwon et al.[30] that focused on the priorities for SCI recovery after novel treatments (e.g. stem cells). Instead, quadriplegics prioritised arm/hand function, whilst paraplegics sexual and bladder/bowel function. These differences relate to their specific significance for patient independence and quality of life.

In DCM, the symptom burden is less well-described[31]·[32] and the relationship between symptom burden or their significance with respect to quality of life in DCM has not been investigated. However, it would seem likely a similar relationship exists.

Limitations

Following recommendation by the James Lind Alliance, which was founded to support priority setting in research,[33]-[34] the present survey was conducted online, as previously described.[27] Respondents belonged to a self-selecting group of individuals who were asked to confirm they had a diagnosis of DCM after being presented with an explanation of the disease for verification purposes. It is possible that some respondents did not have DCM. Reassuringly, respondent demographics were comparable to those of leading prospective surgical studies, with the exception of gender which was not shown to influence patient priorities[8]-[9] (Supplementary Data 1). There are no such comparable series for non-surgical cohorts, but their inclusion provides a further valuable perspective.

The survey questions were not randomly sorted and therefore each respondent answered identical surveys with spinal cord function domains presented in the same order. The last domain assessed was sensation. In keeping with it being the most prevalent DCM symptom,[32] it featured most frequently in the responses, indicating that the order of domains was unlikely to have influenced the rankings. Moreover, answers to demographic questions, which followed the ranking of priorities on the survey, were required to define a complete response in order to be included in the present analysis. Priorities therefore were not influenced by incomplete answers.

Following the qualitative development work and the previous experience of Anderson et al., 2004, the pain domain was kept non-specific, asking patients to rank 'elimination of pain' as a recovery priority (Supplementary Data 2). In contrast however, the pain assessment focused on limb pain, which is classically felt to represent DCM-related pain.[5] Whilst this does not limit the implications of our findings as whole, their interpretation will require a better characterisation of pain in DCM in order to focus research appropriately as other pain foci are reported.[35]

Conclusion

The priorities reported in the present study identify functional domains that are relevant to the quality of life of DCM patients. They provide an important framework for future research and will serve as a valuable reference for the development of a core outcome set relevant to studies in DCM.

Table

Table 1: Summary of respondent demographics.

Respondent Demographics	
Age (Mean ± SD)	53.6 (9.8)
Male Gender (%)	140 (29)
Undergone Surgery (%)	221 (46)
Length of Symptoms (%)	
0 to 1 year	72 (15)
1 to 3 years	140 (29)
3 to 10 years	181 (38)
10 to 25 years	74 (15)
25+ years	14 (3)
mJOA (Mean +SD)	
Upper Limb Function	3.6 (1.0)
Walking	4.4 (1.5)
Upper Limb Sensation	1.7 (0.7)
Bladder Function	2.2 (1.0)
Total	11.9 (3.0)
VAS Limb Pain (Mean ± SD)	3.1 (2.6)

Figure Legends

Figure 1: Overall recovery priorities. The bar chart represents the first choice of patients and the line graph the average ranking for each domain (where the top ranking is 1). Pain was the overall first choice priority of patients, although when priority rankings were averaged, this was closely followed by walking and arm/hand function.

Figure 2: Impact of baseline characteristics on first choice recovery priority. The bar chart represents the first choice of patients. Significant between group differences are denoted by the * symbol. For simplicity, groups were dichotomised as follows: Duration <3years, mJOA upper limb function <3, mJOA lower limb function <4 or feeling <2, mJOA bladder/bowel function <2 and VAS limb pain <3. Those who had undergone surgery were more likely to choose trunk function (p=0.03) or walking function (p<0.005), whereas those who had not yet undergone surgery were more likely to choose arm/hand function (p<0.005). Equally patients with more impairment of walking (p<0.005) or arm/hand function (p<0.005) were more likely to prioritise these domains. Pain remained the priority even in patients reporting less pain.

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Figure 3: Impact of baseline characteristics on recovery priority average rankings. The scatter plot represents the mean ranking for each subgroup investigated. The black line represents the overall average. Despite some discrepancies between subgroups, pain, arm/hand and walking function were consistently the top three priorities for patients. Bladder/bowel function was not a recovery priority.

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Declaration Statement:

JH reports being a Medical Advisor for Depuy Synthes and Ethicon, being an Educational Speaker at Globus Medical and research funding from AO Spine. MGF reports consulting for Fortuna Fix. MRK declares a grant from the National Institute for Health Research, travel support from AO Spine and is founder of Myelopathy.org, the first charity for patients with cervical myelopathy. The remaining authors have nothing to declare.

Author Contributions Statement:

All authors were involved in the interpretation, drafting and final approval of the manuscript. Additionally, authors BMD, IS and MRK were involved in the conception, design and acquisition of data for this study, whilst authors BMD and ODM conducted the data analysis.

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Figure 1: Overall recovery priorities. The bar chart represents the first choice of patients and the line graph the average ranking for each domain (where the top ranking is 1). Pain was the overall first choice priority of patients, although when priority rankings were averaged, this was closely followed by walking and arm/hand function.



Figure 2: Impact of baseline characteristics on first choice recovery priority. The bar chart represents the first choice of patients. Significant between group differences are denoted by the * symbol. For simplicity, groups were dichotomised as follows: Duration <3years, mJOA upper limb function <3, mJOA lower limb function <4 or feeling <2, mJOA bladder/bowel function <2 and VAS limb pain <3. Those who had undergone surgery were more likely to choose trunk function (p=0.03) or walking function (p<0.005), whereas those who had not yet undergone surgery were more likely to choose arm/hand function (p<0.05). Equally patients with more impairment of walking (p<0.005) or arm/hand function (p<0.005) were more likely to prioritise these domains. Pain remained the priority even in patients reporting less pain.

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Figure 3: Impact of baseline characteristics on recovery priority average rankings. The scatter plot represents the mean ranking for each subgroup investigated. The black line represents the overall average. Despite some discrepancies between subgroups, pain, arm/hand and walking function were consistently the top three priorities for patients. Bladder/bowel function was not a recovery priority.

Supplementary Data 1: Results from the missing data analysis. Patients completing the survey in full, were more likely to have undergone surgery than those who did not (p = 0.04). Data is presented as mean +/- standard deviation, unless specified as a percentage. Numbers within brackets indicate data points for the respective variable with incomplete data.

	INCOMPLETE SURVEYS (N<178)	COMPLETED SURVEYS (N=481)	Ρ
%MALE	16.3% (7/43)	28.9%	.076
AGE	55.1 +/-10.9 (43)	53.6 +/- 9.9	.344
%SURGERY	35.9% (46/128)	46.0%	.043
LENGTH OF	19% (128)	73%	.304
SYMPTOMS (YRS)			
0-1	18.0% (23)	15.0% (72)	
1-3	21.9% (28)	29.1% (140)	
3-10	35.2% (45)	37.6% (181)	
10-25	18.8% (24)	15.4% (74)	
25+	6.3% (8)	2.9% (14)	
LIMB PAIN VAS	3.6 +/- 1.9 (17)	3.7 +/- 2.5	.854
MJOA	12.2 +/- 3.2 (47)	11.9 +/- 3.0	.364

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Supplementary Data 2: Questionnaire. The questions relevant to this study were developed, piloted and embedded within an ongoing initiative investigating patient reporting of DCM. The questions pertaining to the data points required for this study are presented, including their question number and options for selection. Question 35 about age was the only question where respondents were asked to type in a specific answer. The answer format was electronically validated to require an integer, prompting users to specify to the nearest year.

Questions	Questions generating data points required for this study						
5	How long have you suffered with cervical myelopathy?						
	0-1 year						
	1-3 years						
	3-10 years						
	10-25 years						
	>25 years						
7	Have you undergone surgery for cervical myelopathy?						
	Yes						
	No						
23	Currently, please indicate the intensity of the current, best and						
	worst pain affecting your arms or legs over the past 24h on a scale of 0						
	(no pain) to 10 (worst pain imaginable)						
	0						
	1						
	2						
	3						
	4						
	6						
	7						
	8						
	9						
20	10						
23	now does cervical myelopathy affect your arms and hands? Choose the						
	statement that best fits: I am						
	- Unable to move my hands						
	- Unable to eat with a spoon but am able to move my hands						
	- Unable to button my shirt but able to eat with a spoon						
	- Able to button my shirt with slight difficulty						
	- Not having any trouble using my hands.						

30	How does cervical myelopathy affect your legs? Choose the statement
	that best fits: I am
	 Completely unable to move legs at all and have no feeling in legs Having feeling in legs but not able to move them at all Able to move my legs but am unable to walk Able to walk on flat floor with a walking aid (cane or crutch) Able to walk up and/or downstairs with the aid of a handrail Able to walk up and/or downstairs without handrail but I notice moderate-to-significant lack of stability/feeling of imbalance when I walk Able to walk unaided (no crutches, canes, walker) with smooth reciprocation (ie, legs move smoothly) but I still notice mild lack of stability/feeling of imbalance when yroblems of imbalance or instability
31	How does cervical myelopathy affect your arms and hands? Choose th
	statement that best fits: I have
	Complete loss of faciling in bands
	- Complete loss of reening in narios
	- Severe loss of feeling, or have pain in my hands
	- Mild loss of feeling in hands
	- No loss of feeling in hands
32	How does cervical myelopathy affect your bladder? Choose the
	statement that best fits: I am
	- Am completely unable to control urination
	- Have marked difficulty controlling urination
	- Have mild to moderate difficulty controlling urination
	- No difficulty controlling urination
33	Effective medical research should target the needs of patients. The

	categories. In DCM, the patient priorities are not known.
	For you as a patient, what are the research priorities for you (please
	rank where 1 is the most important and 7 is the least important)? What
	would you like researchers to focus on?
	-Elimination of Pain -Arm/Hand Function -Walking Function -Bladder/Bowel Function -Sexual Function
	-Upper Body/Trunk Function
34	Are you male or female?
	Male Female
35	How old are you?

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 BMJ Open BMJ Open Supplementary Data 3: Summary of group differences between investigated variables. Highlighted cells representing of ficant differences in the proportion of respondents per group. Further analysis revealed that differences followed a logical course: patients who had had symptoms for longer or undergone surgery were more likely to have severe disease. In addition, patients with more severe disease were likely to have higher boom and scores.

	Ν	Age (±	ESD)	Male ((%)	Unde	rgone	Length	of	mJO, Limb	A Upper Function		A Lower	mJC)A erlimb	mJC Blac)A Ider	Mean	VAS Pain <3
						ourge	,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,,	(%)		<3		o text	ion <4	Sen: <2	sation	Fund	ction <2	LIIIOI	
Gender, Male	140	55.8	10.3			64	46%	71	51%	20	14%	ibad p∉rie and	36%	50	36%	30	21%	88	63%
Gemder, Female	341	52.8	9.5			157	46%	141	41%	38	11%	ed fro sult(A) I data	31%	143	42%	75	22%	205	60%
Jndergone Surgery																		135	61%
	221	53.1	8.7	64	29%			83	38%	31	14%	, <u>,,,,,,,,,,,,,,</u> ,,,,,,,,,,,,,,,,,,,,,	41%	101	46%	53	24%		
Not Undergone Surgery	260	54	10.7	74	28%			129	50%	27	10%	68	26%	92	35%	53	20%	158	61%
mJOA Upper Limb																		21	36%
Function <3	58	56.7	9.3	19	33%	31	53%	13	22%			<u>ම</u> 46 <mark> </mark>	79%	52	90%	29	50%	070	
Tupotion 2	400	F2 2	0.0	110	200/	100	450/	100	470/				260/	1 1 1	220/	70	100/	272	64%
$\sim 100 \text{ Lower Limb}$	423	53.Z	9.0	119	20%	190	40%	199	41%				20%	141	33%	70	10%	95	E / 0/
Function <4	158	55 4	10 1	50	32%	90	57%	51	32%	46	29%	j.o		91	58%	61	30%	05	547
nJOA Lower Limb	100	00.4	10.1	00	0270	50	01 /0	01	0270	40	2070	ön		51	0070	01	0070	208	64%
Function >4	323	52.7	9.5	88	27%	131	41%	161	50%	12	4%	~		102	32%	44	14%		017
nJOA Upper Limb												ar						86	45%
Sensation <2	193	53.7	9.1	48	25%	101	52%	78	40%	52	27%	ត្ថូ91 ឆ្ន	47%			67	35%		
mJOA Upper Limb												ch						207	72%
Sensation >2	288	53.3	10.3	90	31%	120	42%	134	47%	6	2%	<u>67</u>	23%			38	13%		
mJOA Bladder Function <2	105	54	9.6	29	28%	53	50%	40	38%	29	28%	9 61 N	58%	67	64%			58	55%
nJOA Bladder Function >2	376	53.5	9.9	109	29%	168	45%	172	46%	29	8%	<u></u> 87 <u>8</u>	26%	126	34%			235	63%
_ength of Symptoms <3	040	50.0	40.0	74	000/	00	000/			40	00/	. at	0.40/	70	070/	40	400/	136	64%
/ears	212	52.8	10.8	71	33%	83	39%			13	6%	51 ×	24%	78	31%	40	19%	157	E 00/
	260	512	80	67	25%	138	51%			45	17%	107	10%	115	13%	65	24%	157	507
	203	53.3	11	88	30%	135	46%	136	46%	21	7%	85 6	29%	86	29%	58	29%	-	
Best Limb Pain VAS <3	Z 71. 1	00.0		50	200/0	96	16%	76	40%	37	20%	73 D	30%	107	57%	47	25%		

BMJ Open Supplementary Data 4: Mean ranking for recovery domains for baseline characteristics. Based on average rankings, the top ranked domain is highlighted. Whilst pain, walking and arm/hand function remained the priorities, for respondents who were male or had undergoine surgery, or had impaired upper, lower

or bladder function, arm/hand function had the top ranking. For patients with impaired upper limb sensation, walking function was the priority.

	Pain (SD)		Walking (SD)		Arm/Hand		Sexual Function B		B	Bagogr/Bowel		Trunk Function		Sensation (SD)	
					Functio	on (SD)	(SD)		(99)		(SD)				
Gender, Male	3.1	(2.2)	3.2	(1.6)	2.6	(1.6)	4.2	(1.8)	5 to Ta	(1.7)	4.4	(1.6)	4.2	(2.3)	
Gender, Female	2.5	(2.0)	2.9	(1.4)	3.0	(1.7)	4.1	(1.6)	53952	(1.6)	4.6	(1.5)	4.4	(2.0)	
Undergone Surgery	2.8	(2.1)	3.0	(1.4)	2.6	(1.6)	4.1	(1.6)	5 ageria	(1.6)	4.6	(1.6)	4.5	(2.0)	
Not Undergone Surgery	2.6	(2.0)	2.9	(1.5)	3.2	(1.7)	4.2	(1.7)	5 á sế sế sế số	(1.7)	4.4	(1.5)	4.2	(2.1)	
mJOA Upper Limb Function <3	2.9	(2.0)	2.1	(1.3)	2.5	(1.3)	3.7	(1.6)	from h (ABES atanhin	(1.7)	4.4	(1.3)	4.2	(2.1)	
mJOA Upper Limb Function >3	2.6	(2.0)	3.1	(1.5)	3.0	(1.7)	4.2	(1.7)	ing; Al	(1.7)	4.5	(1.6)	4.3	(2.1)	
mJOA Lower Limb Function <4	2.8	(2.0)	2.9	(1.5)	2.4	(1.5)	4.1	(1.6)	njoper 5 ⁵ inii	(1.7)	4.6	(1.4)	4.3	(2.1)	
mJOA Lower Limb Function >4	2.6	(2.1)	3.0	(1.5)	3.2	(1.7)	4.2	(1.7)	ng, an	(1.7)	4.5	(1.6)	4.3	(2.0)	
mJOA Upper Limb Sensation <2	2.7	(2.1)	2.6	(1.3)		(1.5)	4.2	(1.5)	d Simil	(1.6)	4.5	(1.5)	4.2	(2.1)	
mJOA Upper Limb Sensation >2	2.6	(2.0)	3.2	(1.6)	3.0	(1.8)	4.1	(1.8)	n June lar (fe c	(1.7)	4.5	(1.6)	4.4	(2.1)	
mJOA Bladder Function <2	2.6	(1.8)	2.8	(1.6)	2.5	(1.4)	3.6	(1.4)	6780 12	(1.3)	4.7	(1.6)	4.7	(2.0)	
mJOA Bladder Function >2	2.7	(2.1)	3.0	(1.5)	3.1	(1.7)	4.3	(1.7)	5,00 2	(1.7)	4.5	(1.5)	4.2	(2.1)	
Length of Symptoms <3 years	2.9	(2.2)	3.0	(1.5)	3.1	(1.6)	4.2	(1.7)	566 25	(1.7)	4.6	(1.5)	4.1	(2.1)	
Length of Symptoms >3 years	2.5	(1.9)	2.9	(1.4)	2.8	(1.7)	4.1	(1.7)	5.8 🎬	(1.6)	4.5	(1.5)	4.5	(2.1)	
Best Limb Pain VAS <3	2.7	(2.1)	3.0	(1.5)	2.9	(1.7)	4.2	(1.7)	5.7 હૂ	(1.7)	4.4	(1.5)	4.6	(2.0)	
Best Limh Dain VAS 53	25	(2.0)	20	(1 5)	3.0	(1.6)	/1 1	(1.6)	snce -	(1.6)	17	(1.6)	30	(2.2)	
	2.5	(2.0)	2.5	(1.3)	5.0	(1.0)	4.1	(1.0)	<u>5.0</u> B	(1.0)	4./	(1.0)	3.3	(2.2)	
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STRORE Statement

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1 2			STROBE Statement	
3 4	Section/Topic	Item No	Recommendation	Reported on Page No
5 6 7	Title and abstract	1	(a) Indicate the study's design with a commonly used term in the title or the abstract Image: Commonly used term in the title or the abstract (b) Provide in the abstract an informative and balanced summary of what was done and what was formed to the title or the abstract and the title or the title or the abstract and the title or the title or the abstract and the title or the title	2 2 2
8	Introduction			
9	Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	4-5
11	Objectives	3	State specific objectives, including any prespecified hypotheses	5
12	Methods		den g.	
13	Study design	4	Present key elements of study design early in the paper	6-7
15	Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, following and data collection	6-7
17 18 19 20 21 22 22	Participants	6	(a) Cohort study—Give the eligibility criteria, and the sources and methods of selection of participation of the secrible methods of follow-up Case-control study—Give the eligibility criteria, and the sources and methods of case ascertainment of the choice of cases and controls Cross-sectional study—Give the eligibility criteria, and the sources and methods of selection of participation of the choice of cases and controls	6-7
24 25	5 5		(b) Cohort study—For matched studies, give matching criteria and number of exposed and unexposed and $Case-control study$ —For matched studies, give matching criteria and the number of controls per case	
26 27 28	Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	6-7
29 30	Data sources/measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Bescribe comparability of assessment methods if there is more than one group	6-7
31	Bias	9	Describe any efforts to address potential sources of bias	6-7
33	Study size	10	Explain how the study size was arrived at	6-7
34	Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which group ngs were chosen and why	6-7
35			(a) Describe all statistical methods, including those used to control for confounding	7
37	7		(b) Describe any methods used to examine subgroups and interactions	7
38	3		(c) Explain how missing data were addressed	7
39	Statistical methods	12	(d) Cohort study—If applicable, explain how loss to follow-up was addressed	
40			<i>Case-control study</i> —If applicable, explain how matching of cases and controls was addressed	7
42	2		<i>Cross-sectional study</i> —If applicable, describe analytical methods taking account of sampling strategy	
43	3		(e) Describe any sensitivity analyses	
44 45 46	+ 5 5		For peer review only - http://bmjopen.bmj.com/site/about/guidelines.xhtml	1

		BMJ Open do gran	Page 28 of 29
Section/Topic	Item No	Recommendation 2019-0314	Reported on Page No
Results			
7 Bentisinsute	12*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined of receive the study, confirmed eligible, included in the study, completing follow-up, and analysed	8
Participants	13*	(b) Give reasons for non-participation at each stage	N/A
0		(c) Consider use of a flow diagram	N/A
2 3		(a) Give characteristics of study participants (eg demographic, clinical, social) and information on expansion of end of the confounders	8
4 Descriptive data	14*	(b) Indicate number of participants with missing data for each variable of interest	Supplementary Data 2
7		(c) Cohort study—Summarise follow-up time (eg, average and total amount)	N/A
8		Cohort study—Report numbers of outcome events or summary measures over time	N/A
9 Outcome data	15*	Case-control study—Report numbers in each exposure category, or summary measures of exposure	N/A
20 21		Cross-sectional study—Report numbers of outcome events or summary measures	12-13
22		(a) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (e 295% confidence interval).	0
3		Make clear which confounders were adjusted for and why they were included	8
⁴ Main results	16	(b) Report category boundaries when continuous variables were categorized	Supplementary
26			Data 1,3 & 4
.7		(c) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	N/A
⁸ Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	8-9
0 Discussion		ר די עניין די ניין די ניין די ניין די ניין די ניין די ניין געניין געניין געניין געניין געניין געניין געניין געני געניין געניין	
Key results	18	Summarise key results with reference to study objectives	10
2 3 Limitations 34	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	11
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses results from similar studies, and other relevant evidence	10-11
Generalisability	21	Discuss the generalisability (external validity) of the study results	10-11
³⁹ Other Information			
HO HI Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	1
¹² * <i>Give information separa</i>	tely for case	es and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-	
14 15 16		For peer review only - http://bmjopen.bmj.com/site/about/guidelines.xhtml	2
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Pa	age 29 of 29 BMJ Open	.cted by
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Recovery priorities in degenerative cervical myelopathy: a cross-sectional survey of an international, online community of patients

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Recovery priorities in degenerative cervical myelopathy: a cross-sectional

survey of an international, online community of patients

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Abstract

Objectives

To establish the recovery priorities of individuals suffering with degenerative cervical myelopathy (DCM).

Design

A cross-sectional, observational study.

Setting

Patients from across the world with a diagnosis of DCM accessed the survey over an 18month period on Myelopathy.org, an international myelopathy charity.

Participants

481 individuals suffering from DCM completed the online survey fully.

Main outcome measures

Functional recovery domains were established through qualitative interviews and a consensus process. Individuals were asked about their disease characteristics, including limb pain (visual analogue scale) and functional disability (patient derived - modified Japanese Orthopaedic Association score). Individuals ranked recovery domains (arm and hand function, walking, upper body/trunk function, sexual function, elimination of pain, sensation and bladder/bowel function) in order of priority. Priorities were analysed as the modal first priority and mean ranking. The influence of demographics on selection was analysed, with significance p<0.05.

Results

Of 659 survey responses obtained, 481 were complete. Overall, pain was the most popular recovery priority (39.9%) of respondents, followed by walking (20.2%), sensation (11.9%) and arm and hand function (11.5%). Sexual function (5.7%), bladder and bowel (3.7%) or trunk function (3.5%) were chosen less frequently. When considering the average ranking of symptoms, whilst pain remained the priority (2.6±2.0), this was closely followed by walking (2.9±1.7) and arm/hand function (3.0 ±1.4). Sensation ranked much lower (4.3±2.1). With respect to disease characteristics, overall pain remained the recovery priority, with the exception of patients with greater walking impairment (p<0.005) who prioritised walking, even amongst patients with lower pain scores.

Conclusions

This is the first study investigating patient priorities in DCM. The patient priorities reported provide an important framework for future research and will help ensure that it is aligned with patient needs.

Strengths

- This is the <u>largest</u> study of patient perspective in DCM to date and the <u>first</u> to consider patient recovery priorities
- This study is unique in reporting on both surgical and non-surgical DCM patients.
- This study includes a broad demographic representation of patients from across the globe and includes subgroup analysis.

Limitations

- This is an open-access, internet-based survey, a methodology which can lead to a sampling bias.
- Efforts to mitigate against sampling bias, alongside reassuring sub-group analysis suggest this risk is low.

Introduction

 Degenerative cervical myelopathy [DCM] has been coined as an umbrella term for degenerative and congenital or acquired conditions of the cervical spine, such as spondylosis or ossification of the posterior longitudinal ligament (OPLL), which lead to symptomatic cord compression.[1] With an estimated prevalence of up to 5% in individuals above 40 years old,[2]·[3] DCM is the most common cause of spinal cord dysfunction worldwide.[1] Given its degenerative aetiology and the rising age of the population, this incidence is expected to rise.[4]

The cervical spinal cord acts as a processor and conduit of information between the brain and the periphery. Its injury can therefore give rise to a range of possible symptoms.[1] These include pain, paraesthesia, weakness, unsteadiness, frequent falls, bladder or bowel dysfunction and impotence in men.[5] At early stages, individual symptoms may occur in isolation, but more typically occur in combination, especially as the disease advances.

At present, decompressive surgery is the only evidence-based treatment for DCM.[6]. Surgical decompression is able to halt the progression of symptoms and offer limited, albeit clinically relevant[7] improvements across a range of domains.[8]·[9] However, due to the limited intrinsic capacity for the spinal cord to repair, most patients do not make a full recovery, and instead suffer lifelong disabilities.[9] As a consequence, unemployment and/or dependency is prevalent amongst individuals with DCM.[4]·[10]·[11] Moreover, a recent study has identified that DCM severely impacts quality of life with recorded SF-36, patient reported outcome scores amongst the lowest of all chronic disease.[12] Improving recovery is therefore a major unmet clinical need in DCM.[13]

Medical research is primarily designed by health care professionals. This bears the risk of not taking into account actual patient needs. The concept of 'research wastage' has emerged to depict healthcare research that does not yield actual or potential clinical benefit. In the 2014 *Lancet* series, Chalmers et al. estimated that as much as 85% of the US\$240 billion expended on health research in 2010 was wasted and an important contributing factor was the misalignment of patient and clinician research objectives.[14]·[15] As a consequence, several research funding bodies now advocate the involvement of patients in the design and conduct of research. This has demonstrable beneficial impact.[16] Patient and public involvement (PPI) plays a particularly important role in the National Institute for Health Research (NIHR).[17] In addition to participation and engagement in the research process, the involvement of patients in identifying relevant research topics and their prioritisation is particularly encouraged. Organisations such as the James Lind Alliance have
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successfully brought together patients, professionals and industry in order to set research priorities, e.g. for spinal cord injury.[18] However, the research priorities for individuals suffering from DCM have not yet been assessed.

A recent systematic review of DCM research demonstrated a heavy focus on surgical technique.[19]·[20] However, the research needs of patients with DCM and their priorities remain unknown. Moreover, as part of a core-outcomes initiative (REsearch Objectives and COmmon Date Elements in DCM [RECODE-DCM]) we have identified that outcome domains are not consistently reported in current clinical research.[19]

In this study we sought to establish the recovery needs and priorities of individuals suffering from DCM. This will help to determine the outcome assessments that should be included in clinical research and to better direct future research.

Methods

 Reporting adheres to the EQUATOR Network STROBE checklist[21].

Patient and Public Involvement

Individuals with DCM and their caregivers were invited to attend the Myelopathy.org Patient and Public Involvement day, hosted at the University of Cambridge and captured by Cambridge TV in their documentary.[22] Myelopathy.org is an international, charitable organisation for individuals affected by or working with DCM. As part of the event, qualitative interviews (N=9) were used to establish relevant functional domains that affected quality of life of individuals with DCM. These were found to resemble domains previously reported by Anderson et al. (2004), who conducted a survey amongst patients with traumatic spinal cord injury asking them to rank seven domains of spinal cord function in order of priority for recovery.[23] Using this as a template but broadening 'upper body/trunk strength and balance' to upper body/trunk function, the following recovery domains were agreed by participants: elimination of pain, arm and hand function, walking, sexual function, upper body/trunk function, sensation and bladder/bowel function. For brevity, in this article they are referred to as arm/hand, walking, sexual function, pain, sensation, trunk and bladder/bowel.

These questions were embedded into an existing electronic survey initiative, developed using Survey Monkey (California, USA) and following the Checklist for Reporting Results of Internet E-Surveys (CHERRIES),[24] investigating patient reporting of DCM. This iteration was piloted by the lead investigators and a selection of individuals with DCM. Ethical approval was granted by the University of Cambridge. Study objectives were outlined on the initial page, including details of the host organization and estimated time required to complete the survey. This acted as the electronic consent, with continuation into the survey as agreement. Respondents were also presented with a description of DCM, including relevant synonyms, and required to confirm they suffered with the condition.

Respondents were asked to rank recovery domains in order of priority and provide details about their DCM. DCM characteristics included age, gender, history of surgery, best daily limb pain score (using a visual analogue scale), duration of symptoms and disease severity as measured using the self-reported, patient-derived, modified Japanese Orthopaedic Association [**P**-mJOA]. [25] The modified Japanese Orthopaedic scale [mJOA] is amongst the most commonly utilised assessments of disease severity[19]·[20] and is fully validated.[26] It is a composite score based on upper limb function, lower limb function, upper limb sensation and bladder function. The score is valid for analysis in its entirety or per domain. Originally developed as an investigator administered tool, it has recently been

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adapted and validated for use by patients. [25] All questions were mandatory, but respondents were not required to rank every recovery domain, on the basis that some domains may not be a priority for them. The sequence of questions and order of responses was not altered from respondent to respondent.

Survey administration

The survey was accessed via a landing page on Myelopathy.org, allowing assessment of response rates using Google Analytics (California, USA). Individuals with DCM were recruited over an eighteen-month period. The recruitment process has been described in detail previously[27] but in short, the survey was advertised using Google Adwords (California, USA) and through Myelopathy.org and its social media outlets. The survey was voluntary and internet protocol addresses were used to prevent users submitting multiple responses. A missing data analysis was conducted between complete and incomplete survey responses to consider if particular subgroups were more likely to terminate early. Complete responders were defined as having provided answers for all aforementioned variables.

Analysis

Research priorities are presented using summary statistics, including average ranking and overall proportion of patients per domain. Domains which were not ranked by a respondent were omitted from these scores. For subgroup analysis, variables were dichotomised and thresholds were chosen based on the graphical distribution of responses and sample sizes. Categorical variables were compared using the Chi-Squared test. For continuous variables, the Shapiro-Wilks test was used to assess for parametric distribution of data sets. The Mann-Whitney U test was then used to compare the means of non-parametric distributions whilst a two-tailed T-test used to compare the means of parametric distributions. Pearson's correlations were performed to assess between group differences in characteristics, which could have influenced sub-group analysis. Significance was set at p < 0.05.

Results

Respondents

The survey was uniquely accessed 1463 times, with 659 visitors entering the survey (participation rate of 33%). A total of 481 responses contained complete data (completion rate 73%). A missing data analysis was conducted comparing incomplete and complete responses. Patients who completed the survey in full were more likely to have undergone surgery (p = 0.04), otherwise there was no statistical difference within variables of interest (Supplementary Data 1). Only complete responses were analysed in the present study. Of these responses domains were ranked more than 80% of the time: pain (400, 83%), sensation (428, 89%), walking (396, 82%), arm and hand (393, 82%), sexual (388, 81%), bladder and bowel (399, 83%) and trunk function (407, 85%).

On average respondents were more likely to be female (341, 70%) and suffer with moderate myelopathy (11.9 ±3.0) for between 3 and 10 years (181, 31%). Around half of patients (221, 46%) had undergone surgery. Overall respondent demographics are summarised in Table 1. Considering group differences, patients who had suffered from the disease for longer were more likely to have undergone surgery (p < 0.01) and have worse myelopathy (-0.22, p < 0.005). They were also more likely to suffer more pain (-0.14, p < 0.01). Average pain scores were 3.1 (±2.4) for patients suffering with the disease for less than a year, rising to 4.5 (±3.0) for patients suffering for at least 10 years. There was no relationship between severity of myelopathy and pain scores (-0.04, p = 0.36). Between group differences are summarised in Supplementary Data 2.

Ranking of spinal cord dysfunction domains

Overall, pain was the most popular number one ranked recovery domain, chosen by 39.9% of respondents. This was followed by walking (20.2%), sensation (11.9%) and arm and hand function (11.5%). Sexual function (5.7%), bladder and bowel (3.7%) or trunk function (3.5%) were chosen less frequently. When considering the average ranking of symptoms, whilst pain remained the priority (2.6±2.0), this was closely followed by walking (2.9±1.7) and arm/hand function (3.0 ±1.4) (Figure 1). Sensation ranked lower (4.3±2.1).

Impact of baseline characteristics on ranking of spinal cord dysfunction domains

Respondents who had undergone surgery were more likely to prioritise walking (p < 0.005) and trunk function (p = 0.03), whereas patients who had not yet undergone surgery were more likely to prioritise upper limb function (p < 0.05) (Figure 2). Patients with poor upper

limb or lower limb function were more likely to prioritise arm/hand recovery (p < 0.005) and walking (p < 0.005) respectively (Figure 2). Overall, pain remained the priority, with the exception of patients with greater walking impairment (p < 0.005), even amongst patients with lower pain scores (Figure 2).

When considering the average rankings pain, arm/hand function and walking remained the top three recovery priorities (Figure 3). However, amongst the subcategories, the order of these priorities differed slightly (Supplementary Data 3). Patients who were male, or who had undergone surgery, or who had greater lower limb or bladder functional disability, prioritised recovery of walking, over pain and arm/hand function; patients with greater upper limb function or sensory disability prioritised recovery of arm/hand function over pain and walking.

When overall P-mJOA scores were considered to evaluate mild, moderate and severe patients,⁶ no variation was seen in modal or average ranked priorities.

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Discussion

This is the first study to systematically survey functional domains relevant to DCM and to ask patients to rank them in order of importance to their quality of life. The established priorities are likely to reflect symptom prevalence and their impact on day to day life.[23] The analysis of 481 completed answers demonstrated that pain, arm/hand function and walking emerge as the most important spinal cord dysfunction domains. Although based on averaged rankings, there were some subtle differences in ordering of these three domains. With the exception of patients with significant gait impairment, elimination of pain was the recovery priority independent of baseline characteristic.

These findings are surprising: functional disability (specifically recovery of arm/hand and walking function) has been and continues to be a focus for researchers, typically in response to surgery,[8] but more recently with a shift towards enhancing post-surgical recovery.[13]·[27] In contrast, pain is not widely recognised as an important relevant domain. Our recent review of outcome reporting in DCM clinical trials demonstrated that the overwhelming majority of studies (90%) reported outcomes related to function, but only 27% of studies reported outcomes related to pain,[19] despite the fact that pain is a well-recognised feature of DCM,[5] which often improves following surgery.[11] The present findings highlight the fact that systematic research of patient needs is sorely lacking in DCM. A possible explanation for this discrepancy is that surgeons, who play a significant role in the management of DCM and predominate this research field, remain biased towards functional domains because pain is not a recognised indication for surgery in DCM.[6]

The priorities established in the present study differ from those of individuals suffering from spinal cord injury. Although pain is amongst the most prevalent symptoms of traumatic spinal cord injury,[28]·[29] the "elimination of chronic pain" was considered to be a relatively low priority amongst those surveyed in Anderson's study[23] and a similar study by Kwon et al.[30] that focused on the priorities for SCI recovery after novel treatments (e.g. stem cells). Instead, quadriplegics prioritised arm/hand function, whilst paraplegics sexual and bladder/bowel function. These differences relate to their specific significance for patient independence and quality of life.

In DCM, the symptom burden is less well-described[31]·[32] and the relationship between symptom burden or their significance with respect to quality of life in DCM has not been investigated. However, it would seem likely a similar relationship exists.

Limitations

Following recommendation by the James Lind Alliance, which was founded to support priority setting in research,[33]·[34] the present survey was conducted online, as previously described through a DCM charity, Myelopathy.org.[27] Respondents belonged to a selfselecting group of individuals who were asked to confirm they had a diagnosis of DCM by a medical professional, after being presented with an explanation of the disease for verification purposes. It is possible that some respondents did not have DCM. Reassuringly, respondent demographics were comparable to those of leading prospective surgical studies, with the exception of gender which was not shown to influence patient priorities[8]·[9] (Supplementary Data 1). This likely reflects the recognised popularity of online health communities amongst females. There are no such comparable series for non-surgical cohorts, but their inclusion provides a further valuable perspective.

The survey questions were not randomly sorted and therefore each respondent answered identical surveys with spinal cord function domains presented in the same order. The last domain assessed was sensation. In keeping with it being the most prevalent DCM symptom,[32] it featured most frequently in the responses, indicating that the order of domains was unlikely to have influenced the rankings. Moreover, answers to demographic questions, which followed the ranking of priorities on the survey, were required to define a complete response in order to be included in the present analysis. Priorities therefore were not influenced by incomplete answers.

Following the qualitative development work and the previous experience of Anderson et al., 2004, the pain domain was kept non-specific, asking patients to rank 'elimination of pain' as a recovery priority (Supplementary Data 4). In contrast however, the pain assessment focused on limb pain, which is classically felt to represent DCM-related pain.[5] Whilst this does not limit the implications of our findings as whole, their interpretation will require a better characterisation of pain in DCM in order to focus research appropriately as other pain foci are reported.[35]

Conclusion

The priorities reported in the present study identify functional domains that are relevant to the quality of life of DCM patients. They provide an important framework for future research

and will serve as a valuable reference for the development of a core outcome set relevant to studies in DCM.

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Table

Table 1: Summary of respondent demographics.

Respondent Demographics	
Age (Mean ± SD)	53.6 (9.8)
Male Gender (%)	140 (29)
Undergone Surgery (%)	221 (46)
Length of Symptoms (%)	
0 to 1 year	72 (15)
1 to 3 years	140 (29)
3 to 10 years	181 (38)
10 to 25 years	74 (15)
25+ years	14 (3)
P-mJOA (Mean +SD)	
Upper Limb Function	3.6 (1.0)
Walking	4.4 (1.5)
Upper Limb Sensation	1.7 (0.7)
Bladder Function	2.2 (1.0)
Total	11.9 (3.0)
VAS Limb Pain (Mean ± SD)	3.1 (2.6)

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Figure Legends

Figure 1: Overall recovery priorities. The bar chart represents the first choice of patients and the line graph the average ranking for each domain (where the top ranking is 1). Pain was the overall first choice priority of patients, although when priority rankings were averaged, this was closely followed by walking and arm/hand function.

Figure 2: Impact of baseline characteristics on first choice recovery priority. The bar chart represents the first choice of patients. Significant between group differences are denoted by the * symbol. For simplicity, groups were dichotomised as follows: Duration <3years, P-mJOA upper limb function <3, P-mJOA lower limb function <4 or feeling <2, P-mJOA bladder/bowel function <2 and VAS limb pain <3. Those who had undergone surgery were more likely to choose trunk function (p=0.03) or walking function (p<0.005), whereas those who had not yet undergone surgery were more likely to choose arm/hand function (p<0.005). Equally patients with more impairment of walking (p<0.005) or arm/hand function (p<0.005) were more likely to prioritise these domains. Pain remained the priority even in patients reporting less pain.

Figure 3: Impact of baseline characteristics on recovery priority average rankings. The scatter plot represents the mean ranking for each subgroup investigated. The blue line represents the overall average. Despite some discrepancies between subgroups, pain, arm/hand and walking function were consistently the top three priorities for patients. Bladder/bowel function was not a recovery priority.

Declaration Statement:

JH reports being a Medical Advisor for Depuy Synthes and Ethicon, being an Educational Speaker at Globus Medical and research funding from AO Spine. MGF reports consulting for Fortuna Fix. MRK declares a grant from the National Institute for Health Research, travel support from AO Spine and is founder of Myelopathy.org, the first charity for patients with cervical myelopathy. The remaining authors have nothing to declare.

Author Contributions Statement:

BD, OM, IS, BA, BK, SK, JH, JW, RG, MF and MK were involved in the interpretation, drafting and final approval of the manuscript. Additionally, authors BD, IS and MK were involved in the conception, design and acquisition of data for this study, whilst authors BD and OM conducted the data analysis.

Data Availability Statement:

All relevant data to the study are included in the article or uploaded as supplementary information. No additional data are available.

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Figure 1: Overall recovery priorities. The bar chart represents the first choice of patients and the line graph the average ranking for each domain (where the top ranking is 1). Pain was the overall first choice priority of patients, although when priority rankings were averaged, this was closely followed by walking and arm/hand function.



Figure 2: Impact of baseline characteristics on first choice recovery priority. The bar chart represents the first choice of patients. Significant between group differences are denoted by the * symbol. For simplicity, groups were dichotomised as follows: Duration <3years, mJOA upper limb function <3, mJOA lower limb function <4 or feeling <2, mJOA bladder/bowel function <2 and VAS limb pain <3. Those who had undergone surgery were more likely to choose trunk function (p=0.03) or walking function (p<0.005), whereas those who had not yet undergone surgery were more likely to choose arm/hand function (p<0.05).
Equally patients with more impairment of walking (p<0.005) or arm/hand function (p<0.005) were more likely to prioritise these domains. Pain remained the priority even in patients reporting less pain.

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Figure 3: Impact of baseline characteristics on recovery priority average rankings. The scatter plot represents the mean ranking for each subgroup investigated. The black line represents the overall average. Despite some discrepancies between subgroups, pain, arm/hand and walking function were consistently the top three priorities for patients. Bladder/bowel function was not a recovery priority.

Supplementary Data 1: Results from the missing data analysis. Patients completing the survey in full, were more likely to have undergone surgery than those who did not (p = 0.04). Data is presented as mean +/- standard deviation, unless specified as a percentage. Numbers within brackets indicate data points for the respective variable with incomplete data.

	INCOMPLETE SURVEYS (N<178)	COMPLETED SURVEYS (N=481)	Ρ
%MALE	16.3% (7/43)	28.9%	.076
AGE	55.1 +/-10.9 (43)	53.6 +/- 9.9	.344
%SURGERY	35.9% (46/128)	46.0%	.043
LENGTH OF	19% (128)	73%	.304
SYMPTOMS (YRS)			
0-1	18.0% (23)	15.0% (72)	
1-3	21.9% (28)	29.1% (140)	
3-10	35.2% (45)	37.6% (181)	
10-25	18.8% (24)	15.4% (74)	
25+	6.3% (8)	2.9% (14)	
LIMB PAIN VAS	3.6 +/- 1.9 (17)	3.7 +/- 2.5	.854
MJOA	12.2 +/- 3.2 (47)	11.9 +/- 3.0	.364

BMJ Open BMJ Open Supplementary Data 2: Summary of group differences between investigated variables. Highlighted cells representing of ficant differences in the proportion of respondents per group. Further analysis revealed that differences followed a logical course: patients who had had symptoms for longer or undergone surgery were more likely to have severe disease. In addition, patients with more severe disease were likely to have higher imposition scores.

												atio							
	N	Age (±	SD)	Male ((%)	Under Surge	rgone ery (%)	Length Sympto (%)	of oms <3yrs	mJO/ Limb <3	A Upper Function		A Lower tion <4	mJO Uppe Sens	A er Limb sation	mJC Blad Fund)A Ider ction <2	Mean ' Limb F	/AS ain <3
Gender, Male	140	55.8	10.3			64	46%	71	51%	20	14%	t up er	36%	<u><2</u> 50	36%	30	21%	88	63%
												idec idec							
Gemder, Female	341	52.8	9.5			157	46%	141	41%	38	11%	ata Afro	31%	143	42%	75	22%	205	60%
Undergone Surgery																		135	61%
5 5 ,	221	53.1	8.7	64	29%			83	38%	31	14%		41%	101	46%	53	24%		
Not Undergone Surgery	260	54	10.7	74	28%			129	50%	27	10%	68	26%	92	35%	53	20%	158	61%
mJOA Upper Limb																		21	36%
Function <3	58	56.7	9.3	19	33%	31	53%	13	22%			a 46 <mark>9</mark>	79%	52	90%	29	50%		
mJOA Upper Limb												nir e n						272	64%
Function >3	423	53.2	9.8	119	28%	190	45%	199	47%			<u>6</u> 112	26%	141	33%	76	18%		
mJOA Lower Limb									000/	10		.			=00/			85	54%
Function <4	158	55.4	10.1	50	32%	90	57%	51	32%	46	29%	<u>8</u>		91	58%	61	39%	200	C 40/
mJOA Lower Limb	222	F0 7	0.5	00	270/	101	440/	161	E00/	10	40/	m (100	220/	4.4	1 4 0/	208	64%
m IOA Uppor Limb	323	52.7	9.5	00	21%	131	41%	101	50%	12	4%	a J		102	32%	44	14%	86	15%
Sensation <2	193	537	9.1	48	25%	101	52%	78	40%	52	27%		47%			67	35%	00	4070
m.IOA Upper Limb	100	00.7	0.1	40	2070	101	0270	10	-070	52	2170	ect	4770			07	0070	207	72%
Sensation >2	288	53.3	10.3	90	31%	120	42%	134	47%	6	2%		23%			38	13%	201	1270
mJOA Bladder Function <2	105	54	9.6	29	28%	53	50%	40	38%	29	28%	0 61 N	58%	67	64%	00	.0,0	58	55%
mJOA Bladder Function >2	376	53.5	9.9	109	29%	168	45%	172	46%	29	8%	G 97 2	26%	126	34%			235	63%
Length of Symptoms <3												<u>й й</u>						136	64%
years	212	52.8	10.8	71	33%	83	39%			13	6%	51 式	24%	78	37%	40	19%		
Level at Original second												ģ						157	58%
Length of Symptoms >3	269	54.2	8.9	67	25%	138	51%			45	17%	107	40%	115	43%	65	24%		
years				88	30%	135	46%	136	46%	21	7%	85 Ö	29%	86	29%	58	20%		
years Best Limb Pain VAS <3	293	53.3	11	00						~ -	/		/						

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 BMJ Open Supplementary Data 3: Mean ranking for recovery domains for baseline characteristics. Based on average rankings, the top ranked domain is highlighted.

Whilst pain, walking and arm/hand function remained the priorities, for respondents who were male or had undergoine surgery, or had impaired upper, lower

or bladder function, arm/hand function had the top ranking. For patients with impaired upper limb sensation, walking function was the priority.

	Pain (S	D)	Walkir	ng (SD)	Arm/Ha	nd	Sexual	Function	B	/ Bowel	Trunk	Function	Sensa	tion (SD)
					Functio	n (SD)	(SD)		(\$ <u>9</u> 9 <u></u> 8 <u>9</u> 9		(SD)			
Gender, Male	3.1	(2.2)	3.2	(1.6)	2.6	(1.6)	4.2	(1.8)	53 at o	(1.7)	4.4	(1.6)	4.2	(2.3)
Gender, Female	2.5	(2.0)	2.9	(1.4)	3.0	(1.7)	4.1	(1.6)	5 3 5	(1.6)	4.6	(1.5)	4.4	(2.0)
Jndergone Surgery	2.8	(2.1)	3.0	(1.4)	2.6	(1.6)	4.1	(1.6)	5 and a d	(1.6)	4.6	(1.6)	4.5	(2.0)
Iot Undergone Surgery	2.6	(2.0)	2.9	(1.5)	3.2	(1.7)	4.2	(1.7)	5 a f i i i i i i i i i i i i i i i i i i	(1.7)	4.4	(1.5)	4.2	(2.1)
nJOA Upper Limb Function 3	2.9	(2.0)	2.1	(1.3)	2.5	(1.3)	3.7	(1.6)	from h (ABES) ta9hir	(1.7)	4.4	(1.3)	4.2	(2.1)
nJOA Upper Limb Function 3	2.6	(2.0)	3.1	(1.5)	3.0	(1.7)	4.2	(1.7)	5) 5) 1007, A	(1.7)	4.5	(1.6)	4.3	(2.1)
nJOA Lower Limb Function	2.8	(2.0)	2.9	(1.5)	2.4	(1.5)	4.1	(1.6)	njope 1 trazini	(1.7)	4.6	(1.4)	4.3	(2.1)
nJOA Lower Limb Function 4	2.6	(2.1)	3.0	(1.5)	3.2	(1.7)	4.2	(1.7)	n.bmj. ng,`an	(1.7)	4.5	(1.6)	4.3	(2.0)
nJOA Upper Limb Sensation	2.7	(2.1)	2.6	(1.3)		(1.5)	4.2	(1.5)	d Simi	(1.6)	4.5	(1.5)	4.2	(2.1)
nJOA Upper Limb Sensation	2.6	(2.0)	3.2	(1.6)	3.0	(1.8)	4.1	(1.8)	n Jun lar fle c	(1.7)	4.5	(1.6)	4.4	(2.1)
nJOA Bladder Function <2	2.6	(1.8)	2.8	(1.6)	2.5	(1.4)	3.6	(1.4)		(1.3)	4.7	(1.6)	4.7	(2.0)
nJOA Bladder Function >2	2.7	(2.1)	3.0	(1.5)	3.1	(1.7)	4.3	(1.7)	5,00 N	(1.7)	4.5	(1.5)	4.2	(2.1)
ength of Symptoms <3 years	2.9	(2.2)	3.0	(1.5)	3.1	(1.6)	4.2	(1.7)	5 gg 25	(1.7)	4.6	(1.5)	4.1	(2.1)
ength of Symptoms >3 years	2.5	(1.9)	2.9	(1.4)	2.8	(1.7)	4.1	(1.7)	5.8 🎬	(1.6)	4.5	(1.5)	4.5	(2.1)
Best Limb Pain VAS <3	2.7	(2.1)	3.0	(1.5)	2.9	(1.7)	4.2	(1.7)	5.7 Agen	(1.7)	4.4	(1.5)	4.6	(2.0)
	25	(2.0)	2.9	(1.5)	3.0	(1.6)	4.1	(1.6)	5.8 DD	(1.6)	4.7	(1.6)	3.9	(2.2)

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Supplementary Data 4: Questionnaire. The questions relevant to this study were developed, piloted and embedded within an ongoing initiative investigating patient reporting of DCM. The questions pertaining to the data points required for this study are presented, including their question number and options for selection. Question 35 about age was the only question where respondents were asked to type in a specific answer. The answer format was electronically validated to require an integer, prompting users to specify to the nearest year.

Questions gen	erating data points required for this study
5	How long have you suffered with cervical myelopathy?
	0-1 year
	1-3 years
	3-10 years
	10-25 years
	>25 years
7	Have you undergone surgery for cervical myelopathy?
	Yes
	No
23	Currently, please indicate the intensity of the current, best and
	worst pain affecting your arms or legs over the past 24h on a scale of 0
	(no pain) to 10 (worst pain imaginable)
	4
	0
	2 3
	4
	5
	6
	7
	8 9
	10
29	How does cervical myelopathy affect your arms and hands? Choose the
	statement that best fits: I am
	- Unable to move my hands
	- Unable to eat with a spoon but am able to move my hands
	- Unable to button my shirt but able to eat with a spoon
	- Able to button my shirt with great difficulty
	- Able to button my shirt with slight difficulty
1	- Not having any trouble using my hands.

30	How does cervical myelopathy affect your legs? Choose the statement
	that best fits: I am
	 Completely unable to move legs at all and have no feeling in legs Having feeling in legs but not able to move them at all Able to move my legs but am unable to walk Able to walk on flat floor with a walking aid (cane or crutch) Able to walk up and/or downstairs with the aid of a handrail Able to walk up and/or downstairs without handrail but I notice moderate-to-significant lack of stability/feeling of imbalance when I walk Able to walk unaided (no crutches, canes, walker) with smooth reciprocation (ie, legs move smoothly) but I still notice mild lack of stability/feeling of imbalance when yroblems of imbalance or instability
31	How does cervical myelopathy affect your arms and hands? Choose the
	statement that best fits: I have
	- Complete loss of feeling in hands
	- Severe loss of feeling, or have pain in my hands
	- Mild loss of feeling in hands
	- No loss of feeling in hands
32	How does cervical myelopathy affect your bladder? Choose the
	statement that best fits: I am
	- Am completely unable to control urination
	- Have marked difficulty controlling urination
	- Have mild to moderate difficulty controlling urination
	- No difficulty controlling urination
33	Effective medical research should target the needs of patients. The
00	

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categories. In DCM, the patient priorities are not known.
For you as a patient, what are the research priorities for you (please
rank where 1 is the most important and 7 is the least important)? What
would you like researchers to focus on?
-Elimination of Pain -Arm/Hand Function -Walking Function -Bladder/Bowel Function -Sexual Function -Upper Body/Trunk Function -Normal Sensation
Are you male or female?
Male Female
How old are you?

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STRORE Statement

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1 2			STROBE Statement	
3 4	Section/Topic	Item No	Recommendation	Reported on Page No
5 6 7	Title and abstract	1	(a) Indicate the study's design with a commonly used term in the title or the abstract 5 6 (b) Provide in the abstract an informative and balanced summary of what was done and what was formed at the study and the statement of the stat	2 2
8	Introduction			
9	Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	4-5
11	Objectives	3	State specific objectives, including any prespecified hypotheses	5
12	Methods		teen 19.	
13	Study design	4	Present key elements of study design early in the paper	6-7
15 16	Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, following and data collection	6-7
17 18 19 20 21 22	Participants	6	(a) Cohort study—Give the eligibility criteria, and the sources and methods of selection of participation of the secrible methods of <i>Case-control study</i> —Give the eligibility criteria, and the sources and methods of case ascertainment of the choice of cases and controls <i>Cross-sectional study</i> —Give the eligibility criteria, and the sources and methods of selection of participation of the choice of cases and controls <i>Cross-sectional study</i> —Give the eligibility criteria, and the sources and methods of selection of participation of the choice of cases and controls <i>Cross-sectional study</i> —Give the eligibility criteria, and the sources and methods of selection of participation of the choice of cases and controls <i>Cross-sectional study</i> —Give the eligibility criteria, and the sources and methods of selection of participation of the choice of cases and controls <i>Cross-sectional study</i> —Give the eligibility criteria, and the sources and methods of selection of participation of the choice of cases and controls <i>Cross-sectional study</i> —Give the eligibility criteria, and the sources and methods of selection of participation of the choice of cases and controls <i>Cross-sectional study</i> —Give the eligibility criteria, and the sources and methods of selection of participation of the choice of cases and controls <i>Cross-sectional study</i> —Give the eligibility criteria, and the sources and methods of selection of participation of the choice of cases and controls <i>Cross-sectional study</i> —Give the eligibility criteria, and the sources and methods of selection of participation of the choice of cases and controls <i>Cross-sectional study</i> —Give the eligibility criteria, and the sources and methods of selection of participation of the choice of cases and controls <i>Cross-sectional study</i> —Give the eligibility criteria, and the sources and methods of selection of participation of the choice of cases and controls <i>Cross-sectional study</i> —Give the eligibility criteria, and the sources and methods of selectipation of participation of the choice of ca	6-7
22 24 25			(b) Cohort study—For matched studies, give matching criteria and number of exposed and unexposed and	
26 27 28	Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	6-7
29 30	Data sources/measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measuremeart). Bescribe comparability of assessment methods if there is more than one group	6-7
31	Bias	9	Describe any efforts to address potential sources of bias	6-7
33	Study size	10	Explain how the study size was arrived at	6-7
34	Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which group ngs were chosen and why	6-7
35			(a) Describe all statistical methods, including those used to control for confounding	7
37	,		(b) Describe any methods used to examine subgroups and interactions	7
38	}		(c) Explain how missing data were addressed	7
39 4(Statistical methods	12	(d) Cohort study—If applicable, explain how loss to follow-up was addressed	
41			<i>Case-control study</i> —If applicable, explain how matching of cases and controls was addressed	7
42	2		Cross-sectional study—If applicable, describe analytical methods taking account of sampling strategy	
43	5 		(e) Describe any sensitivity analyses	
45			For peer review only - http://bmjopen.bmj.com/site/about/guidelines.xhtml	1

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1 2 3 Section/Topic 4	Item No	Recommendation	Reported on Page No
Results			
7 3 Denticipante	10*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined or eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	8
Participants	13*	(b) Give reasons for non-participation at each stage	N/A
10		(c) Consider use of a flow diagram	N/A
12 13		(a) Give characteristics of study participants (eg demographic, clinical, social) and information on expansion of study participants (eg demographic, clinical, social) and information on expansion of study participants (eg demographic, clinical, social) and information on expansion of study participants (eg demographic, clinical, social) and information on expansion of study participants (eg demographic, clinical, social) and information on expansion of expansion of the study participants (eg demographic, clinical, social) and information on expansion of the study participants (eg demographic, clinical, social) and information on expansion of the study participants (eg demographic, clinical, social) and information on expansion of the study participants (eg demographic, clinical, social) and information on expansion of the study participants (eg demographic, clinical, social) and information on expansion of the study participants (eg demographic, clinical, social) and information on expansion of the study participants (eg demographic, clinical, social) and information on expansion of the study participants (eg demographic, clinical, social) and information on expansion of the study participants (eg demographic, clinical, social) and information on expansion of the study participants (eg demographic, clinical, social) and information on expansion of the study participants (eg demographic, clinical, social) and information on expansion of the study participants (eg demographic, clinical, social) and information on expansion of the study participants (eg demographic, clinical, social) and information on expansion of the study participants (eg demographic, clinical, social) and information of the study participants (eg demographic, clinical, social) and (eg demographic, clinical, socia	8
 Descriptive data 	14*	(b) Indicate number of participants with missing data for each variable of interest	Supplementary Data 2
10		(c) Cohort study—Summarise follow-up time (eg, average and total amount)	N/A
18		Cohort study—Report numbers of outcome events or summary measures over time	N/A
19 Outcome data	15*	Case-control study—Report numbers in each exposure category, or summary measures of exposure	N/A
20 21		Cross-sectional study—Report numbers of outcome events or summary measures	12-13
22		(a) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (e 295 confidence interval).	0
23		Make clear which confounders were adjusted for and why they were included	8
Main results	16	(b) Report category boundaries when continuous variables were categorized	Supplementary
26			Data 1,3 & 4
27		(c) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time refering	N/A
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	8-9
0 Discussion		r و ا	
Key results	18	Summarise key results with reference to study objectives	10
32 33 Limitations 34	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	11
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses results from similar studies, and other relevant evidence	10-11
Generalisability	21	Discuss the generalisability (external validity) of the study results	10-11
³⁹ Other Information			
40 41 Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	1
¹² * <i>Give information separa</i>	tely for case	es and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-	
14 15 16		For peer review only - http://bmjopen.bmj.com/site/about/guidelines.xhtml	2
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$\begin{array}{c} 1 \\ 2 \\ 3 \\ 4 \\ 5 \\ 6 \\ 7 \\ 8 \\ 9 \\ 1 \\ 1 \\ 1 \\ 1 \\ 1 \\ 1 \\ 1 \\ 1 \\ 1$	Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of the background and the web sites of PLoS Medicine at http://www.oposmedicine.org/. Annals of InterEpidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at www.strobe-statement.org.	rangenetit reporting. The STROBE checklist is matrixed by the strength of the
45 46 47	For peer review only - http://bmjopen.bmj.com/site/about/guidelines.xhtml	

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Recovery priorities in degenerative cervical myelopathy: a cross-sectional survey of an international, online community of patients

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Recovery priorities in degenerative cervical myelopathy: a cross-sectional

survey of an international, online community of patients

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Abstract

Objectives

To establish the recovery priorities of individuals suffering with degenerative cervical myelopathy (DCM).

Design

A cross-sectional, observational study.

Setting

Patients from across the world with a diagnosis of DCM accessed the survey over an 18month period on Myelopathy.org, an international myelopathy charity.

Participants

481 individuals suffering from DCM completed the online survey fully.

Main outcome measures

Functional recovery domains were established through qualitative interviews and a consensus process. Individuals were asked about their disease characteristics, including limb pain (visual analogue scale) and functional disability (patient derived - modified Japanese Orthopaedic Association score). Individuals ranked recovery domains (arm and hand function, walking, upper body/trunk function, sexual function, elimination of pain, sensation and bladder/bowel function) in order of priority. Priorities were analysed as the modal first priority and mean ranking. The influence of demographics on selection was analysed, with significance p<0.05.

Results

Of 659 survey responses obtained, 481 were complete. Overall, pain was the most popular recovery priority (39.9%) of respondents, followed by walking (20.2%), sensation (11.9%) and arm and hand function (11.5%). Sexual function (5.7%), bladder and bowel (3.7%) or trunk function (3.5%) were chosen less frequently. When considering the average ranking of symptoms, whilst pain remained the priority (2.6±2.0), this was closely followed by walking (2.9±1.7) and arm/hand function (3.0 ±1.4). Sensation ranked much lower (4.3±2.1). With respect to disease characteristics, overall pain remained the recovery priority, with the exception of patients with greater walking impairment (p<0.005) who prioritised walking, even amongst patients with lower pain scores.

Conclusions

This is the first study investigating patient priorities in DCM. The patient priorities reported provide an important framework for future research and will help ensure that it is aligned with patient needs.

Strengths

- This is the <u>largest</u> study of patient perspective in DCM to date and the <u>first</u> to consider patient recovery priorities
- This study is unique in reporting on both surgical and non-surgical DCM patients.
- This study includes a broad demographic representation of patients from across the globe and includes subgroup analysis.

Limitations

- This is an open-access, internet-based survey, a methodology which can lead to a sampling bias.
- Efforts to mitigate against sampling bias, alongside reassuring sub-group analysis suggest this risk is low.

Introduction

 Degenerative cervical myelopathy [DCM] has been coined as an umbrella term for degenerative and congenital or acquired conditions of the cervical spine, such as spondylosis or ossification of the posterior longitudinal ligament (OPLL), which lead to symptomatic cord compression.[1] With an estimated prevalence of up to 5% in individuals above 40 years old,[2]·[3] DCM is the most common cause of spinal cord dysfunction worldwide.[1] Given its degenerative aetiology and the rising age of the population, this incidence is expected to rise.[4]

The cervical spinal cord acts as a processor and conduit of information between the brain and the periphery. Its injury can therefore give rise to a range of possible symptoms.[1] These include pain, paraesthesia, weakness, unsteadiness, frequent falls, bladder or bowel dysfunction and impotence in men.[5] At early stages, individual symptoms may occur in isolation, but more typically occur in combination, especially as the disease advances.

At present, decompressive surgery is the only evidence-based treatment for DCM.[6]. Surgical decompression is able to halt the progression of symptoms and offer limited, albeit clinically relevant[7] improvements across a range of domains.[8]·[9] However, due to the limited intrinsic capacity for the spinal cord to repair, most patients do not make a full recovery, and instead suffer lifelong disabilities.[9] As a consequence, unemployment and/or dependency is prevalent amongst individuals with DCM.[4]·[10]·[11] Moreover, a recent study has identified that DCM severely impacts quality of life with recorded SF-36, patient reported outcome scores amongst the lowest of all chronic disease.[12] Improving recovery is therefore a major unmet clinical need in DCM.[13]

Medical research is primarily designed by health care professionals. This bears the risk of not taking into account actual patient needs. The concept of 'research wastage' has emerged to depict healthcare research that does not yield actual or potential clinical benefit. In the 2014 *Lancet* series, Chalmers et al. estimated that as much as 85% of the US\$240 billion expended on health research in 2010 was wasted and an important contributing factor was the misalignment of patient and clinician research objectives.[14]·[15] As a consequence, several research funding bodies now advocate the involvement of patients in the design and conduct of research. This has demonstrable beneficial impact.[16] Patient and public involvement (PPI) plays a particularly important role in the National Institute for Health Research (NIHR).[17] In addition to participation and engagement in the research process, the involvement of patients in identifying relevant research topics and their prioritisation is particularly encouraged. Organisations such as the James Lind Alliance have

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successfully brought together patients, professionals and industry in order to set research priorities, e.g. for spinal cord injury.[18] However, the research priorities for individuals suffering from DCM have not yet been assessed.

A recent systematic review of DCM research demonstrated a heavy focus on surgical technique.[19]·[20] However, the research needs of patients with DCM and their priorities remain unknown. Moreover, as part of a core-outcomes initiative (REsearch Objectives and COmmon Date Elements in DCM [RECODE-DCM]) we have identified that outcome domains are not consistently reported in current clinical research.[19]

In this study we sought to establish the recovery needs and priorities of individuals suffering from DCM. This will help to determine the outcome assessments that should be included in clinical research and to better direct future research.

Methods

 Reporting adheres to the EQUATOR Network STROBE checklist[21].

Survey design

Individuals with DCM and their caregivers were invited to attend the Myelopathy.org Patient and Public Involvement day, hosted at the University of Cambridge and captured by Cambridge TV in their documentary.[22] Myelopathy.org is an international, charitable organisation for individuals affected by or working with DCM. As part of the event, qualitative interviews (N=9) were used to establish relevant functional domains that affected quality of life of individuals with DCM. These were found to resemble domains previously reported by Anderson et al. (2004), who conducted a survey amongst patients with traumatic spinal cord injury asking them to rank seven domains of spinal cord function in order of priority for recovery.[23] Using this as a template but broadening 'upper body/trunk strength and balance' to upper body/trunk function, the following recovery domains were agreed by participants: elimination of pain, arm and hand function, walking, sexual function, upper body/trunk function, sensation and bladder/bowel function. For brevity, in this article they are referred to as arm/hand, walking, sexual function, pain, sensation, trunk and bladder/bowel.

These questions were embedded into an existing electronic survey initiative, developed using Survey Monkey (California, USA) and following the Checklist for Reporting Results of Internet E-Surveys (CHERRIES),[24] investigating patient reporting of DCM. This iteration was piloted by the lead investigators and a selection of individuals with DCM. Ethical approval was granted by the University of Cambridge. Study objectives were outlined on the initial page, including details of the host organization and estimated time required to complete the survey. This acted as the electronic consent, with continuation into the survey as agreement. Respondents were also presented with a description of DCM, including relevant synonyms, and required to confirm they suffered with the condition.

Respondents were asked to rank recovery domains in order of priority and provide details about their DCM. DCM characteristics included age, gender, history of surgery, best daily limb pain score (using a visual analogue scale), duration of symptoms and disease severity as measured using the self-reported, patient-derived, modified Japanese Orthopaedic Association [**P**-mJOA]. [25] The modified Japanese Orthopaedic scale [mJOA] is amongst the most commonly utilised assessments of disease severity[19]·[20] and is fully validated.[26] It is a composite score based on upper limb function, lower limb function, upper limb sensation and bladder function. The score is valid for analysis in its entirety or per domain. Originally developed as an investigator administered tool, it has recently been

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adapted and validated for use by patients. [25] All questions were mandatory, but respondents were not required to rank every recovery domain, on the basis that some domains may not be a priority for them. The sequence of questions and order of responses was not altered from respondent to respondent.

Survey administration

The survey was accessed via a landing page on Myelopathy.org, allowing assessment of response rates using Google Analytics (California, USA). Individuals with DCM were recruited over an eighteen-month period. The recruitment process has been described in detail previously[27] but in short, the survey was advertised using Google Adwords (California, USA) and through Myelopathy.org and its social media outlets. The survey was voluntary and internet protocol addresses were used to prevent users submitting multiple responses. A missing data analysis was conducted between complete and incomplete survey responses to consider if particular subgroups were more likely to terminate early. Complete responders were defined as having provided answers for all aforementioned variables.

Analysis

Research priorities are presented using summary statistics, including average ranking and overall proportion of patients per domain. Domains which were not ranked by a respondent were omitted from these scores. For subgroup analysis, variables were dichotomised and thresholds were chosen based on the graphical distribution of responses and sample sizes. Categorical variables were compared using the Chi-Squared test. For continuous variables, the Shapiro-Wilks test was used to assess for parametric distribution of data sets. The Mann-Whitney U test was then used to compare the means of non-parametric distributions whilst a two-tailed T-test used to compare the means of parametric distributions. Pearson's correlations were performed to assess between group differences in characteristics, which could have influenced sub-group analysis. Significance was set at p < 0.05.

Patient and Public Involvement

Patients were involved in the design, development, recruitment and conduct of this study. At a patient and public involvement day hosted at the University of Cambridge, a focus group of DCM patients evaluated and confirmed the recovery domains in DCM. DCM patients were used to pilot the subsequent survey, including optimising its design to reduce the time taken to complete and clarify questions. The online survey for the study was hosted on Myelopathy.org, an international DCM charity run largely by DCM patients. Patients were therefore active in disseminating the survey via online DCM support groups, including

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Myelopathy Support, led by Iwan Sadler, patient and co-author. Patients who were involved in preparation of the manuscript are amongst the authors. In addition, all patients who participated in the research are recognised in the acknowledgement statement. DCM patients are involved in plans to disseminate this research to the patient community, including blog articles on Myelopathy.org, posts in online patient support groups and presence at spinal conferences in the United Kingdom
Results

Respondents

The survey was uniquely accessed 1463 times, with 659 visitors entering the survey (participation rate of 33%). A total of 481 responses contained complete data (completion rate 73%). A missing data analysis was conducted comparing incomplete and complete responses. Patients who completed the survey in full were more likely to have undergone surgery (p = 0.04), otherwise there was no statistical difference within variables of interest (Supplementary Data 1). Only complete responses were analysed in the present study. Of these responses domains were ranked more than 80% of the time: pain (400, 83%), sensation (428, 89%), walking (396, 82%), arm and hand (393, 82%), sexual (388, 81%), bladder and bowel (399, 83%) and trunk function (407, 85%).

On average respondents were more likely to be female (341, 70%) and suffer with moderate myelopathy (11.9 ±3.0) for between 3 and 10 years (181, 31%). Around half of patients (221, 46%) had undergone surgery. Overall respondent demographics are summarised in Table 1. Considering group differences, patients who had suffered from the disease for longer were more likely to have undergone surgery (p < 0.01) and have worse myelopathy (-0.22, p < 0.005). They were also more likely to suffer more pain (-0.14, p < 0.01). Average pain scores were 3.1 (±2.4) for patients suffering with the disease for less than a year, rising to 4.5 (±3.0) for patients suffering for at least 10 years. There was no relationship between severity of myelopathy and pain scores (-0.04, p = 0.36). Between group differences are summarised in Supplementary Data 2.

Ranking of spinal cord dysfunction domains

Overall, pain was the most popular number one ranked recovery domain, chosen by 39.9% of respondents. This was followed by walking (20.2%), sensation (11.9%) and arm and hand function (11.5%). Sexual function (5.7%), bladder and bowel (3.7%) or trunk function (3.5%) were chosen less frequently. When considering the average ranking of symptoms, whilst pain remained the priority (2.6±2.0), this was closely followed by walking (2.9±1.7) and arm/hand function (3.0±1.4) (Figure 1). Sensation ranked lower (4.3±2.1).

Impact of baseline characteristics on ranking of spinal cord dysfunction domains

Respondents who had undergone surgery were more likely to prioritise walking (p < 0.005) and trunk function (p = 0.03), whereas patients who had not yet undergone surgery were more likely to prioritise upper limb function (p < 0.05) (Figure 2). Patients with poor upper

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limb or lower limb function were more likely to prioritise arm/hand recovery (p < 0.005) and walking (p < 0.005) respectively (Figure 2). Overall, pain remained the priority, with the exception of patients with greater walking impairment (p < 0.005), even amongst patients with lower pain scores (Figure 2).

When considering the average rankings pain, arm/hand function and walking remained the top three recovery priorities (Figure 3). However, amongst the subcategories, the order of these priorities differed slightly (Supplementary Data 3). Patients who were male, or who had undergone surgery, or who had greater lower limb or bladder functional disability, prioritised recovery of walking, over pain and arm/hand function; patients with greater upper limb function or sensory disability prioritised recovery of arm/hand function over pain and walking.

When overall P-mJOA scores were considered to evaluate mild, moderate and severe patients,⁶ no variation was seen in modal or average ranked priorities.

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Discussion

This is the first study to systematically survey functional domains relevant to DCM and to ask patients to rank them in order of importance to their quality of life. The established priorities are likely to reflect symptom prevalence and their impact on day to day life.[23] The analysis of 481 completed answers demonstrated that pain, arm/hand function and walking emerge as the most important spinal cord dysfunction domains. Although based on averaged rankings, there were some subtle differences in ordering of these three domains. With the exception of patients with significant gait impairment, elimination of pain was the recovery priority independent of baseline characteristic.

These findings are surprising: functional disability (specifically recovery of arm/hand and walking function) has been and continues to be a focus for researchers, typically in response to surgery,[8] but more recently with a shift towards enhancing post-surgical recovery.[13]·[27] In contrast, pain is not widely recognised as an important relevant domain. Our recent review of outcome reporting in DCM clinical trials demonstrated that the overwhelming majority of studies (90%) reported outcomes related to function, but only 27% of studies reported outcomes related to pain,[19] despite the fact that pain is a well-recognised feature of DCM,[5] which often improves following surgery.[11] The present findings highlight the fact that systematic research of patient needs is sorely lacking in DCM. A possible explanation for this discrepancy is that surgeons, who play a significant role in the management of DCM and predominate this research field, remain biased towards functional domains because pain is not a recognised indication for surgery in DCM.[6]

The priorities established in the present study differ from those of individuals suffering from spinal cord injury. Although pain is amongst the most prevalent symptoms of traumatic spinal cord injury,[28]·[29] the "elimination of chronic pain" was considered to be a relatively low priority amongst those surveyed in Anderson's study[23] and a similar study by Kwon et al.[30] that focused on the priorities for SCI recovery after novel treatments (e.g. stem cells). Instead, quadriplegics prioritised arm/hand function, whilst paraplegics sexual and bladder/bowel function. These differences relate to their specific significance for patient independence and quality of life.

In DCM, the symptom burden is less well-described[31]·[32] and the relationship between symptom burden or their significance with respect to quality of life in DCM has not been investigated. However, it would seem likely a similar relationship exists.

Limitations

 Following recommendation by the James Lind Alliance, which was founded to support priority setting in research,[33]·[34] the present survey was conducted online, as previously described through a DCM charity, Myelopathy.org.[27] Respondents belonged to a self-selecting group of individuals who were asked to confirm they had a diagnosis of DCM by a medical professional, after being presented with an explanation of the disease for verification purposes. It is possible that some respondents did not have DCM. Reassuringly, respondent demographics were comparable to those of leading prospective surgical studies, with the exception of gender which was not shown to influence patient priorities[8]·[9] (Supplementary Data 1). This likely reflects the recognised popularity of online health communities amongst females. There are no such comparable series for non-surgical cohorts, but their inclusion provides a further valuable perspective.

The survey questions were not randomly sorted and therefore each respondent answered identical surveys with spinal cord function domains presented in the same order. The last domain assessed was sensation. In keeping with it being the most prevalent DCM symptom,[32] it featured most frequently in the responses, indicating that the order of domains was unlikely to have influenced the rankings. Moreover, answers to demographic questions, which followed the ranking of priorities on the survey, were required to define a complete response in order to be included in the present analysis. Priorities therefore were not influenced by incomplete answers.

Following the qualitative development work and the previous experience of Anderson et al., 2004, the pain domain was kept non-specific, asking patients to rank 'elimination of pain' as a recovery priority (Supplementary Data 4). In contrast however, the pain assessment focused on limb pain, which is classically felt to represent DCM-related pain.[5] Whilst this does not limit the implications of our findings as whole, their interpretation will require a better characterisation of pain in DCM in order to focus research appropriately as other pain foci are reported.[35]

Conclusion

The priorities reported in the present study identify functional domains that are relevant to the quality of life of DCM patients. They provide an important framework for future research

and will serve as a valuable reference for the development of a core outcome set relevant to
studies in DCM.

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Table

Table 1: Summary of respondent demographics.

Respondent Demographics	
Age (Mean ± SD)	53.6 (9.8)
Male Gender (%)	140 (29)
Undergone Surgery (%)	221 (46)
Length of Symptoms (%)	
0 to 1 year	72 (15)
1 to 3 years	140 (29)
3 to 10 years	181 (38)
10 to 25 years	74 (15)
25+ years	14 (3)
P-mJOA (Mean +SD)	
Upper Limb Function	3.6 (1.0)
Walking	4.4 (1.5)
Upper Limb Sensation	1.7 (0.7)
Bladder Function	2.2 (1.0)
Total	11.9 (3.0)
VAS Limb Pain (Mean ± SD)	3.1 (2.6)

Figure Legends

Figure 1: Overall recovery priorities. The bar chart represents the first choice of patients and the line graph the average ranking for each domain (where the top ranking is 1). Pain was the overall first choice priority of patients, although when priority rankings were averaged, this was closely followed by walking and arm/hand function.

Figure 2: Impact of baseline characteristics on first choice recovery priority. The bar chart represents the first choice of patients. Significant between group differences are denoted by the * symbol. For simplicity, groups were dichotomised as follows: Duration <3years, P-mJOA upper limb function <3, P-mJOA lower limb function <4 or feeling <2, P-mJOA bladder/bowel function <2 and VAS limb pain <3. Those who had undergone surgery were more likely to choose trunk function (p=0.03) or walking function (p<0.005), whereas those who had not yet undergone surgery were more likely to choose arm/hand function (p<0.005). Equally patients with more impairment of walking (p<0.005) or arm/hand function (p<0.005) were more likely to prioritise these domains. Pain remained the priority even in patients reporting less pain.

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Figure 3: Impact of baseline characteristics on recovery priority average rankings. The scatter plot represents the mean ranking for each subgroup investigated. The blue line represents the overall average. Despite some discrepancies between subgroups, pain, arm/hand and walking function were consistently the top three priorities for patients. Bladder/bowel function was not a recovery priority.

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Declaration Statement:

JH reports being a Medical Advisor for Depuy Synthes and Ethicon, being an Educational Speaker at Globus Medical and research funding from AO Spine. MGF reports consulting for Fortuna Fix. MRK declares a grant from the National Institute for Health Research, travel support from AO Spine and is founder of Myelopathy.org, the first charity for patients with cervical myelopathy. The remaining authors have nothing to declare.

Author Contributions Statement:

BD, OM, IS, BA, BK, SK, JH, JW, RG, MF and MK were involved in the interpretation, drafting and final approval of the manuscript. Additionally, authors BD, IS and MK were involved in the conception, design and acquisition of data for this study, whilst authors BD and OM conducted the data analysis.

Data Availability Statement:

All relevant data to the study are included in the article or uploaded as supplementary information. No additional data are available.

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Figure 1: Overall recovery priorities. The bar chart represents the first choice of patients and the line graph the average ranking for each domain (where the top ranking is 1). Pain was the overall first choice priority of patients, although when priority rankings were averaged, this was closely followed by walking and arm/hand function. BMJ Open: first published as 10.1136/bmjopen-2019-031486 on 10 October 2019. Downloaded from http://bmjopen.bmj.com/ on June 12, 2025 at Agence Bibliographique de I Enseignement Superieur (ABES) . Protected by copyright, including for uses related to text and data mining, Al training, and similar technologies.

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Figure 2: Impact of baseline characteristics on first choice recovery priority. The bar chart represents the first choice of patients. Significant between group differences are denoted by the * symbol. For simplicity, groups were dichotomised as follows: Duration <3years, mJOA upper limb function <3, mJOA lower limb function <4 or feeling <2, mJOA bladder/bowel function <2 and VAS limb pain <3. Those who had undergone surgery were more likely to choose trunk function (p=0.03) or walking function (p<0.005), whereas those who had not yet undergone surgery were more likely to choose arm/hand function (p<0.05). Equally patients with more impairment of walking (p<0.005) or arm/hand function (p<0.005) were more likely to prioritise these domains. Pain remained the priority even in patients reporting less pain.





Figure 3: Impact of baseline characteristics on recovery priority average rankings. The scatter plot represents the mean ranking for each subgroup investigated. The black line represents the overall average. Despite some discrepancies between subgroups, pain, arm/hand and walking function were consistently the top three priorities for patients. Bladder/bowel function was not a recovery priority.

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Supplementary Data 1: Results from the missing data analysis. Patients completing the survey in full, were more likely to have undergone surgery than those who did not (p = 0.04). Data is presented as mean +/- standard deviation, unless specified as a percentage. Numbers within brackets indicate data points for the respective variable with incomplete data.

	INCOMPLETE	COMPLETED	Р
	SURVEYS	SURVEYS	
	(N<178)	(N=481)	
%MALE	16.3% (7/43)	28.9%	.076
AGE	55.1 +/-10.9 (43)	53.6 +/- 9.9	.344
%SURGERY	35.9% (46/128)	46.0%	.043
LENGTH OF	19% (128)	73%	.304
SYMPTOMS (YRS)			
0-1	18.0% (23)	15.0% (72)	
1-3	21.9% (28)	29.1% (140)	
3-10	35.2% (45)	37.6% (181)	
10-25	18.8% (24)	15.4% (74)	
25+	6.3% (8)	2.9% (14)	
LIMB PAIN VAS	3.6 +/- 1.9 (17)	3.7 +/- 2.5	.854
MJOA	12.2 +/- 3.2 (47)	11.9 +/- 3.0	.364

 BMJ Open BMJ Open Supplementary Data 2: Summary of group differences between investigated variables. Highlighted cells representing of ficant differences in the proportion of respondents per group. Further analysis revealed that differences followed a logical course: patients who had had symptoms for longer or undergone surgery were more likely to have severe disease. In addition, patients with more severe disease were likely to have higher boom and scores.

											<u>a 7 0</u>							
Ν	Age (±	SD)	Male (%)	Under	rgone	Length	of	mJO/	A Upper		A Lower	mJO	A	mJO)A	Mean	VAS
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							(70)		~0		ext stur		<2	ballon	i un			
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											ieu ieu							
341	52.8	9.5			157	46%	141	41%	38	11%	ăŭ	31%	143	42%	75	22%	205	60%
											nini ES						135	61%
221	53.1	8.7	64	29%			83	38%	31	14%	<u>200</u>	41%	101	46%	53	24%		
260	54	10.7	74	28%			129	50%	27	10%	68	26%	92	35%	53	20%	158	61%
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123	53.2	0.8	110	28%	100	15%	100	17%				26%	1/1	33%	76	18%	212	04%
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193	53.7	9.1	48	25%	101	52%	78	40%	52	27%		47%			67	35%		
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105	54 52 5	9.6	29	28%	53	50%	40	38%	29	28%	2 61 2 0	58%	67	64%			58	55%
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BMJ Open BMJ Open Supplementary Data 3: Mean ranking for recovery domains for baseline characteristics. Based on average rankings, the top ranked domain is highlighted. Whilst pain, walking and arm/hand function remained the priorities, for respondents who were male or had undergoine surgery, or had impaired upper, lower

or bladder function, arm/hand function had the top ranking. For patients with impaired upper limb sensation, walking function was the priority.

	Pain (SD)		Pain (SD)		ain (SD) Walking (SD)		Arm/Hand		Sexual Function		Bagogr/Bowel		Trunk Function		Sensation (SD)	
					Functio	on (SD)	(SD)		(99) 39 9		(SD)					
Gender, Male	3.1	(2.2)	3.2	(1.6)	2.6	(1.6)	4.2	(1.8)		(1.7)	4.4	(1.6)	4.2	(2.3)		
Gender, Female	2.5	(2.0)	2.9	(1.4)	3.0	(1.7)	4.1	(1.6)	5% 25 2	(1.6)	4.6	(1.5)	4.4	(2.0)		
Undergone Surgery	2.8	(2.1)	3.0	(1.4)	2.6	(1.6)	4.1	(1.6)	5 ageria	(1.6)	4.6	(1.6)	4.5	(2.0)		
Not Undergone Surgery	2.6	(2.0)	2.9	(1.5)	3.2	(1.7)	4.2	(1.7)	5 ରୁ ଜିଙ୍କ ପ	(1.7)	4.4	(1.5)	4.2	(2.1)		
mJOA Upper Limb Function <3	2.9	(2.0)	2.1	(1.3)	2.5	(1.3)	3.7	(1.6)	from h (ABES tanhir	(1.7)	4.4	(1.3)	4.2	(2.1)		
mJOA Upper Limb Function >3	2.6	(2.0)	3.1	(1.5)	3.0	(1.7)	4.2	(1.7)	s) ing, A	(1.7)	4.5	(1.6)	4.3	(2.1)		
mJOA Lower Limb Function <4	2.8	(2.0)	2.9	(1.5)	2.4	(1.5)	4.1	(1.6)	njope 5 trajni	(1.7)	4.6	(1.4)	4.3	(2.1)		
mJOA Lower Limb Function >4	2.6	(2.1)	3.0	(1.5)	3.2	(1.7)	4.2	(1.7)	n.bmj. ng,∑an	(1.7)	4.5	(1.6)	4.3	(2.0)		
mJOA Upper Limb Sensation <2	2.7	(2.1)	2.6	(1.3)		(1.5)	4.2	(1.5)	d Simi	(1.6)	4.5	(1.5)	4.2	(2.1)		
mJOA Upper Limb Sensation >2	2.6	(2.0)	3.2	(1.6)	3.0	(1.8)	4.1	(1.8)	Iarfec	(1.7)	4.5	(1.6)	4.4	(2.1)		
mJOA Bladder Function <2	2.6	(1.8)	2.8	(1.6)	2.5	(1.4)	3.6	(1.4)	6 m t	(1.3)	4.7	(1.6)	4.7	(2.0)		
mJOA Bladder Function >2	2.7	(2.1)	3.0	(1.5)	3.1	(1.7)	4.3	(1.7)	5,00 N	(1.7)	4.5	(1.5)	4.2	(2.1)		
Length of Symptoms <3 years	2.9	(2.2)	3.0	(1.5)	3.1	(1.6)	4.2	(1.7)	5 gg 25	(1.7)	4.6	(1.5)	4.1	(2.1)		
Length of Symptoms >3 years	2.5	(1.9)	2.9	(1.4)	2.8	(1.7)	4.1	(1.7)	5.8 🛱	(1.6)	4.5	(1.5)	4.5	(2.1)		
Best Limb Pain VAS <3	2.7	(2.1)	3.0	(1.5)	2.9	(1.7)	4.2	(1.7)	5.7 B	(1.7)	4.4	(1.5)	4.6	(2.0)		
Best Limb Pain VAS >3	2.5	(2.0)	2.9	(1.5)	3.0	(1.6)	4.1	(1.6)	5.8 B	(1.6)	4.7	(1.6)	3.9	(2.2)		
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Supplementary Data 4: Questionnaire. The questions relevant to this study were developed, piloted and embedded within an ongoing initiative investigating patient reporting of DCM. The questions pertaining to the data points required for this study are presented, including their question number and options for selection. Question 35 about age was the only question where respondents were asked to type in a specific answer. The answer format was electronically validated to require an integer, prompting users to specify to the nearest year.

Questions	generating data points required for this study
5	How long have you suffered with cervical myelonathy?
U	
	0-1 year
	1-3 years
	3-10 years
	10-25 years
	>25 years
7	Have you undergone surgery for cervical myelopathy?
	Yes
	No
23	Currently, please indicate the intensity of the current, best and
	worst pain affecting your arms or legs over the past 24h on a scale of 0
	(no pain) to 10 (worst pain imaginable)
	· L
	0
	2 3
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	5
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20	10
29	How does cervical myelopathy affect your arms and hands? Choose the
	statement that best fits: I am
	- Unable to move my hands
	- Unable to eat with a spoon but am able to move my hands
	 Unable to button my shift but able to eat with a spoon Able to button my shift with great difficulty
	- Able to button my shirt with slight difficulty
	- Not having any trouble using my hands.

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30	How does cervical myelopathy affect your legs? Choose the statement
	that best fits: I am
	 Completely unable to move legs at all and have no feeling in legs Having feeling in legs but not able to move them at all Able to move my legs but am unable to walk Able to walk on flat floor with a walking aid (cane or crutch) Able to walk up and/or downstairs with the aid of a handrail Able to walk up and/or downstairs without handrail but I notice moderate-to-significant lack of stability/feeling of imbalance when I walk Able to walk unaided (no crutches, canes, walker) with smooth reciprocation (ie, legs move smoothly) but I still notice mild lack of stability/feeling of imbalance when walking Able to walk without any problems of imbalance or instability
31	How does cervical myelopathy affect your arms and hands? Choose the
	statement that best fits: I have
	- Complete loss of feeling in hands
	- Severe loss of feeling, or have pain in my hands
	- Mild loss of feeling in hands
	- No loss of feeling in hands
32	How does cervical myelopathy affect your bladder? Choose the
	statement that best fits: I am
	- Am completely unable to control urination
	- Have marked difficulty controlling urination
	- Have mild to moderate difficulty controlling urination
	- No difficulty controlling urination
33	Effective medical research should target the needs of patients. The
	consequences of spinal cord injury can be classified into 7 different

	categories. In DCM, the patient priorities are not known.
	For you as a patient, what are the research priorities for you (please
	rank where 1 is the most important and 7 is the least important)? What
	would you like researchers to focus on?
	-Elimination of Pain -Arm/Hand Function -Walking Function -Bladder/Bowel Function -Sexual Function
	-Upper Body/Trunk Function -Normal Sensation
34	Are you male or female?
	Male
35	How old are you?
	C.

STROBE Statement

			BMJ Open Steel BMJ Open Steel BMJ Open	Page 30 of
1 2			STROBE Statement	
3 4	Section/Topic	Item No	Recommendation	Reported on Page No
5 6	Title and abstract	1	(a) Indicate the study's design with a commonly used term in the title or the abstract	2
7		-	(b) Provide in the abstract an informative and balanced summary of what was done and what was for the stract and the stract and balanced summary of what was done and what was for the stract and the str	2
8	Introduction			
9 1(Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	4-5
11	Objectives	3	State specific objectives, including any prespecified hypotheses	5
12	Methods		teeme	
1:	1 Study design	4	Present key elements of study design early in the paper	6-7
15 16	Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, following and data collection	6-7
18 19 20 21 22 23	3 9 0 1 Participants 2	6	(a) Cohort study—Give the eligibility criteria, and the sources and methods of selection of participation of the secret be methods of follow-up Case-control study—Give the eligibility criteria, and the sources and methods of case ascertainment of the choice of cases and controls Cross-sectional study—Give the eligibility criteria, and the sources and methods of selection of participation of the secret because of	6-7
24 25	4 5		(b) Cohort study—For matched studies, give matching criteria and number of exposed and unexposed Case-control study—For matched studies, give matching criteria and the number of controls per case	
26 27 28	7 Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	6-7
29 30	Data sources/measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measuremetit). Bescribe comparability of assessment methods if there is more than one group	6-7
3	Bias	9	Describe any efforts to address potential sources of bias	6-7
33	3 Study size	10	Explain how the study size was arrived at	6-7
34	4 Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which group ness were chosen and why	6-7
35			(a) Describe all statistical methods, including those used to control for confounding	7
37	7		(b) Describe any methods used to examine subgroups and interactions	7
38	3		(c) Explain how missing data were addressed	7
39 40 41 41	Statistical methods	12	(<i>d</i>) Cohort study—If applicable, explain how loss to follow-up was addressed <i>Case-control study</i> —If applicable, explain how matching of cases and controls was addressed <i>Cross-sectional study</i> —If applicable, describe analytical methods taking account of sampling strategy	7
43	- 3		(e) Describe any sensitivity analyses	
44 45 46 47	1 5 5 7		For peer review only - http://bmjopen.bmj.com/site/about/guidelines.xhtml	1

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1 2 3 4	Section/Topic	Item No	Recommendation	Reported on Page No
5	Results			
6 7			(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined of eligibility, confirmed	0
, 8	Dentisinanta	12*	eligible, included in the study, completing follow-up, and analysed	8
9	Participants	13*	(b) Give reasons for non-participation at each stage	N/A
10			(c) Consider use of a flow diagram	N/A
12 13			(a) Give characteristics of study participants (eg demographic, clinical, social) and information on examples and potential confounders	8
14 15	Descriptive data	14*	(b) Indicate number of participants with missing data for each variable of interest	Supplementary Data 2
16 17			(c) Cohort study—Summarise follow-up time (eg. average and total amount)	N/A
18			Cohort study—Report numbers of outcome events or summary measures over time $\vec{a} \geq \vec{c}$	N/A
19	Outcome data	15*	Case-control study—Report numbers in each exposure category, or summary measures of exposure	N/A
20			Cross-sectional study—Report numbers of outcome events or summary measures	12-13
21			(a) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision ($e \ge 95^{\circ}$ confidence interval).	
23			Make clear which confounders were adjusted for and why they were included	8
24	Main results	16	(b) Report category boundaries when continuous variables were categorized	Supplementary
25 26				Data 1,3 & 4
27			(c) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	N/A
28	Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	8-9
29 30	Discussion		ar te	
31	Key results	18	Summarise key results with reference to study objectives	10
32 33 34	Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss b oth direction and magnitude of any potential bias	11
35 36	Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses results from similar studies, and other relevant evidence	10-11
37	Generalisability	21	Discuss the generalisability (external validity) of the study results	10-11
39	Other Information			
40 41 42	Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	1
43	*Give information separate	ely for case	es and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-	
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