

EXPLORING EXPERIENCES OF THE ILLNESS, ITS CARE AND RECOVERY FOLLOWING GUILLAIN-BARRE SYNDROME - FINAL REPORT

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Short title: Exploring experiences of Guillain-Barré Syndrome

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EXECUTIVE SUMMARY

This study was commissioned and developed in discussion with the chief executive and chair of the board of trustees of the charity, GAIN ('Guillain-Barré and associated Inflammatory Neuropathies') to explore the experiences of the illness, its care and how people with Guillain-Barré Syndrome (GBS) and Chronic Inflammatory Demyelinating Polyneuropathy (CIDP) living in the United Kingdom and Republic of Ireland can return to their lives following discharge from hospital, functioning as well as possible from a physical, psychological and social perspective.

We conducted a series of studies: a systematic review of the published research on the experiences and views of people living after GBS and its variants; an interview study exploring experiences of the illness, its care and return to function in more depth; a survey to gather information more widely from people with these conditions; and the findings from each were brought together at the end.

These parts of the study were linked, each informing the next.

In the first investigation, the systematic review, we aimed to review qualitative studies of patients' experiences and perceptions of GBS (and variants) at diagnosis, discharge and during recovery. We searched twelve electronic databases, supplemented with internet searches and forward and backward citation tracking from the included studies and review articles. Data were synthesised thematically, and the quality of included studies was assessed. We identified 5,282 citations and after removing duplicates and excluding citations based on title and abstract, and full-text screening, included five studies, all of acceptable quality, in the review and metasynthesis. Through constant discussions and an iterative approach, we developed six analytical themes following a patient's journey from suspecting that they had a health problem, through to being hospitalised, experiencing ongoing difficulties, slowly recovering from GBS, adjusting to their new circumstances, and re-evaluating their lives. Being diagnosed with and surviving GBS was a life-changing experience for all participants.

In the second study we aimed to explore the experiences of people with GBS in the UK using qualitative (face-to-face and telephone) interviews. Audio-recorded data were transcribed verbatim and analysed using the Framework Method supported by NVivo 11. We purposively recruited a sample of 16 volunteers with a prior diagnosis of GBS of varying age, sex, ethnicity, location, marital status, time since diagnosis and length of hospital stay to maximize differences in experience. Interviewees were required to have been discharged from hospital, able to give informed consent, able to speak and understand English and currently resident in the United Kingdom. The key themes arising from the analysis were as follows: the importance of early diagnosis; the experiences of inpatient care; the importance of active support for recovery; the need for communication throughout the course of the illness; the need for greater

awareness, knowledge and provision of information by healthcare staff; and path to achieving function. This, the first qualitative study exploring experiences of people with GBS in the UK through their whole illness journey, from onset to recovery, helped to increase our understanding of the experiences and support needs of people recovering from GBS.

In the final study we aimed to develop and validate a survey to quantify the experiences of people with GBS. We employed a cross-sectional design using a self-administered online questionnaire survey. The questionnaire was based on the systematic review and qualitative study. It included question domains covering experiences of GBS, symptom severity at each stage, healthcare, and factors that supported or hindered recovery. People with a previous diagnosis of GBS were invited to participate in the online survey through a social media advert. We conducted descriptive, exploratory factor and reliability analyses and undertook multivariable regression analysis to investigate relationships between variables of interest, explore reliability and validity of the questionnaire, and identify factors predicting recovery. There were 291 responders including people with previous GBS of different sex (male 51.9%, female 48.1%) and marital status (married, 61.1%, civil partnership 19.6%, single or undeclared 12.0%). Most responders were from the UK (86.6%); 43% were diagnosed between 2015 and 2019 and 61.7% within 7 days of onset compared with later (38.3%). First medical help was more often from a GP (62.2%) than the Emergency Department (25.6%). Factor analysis showed four main scales, with excellent internal consistency for symptoms ($\alpha > 0.9$), good for information provided and for factors affecting recovery ($\alpha > 0.8$), and acceptable for care received ($\alpha \geq 0.7$). A combination of physical, psychological and social factors was associated with recovery. Factors considered by responders to be most important for recovery were positive social interactions, physical activity including physiotherapy and movement, changes made at home, and immunoglobulin treatment. Responders were more satisfied with information provided by specialists rather than non-specialists. Multivariable models showed that immunoglobulin and /or plasma exchange treatment were significant predictors of recovery. Employment and recovery factors combined (positive interactions, work support and changes at work or home, physical activity and therapy) were associated with recovery, but did not reach statistical significance. We found that the questionnaire demonstrated good internal reliability of scales and subscales and findings also supported the construct validity of the questionnaire for people with GBS.

Together, these studies provide key new evidence needed by the charity to provide support and advice to people with GBS and related conditions to help them return to good function. We hope it will also provide valuable information for patients, their relatives, health professionals, commissioners of services and academics with an interest in this field. The questionnaire can be repeated in future and can be used to see whether changes in care or support to people with GBS improve their experiences.

BACKGROUND

GAIN - Guillain-Barré and associated Inflammatory Neuropathies, is a charity 'governed by a voluntary board of trustees, most of whom have direct experience of living with Guillain-Barré Syndrome (GBS) or Chronic Inflammatory Demyelinating Polyneuropathy (CIDP), either because they have had the condition themselves, or because someone close to them has.' The trustees, who are responsible for the Charity's strategy and policies and, ultimately, for everything the Charity does on behalf of people affected by GBS, CIDP and associated inflammatory neuropathies, approached researchers at the University of Lincoln to discuss ideas for research to support GAIN's aim of providing support and advice to people with GBS and CIDP.

A meeting took place on 14 May 2018 between Caroline Morrice, chief executive of GAIN, Chris Fuller, chair of the board of trustees, one of the trustees, Susan McAllister, and academics from the University of Lincoln to discuss the development of a research proposal based on initial ideas presented by the trustees. The ideas involved exploring patients' experience of care, particularly focussing on the period following discharge from hospital and return to the community, from a physical, psychological and social perspective.

The study was envisaged to involve three stages: firstly, a literature review; secondly, a qualitative study; and finally, a survey. It was expressed during the meeting and in a document circulated beforehand that the research needed to be time-limited and focussed, but rigorously designed and conducted, so that it would inform the trustees on how they could, as a charity, best support the needs of patients, their family and carers, but also so that the findings would be beneficial to service users, patient groups, academics and commissioners of services.

INTRODUCTION

Guillain Barré Syndrome (GBS) is the commonest form of acute polyneuropathy with an incidence of 1-2/100,000 per year.[1] It is more common in males and with increasing age in both sexes, affecting around 100,000 people per year worldwide.[2] It has an acute (4 hours) to subacute (up to 1 week) onset with symmetrical weakness and numbness of the limbs progressing proximally usually over 2-4 weeks, causing loss of reflexes. There are several variants of GBS, including Miller-Fisher syndrome, which affects the cranial nerves and brainstem, but the diagnosis is largely made clinically.[3] The severity is variable with mild cases causing little disability and recovering spontaneously, but in 20-30% of cases a more severe generalised form rapidly progresses to affect facial and respiratory muscles, and causes autonomic dysfunction with sweats, cardiac arrhythmias, hyper- or hypo-tension, and, in 5%, even death.[2]

In two thirds of cases GBS is preceded by a diarrhoeal or upper respiratory infection, the commonest micro-organism implicated being *Campylobacter jejuni* (25-50%). Other heralding infections include cytomegalovirus (CMV), Epstein-Barr virus, influenza A, *Mycoplasma pneumoniae*, and *Haemophilus influenzae*, but in a third, no cause is found. Preceding infection explains the seasonal variation in GBS,[4] and also the mechanism of demyelination or axonal (nerve) damage in GBS, which is thought to be due to antibodies directed against the microorganism liposaccharides in the cell wall cross-reacting with neuronal or myelin gangliosides in particular GM1 and GD1a, through so-called 'molecular mimicry'. [2, 5]

Effective treatment, particularly for more severe or rapidly progressing cases, involves intensive care, supportive therapy for respiratory and cardiac problems, intravenous immunoglobulins (IVIg) or plasma exchange, with steroids being less effective. [6, 7] Prevention of complications such as pneumonia, deep vein thrombosis, urinary retention or bowel obstruction is important.[2, 5] Other treatments have not been shown to be definitively effective.[8] Although people with milder forms of GBS can recover completely within a year, many of those affected will have pain and fatigue, 20% will be unable to walk without an aid at 6 months, most have residual pain and fatigue,[2, 5] and others have sleep disturbances, anxiety or post-traumatic stress disorder.[9] These symptoms can affect work or other activities, although recovery can continue even after 3 years or more. There is lack of evidence for longer term care for pain[10] or exercise interventions.[11]

Chronic inflammatory demyelinating poly(radiculo)neuropathy (CIDP) is a closely related condition distinguished by a longer duration of onset.[12] It affects 0.5-1 people per 100,000 per year and is also thought to be immunological in origin, presenting with sensory and motor symptoms in the distal and proximal segments of the limbs over more than 8 weeks, with absent reflexes, raised protein concentrations in the cerebrospinal fluid, and heterogeneous slowing of nerve conduction being typical of the condition.[12] The disease course and disability is often variable resulting in the condition being underdiagnosed. There is weak evidence for effectiveness of steroids; intravenous immunoglobulins reduce disability short-term, but with more adverse effects; plasma exchange has similar short-term effects to immunoglobulins; and the immunosuppressant drugs tested so far have been largely ineffective.[13]

Although a number of qualitative studies have been published of GBS sufferers' experiences of their initial illness,[14] initial recovery,[15], return to work[16] ongoing recovery,[17] and use of healthcare or other support,[18] most studies have been conducted in Scandinavia, with limited research on the experiences and needs of people in the UK or elsewhere. A systematic review of health related quality of life in GBS[19] found that most studies were of low quality and there was a need for 'high-quality studies on patterns of patient-perceived recovery after GBS onset.'

We conducted a systematic review and metasynthesis of qualitative studies, followed by a qualitative study in the UK setting, and from these we developed and validated a survey designed to elicit people's experiences of illness and recovery, together with their health, psychological and social needs during rehabilitation.

AIM

Our overall aim was to explore the experiences and return to function of people with Guillain-Barré Syndrome.

OBJECTIVES

Our objectives were:

1. To review patients' experiences of GBS and CIDP at diagnosis, discharge and during recovery by conducting a systematic review and thematic metasynthesis of qualitative studies of patients' experiences of GBS and CIDP.
2. To explore experiences particularly focussing on the period post-discharge from hospital and return to the community, of people living in the United Kingdom and Republic of Ireland who have had GBS and CIDP, from physical, psychological (cognitive and emotional) and social, including occupational (e.g., job stability and security, financial difficulties, reduced work capacity), perspectives using qualitative (individual or focus group) interviews to understand health and social care needs, facilitators and barriers to recovery and return to function.
3. To develop, administer and validate a questionnaire survey to elicit the range of experiences, health and social care needs, and facilitators and barriers to recovery and return to function of people with GBS and related conditions, particularly those resident in the United Kingdom and Republic of Ireland.
4. To integrate and report on findings from 1-3 above and to report on these.

METHODOLOGY

We used a mixed methods sequential design [20] with the following component work streams:

1. Systematic review and qualitative metasynthesis.
2. Qualitative interview study.
3. Self-administered questionnaire survey of people with GBS and related conditions.
4. Integration and synthesis of data from 1-3 to produce a report to inform recommendations for support.

The first two studies have been published, so we have included the abstracts below and links to the full articles which are available through open access. The third study has been submitted for publication and the full report is included below.

1. Patients' experiences and perceptions of Guillain-Barré syndrome: A systematic review and meta-synthesis of qualitative research

Summary

Background: The aim of this study was to review patients' experiences and perceptions of GBS and its variants at diagnosis, discharge and during recovery, by conducting a systematic review and thematic metasynthesis of qualitative studies of patients' experiences of GBS (and its variants).

Methods: We searched twelve electronic databases, supplemented with internet searches and forward and backward citation tracking from the included studies and review articles. Data were synthesised thematically following the Thomas and Harden approach. The CASP Qualitative Checklist was used to assess the quality of the included studies of this review.

Results: Our search strategy identified a total of 5,282 citations and after removing duplicates and excluding citations based on title and abstract, and full-text screening, five studies were included in the review and meta-synthesis; all included studies were considered of acceptable quality. Through constant discussions and an iterative approach, we developed six analytical themes following a patient's journey from suspecting that they had a health problem, through to being hospitalised, experiencing ongoing difficulties, slowly recovering from GBS, adjusting to their new circumstances, and re-evaluating their lives.

Conclusions: Despite the variety of experiences, it was evident from all included studies that being diagnosed with and surviving GBS was a life-changing experience for all participants.

Trial registration: Protocol was registered (CRD42019122199) on the International Prospective Register of Systematic Reviews (<http://www.crd.york.ac.uk/PROSPERO>).

The study was published in the international peer-reviewed journal PLoS ONE and is available at: Laparidou D, Curtis F, Akanuwe J, Jackson J, Hodgson TL, Siriwardena AN (2021) Patients' experiences and perceptions of Guillain-Barre' syndrome: A systematic review and metasynthesis of qualitative research. PLoS ONE 16(2): e0245826. <https://doi.org/10.1371/journal.pone.0245826>.

2. Exploring the experiences of having Guillain-Barré Syndrome: A qualitative interview study

Summary

Background: Evidence describing the experiences of people with GBS is limited. We aimed to explore the experiences of people with GBS in the UK.

Methods: We used qualitative (face-to-face and telephone) interviews to explore experiences of people with GBS. Audio-recorded data were transcribed verbatim and analysed using the Framework Method supported by NVivo 11. We purposively recruited a sample of 16 volunteers with a prior diagnosis of GBS of varying age, sex, ethnicity, location, marital status, time since diagnosis and length of hospital stay to maximize differences in experience. Interviewees were required to have been discharged from hospital, able to give informed consent, able to speak and understand English and currently resident in the United Kingdom.

Results: The key themes arising from the analysis were as follows: the importance of early diagnosis; the experiences of inpatient care; the importance of active support for recovery; the need for communication throughout the course of the illness; the need for greater awareness, knowledge and provision of information by healthcare staff; and path to achieving function.

Conclusion: This is the first qualitative study exploring experiences of people with GBS in the UK through their whole illness journey from onset to recovery. The findings contribute to our understanding of the experiences and support needs of people recovering from GBS.

The study was published in the international peer-reviewed journal *Health Expectations* and is available at: Akanuwe JNA, Laparidou D, Curtis F, Jackson J, Hodgson TL, Siriwardena AN. Exploring the experiences of having Guillain-Barré Syndrome: A qualitative interview study. *Health Expect.* 2020;23:1338–1349. <https://doi.org/10.1111/hex.13116>.

3. Patient symptoms and experience following Guillain-Barré and associated syndromes: questionnaire development and validation

Summary

Background: There is lack of evidence on experiences of people with GBS and how to measure this. We aimed to develop and validate a survey to quantify the experiences of people with GBS.

Methods: We employed a cross-sectional design using a self-administered online questionnaire survey. The questionnaire was developed from a previous systematic review and qualitative study of people's experiences of and recovery from GBS. People with a previous diagnosis of GBS were invited to participate in an online survey through a social media advert. We included question domains covering experiences of GBS, symptom severity at each stage, healthcare, and factors that supported or hindered recovery. We conducted descriptive, exploratory factor and reliability analyses and undertook multivariable regression analysis to investigate relationships between variables of interest, explore reliability and validity of the questionnaire, and identify factors predicting recovery.

Results: The 291 responders included people with previous GBS of different sex (male 51.9%, female 48.1%), marital status (married, 61.1%, civil partnership 19.6%, single or undeclared 12.0%) and country of origin (UK 86.6%). Most (43%) were diagnosed between 2015 and 2019 and within 7 days of onset compared with later (61.7% vs 38.3%). First medical help was more often from a GP (62.2%) than the Emergency Department (25.6%). Factor analysis showed four main scales, with excellent internal consistency for symptoms ($\alpha > 0.9$), good for information provided and for factors affecting recovery ($\alpha > 0.8$), and acceptable for care received ($\alpha \geq 0.7$). A combination of physical, psychological and social factors was associated with recovery. Factors considered by responders to be most important for recovery were positive social interactions, physical activity including physiotherapy and movement, changes made at home, and immunoglobulin treatment. Responders were more satisfied with information provided by specialists rather than non-specialists. Multivariable models showed that immunoglobulin and /or plasma exchange treatment were significant predictors of recovery. Employment and recovery factors combined (positive interactions, work support and changes at work or home, physical activity and therapy) were associated with recovery, but did not reach statistical significance.

Conclusion: The questionnaire demonstrated good internal reliability of scales and subscales. The findings also supported the construct validity of the questionnaire for people with GBS.

Patient symptoms and experience following Guillain-Barré and associated syndromes: questionnaire development and validation

METHODS

Design

We employed a cross-sectional design using a self-administered online questionnaire survey designed to explore symptoms, care experiences and recovery in people who previously had GBS.

Questionnaire development

Questionnaire domains and items were based on a systematic review and metasyntesis of qualitative studies [21] and an interview study of people with the condition.[22] The domains for people who previously had GBS included: participant characteristics; severity of symptoms (physical, psychological and social) at each stage of illness; medical health seeking experience; treatment and care experiences; follow-up and support; and social or work-related experience.

The initial questionnaire was piloted with four people who had recovered from GBS, of which two had taken part in an earlier interview study.[22] The questionnaire was also discussed with the Guillain Barré Syndrome and Associated Inflammatory Neuropathies (GAIN) charity, the Healthier Ageing Patient and Public Involvement (PPI) group at the University of Lincoln and members of the research team. Comments and suggestions were used to revise some of the questions to ensure they were appropriate for the intended population of GBS patients.

Participants recruitment and data collection

Ethical approval was gained from the University of Lincoln Human Ethics Committee (2019-Jul-0738). A convenience sample of people with GBS living in the UK was recruited through a social media advertisement posted on Twitter and the UK GBS charity, Guillain-Barré and Associated Inflammatory Neuropathies (GAIN) website, Facebook page and member list. Information about the research (including consent and a link to the questionnaire) was posted at the GAIN and University of Lincoln Community and Health Research Unit websites accessible to potential participants. Participants, who self-identified with a diagnosis of GBS, consented and completed the questionnaire online. Participants were encouraged to contact a member of the research team (JA) if they needed further information or assistance to complete the questionnaire. The survey remained open for two months (August and September 2019), and once completed by participants, the questionnaire was retrieved and stored securely for analysis.

Data analysis

The internal consistency of the GBS questionnaire was assessed using Cronbach's alpha (α). [23] This test was used to establish the level of agreement between items belonging to the same scale. Four main scales were developed which contained items scored on a 7-point Likert scale including: *symptoms*, *care received*, *factors affecting recovery*, and *information provided*. Some of these scales were divided into further subscales: initial, in hospital, after hospital, and current symptoms, as well as care received in hospital and after discharge from hospital.

Factor analyses were run to identify questionnaire subscales. The scales included were suitable for this type of analysis as indicated by Kaiser-Meyer-Olkin measure (KMO), which was higher than 0.7 for all of them. Retained factors were those with eigenvalues greater than 1 and items with loadings higher than 0.4 [24]. As such, the scales included in the factor analysis were symptoms (initial, in hospital, residual, and current), factors affecting recovery, and information provided.

Multivariate linear regression models were used to identify the factors predicting recovery rate. Two regression models were run: the first one using the scales of the questionnaire as predictors and the second one using the subscales derived from factor analyses as main predictors together with demographic characteristics which might have influenced the outcome. These demographic predictors included age and the binary variables: gender (female or male), employment status (employed or unemployed), living with someone else or alone. The recovery score, which was used as main outcome, was computed using the formula: *recovery score = mean score of in hospital symptoms – mean score of present symptoms*.

The assumption of normality was met as indicated by both histograms and P-P plots of residuals.

Homoscedasticity was present as indicated by scatterplots. The assumption of no multicollinearity was also met for both models as indicated by Durbin Watson tests with values close to 2 (1.93 for the first model and 2.12 for the second model), tolerance values higher than 1 and VIF values smaller than 10.

RESULTS

Responder characteristics

In total, 291 participants responded fully or partially to the questionnaire. Table 1 shows the demographic characteristics of participants who responded. Of the responders 123 (45.6%) were aged between 60 and 79 years; 140 (51.9%) were male and 130 (48.1%) were female. Most were of white ethnicity (264, 97.8%) compared with the minority, who were either BAME (2, 0.7%) or mixed race (3, 1.1%) or other (1, 0.4%). One hundred and seventy eight (65.9%) were married or in civil partnership compared with 57 (21.1%)

single or 35 (13.0%) who did not declare their marital status. At the time of the survey, most participants (252, 86.6%) resided in the UK compared with non-UK residence (39, 13.4%). More participants were retired from work (89, 38%) compared with those in full-time employment (55, 23.5%) or part-time work (31, 13.2%) or those on disability and/or other benefits and not working (30, 12.8%). At the time of the study, majority of participants (177, 65.6%) were living with their spouse compared with other family member (27, 10%) or alone (38, 14.1%) or other (27, 10%).

Disease characteristics of responders are shown in Table 2. Most responders had a diagnosis of GBS (202, 74.8%) compared with CIDP (46, 17.0%) or a related condition (22, 8.1%). Most responders sought help within the first 3 days (158, 60.3%) of feeling unwell and help was most often sought from a GP surgery (163, 62.2%) compared with the Emergency Department (67, 25.6%). Most responders (106, 44.9%) were diagnosed on their first visit with most (166, 64.6%) receiving a GBS diagnosis or its variant rather than a different diagnosis (91, 35.4%).

Overall, 116 (of 291 responders, i.e. 43%) were diagnosed between 2015 and 2019 and time to diagnosis was usually 1-7 days (161, 61.7%) compared with later (100, 38.3%). Responders were generally treated in a hospital general ward or intensive care unit (76, 31.8%) and hospital general ward or regional neurological unit (71, 29.7%).

Table 1 Participant demographic characteristics

Characteristic		Number (N)	Percentage (%)
Age	Below 18 years	2	0.7
	19-39 years	40	14.8
	40-59 years	96	35.6
	60-79 years	123	45.6
	80 + years	9	3.3
	Total	270	100
Sex	Female	130	48.1
	Male	140	51.9
	Total	270	100
Ethnicity	White	264	97.8
	BAME	2	0.7
	Mixed race	3	1.1
	Other	1	0.4
	Total	270	100
Marital Status	Married	168	62.2
	Civil partnership	10	3.7
	Single	57	21.1
	Other	34	12.6
	Prefer not to say	1	0.4
	Total	270	100
Residence	United Kingdom	252	86.6
	Non-United Kingdom	39	13.4
	Total	291	100
Employment status	In full-time time work	55	23.5
	In part-time work	31	13.2
	In work with disability and/or other benefits	6	2.6
	On disability and/or other benefits and not working	30	12.8
	Unemployed	3	1.3
	Retired	89	38
	Other	20	8.5
	Total	234	100
Household status	Spouse	177	65.6
	Other family member	27	10
	Alone	38	14.1
	Other	27	10
	Prefer not to say	1	0.4
	Total	270	100

Table 2 Participant disease characteristics

Characteristic		Number (N)	Percentage (%)
Diagnosis	GBS	202	74.8
	CIDP	46	17.0
	Related condition	22	8.1
	Total	270	100
Help sought after	1-3 days	158	60.3
	4-6 days	40	15.3
	7-9 days	19	7.3
	10-14 days	14	5.3
	15-28 days	11	4.2
	>28 days	20	7.6
	Total	262	100
Help sought from	General Practitioner (GP) surgery	163	62.2
	Emergency Department	67	25.6
	Other (please state)	32	12.2
	Total	262	100.0
Delay in days after first visit	1-7 days	161	61.7
	8-14 days	31	11.9
	15-28 days	28	10.7
	More than 4 weeks (please state how many weeks approximately)	41	15.7
	Total		
Number of consultations before diagnosis	1	106	44.9
	2	42	17.8
	3	38	16.1
	4	18	7.6
	5	13	5.5
	≥6	5	7.8
	Too many to recall	6	2.5
	Total	236	100.0
Another/other diagnosis	No	166	64.6
	Yes	91	35.4
Place treatment received	Intensive Care Unit (ICU)	40	16.7
	Hospital general ward	36	15.1
	Hospital general ward and ICU	76	31.8
	Hospital general ward, outpatient	16	6.7
	Hospital general ward, Regional Neurological centre	71	29.7
	Total	239	100
Year of diagnosis	Before 2000	49	18.1
	2000-2009	51	19.9
	2010-2019	170	63.0
	Total	270	100.0

Reliability of scales and subscales

The reliability (internal consistency) of the main scales was excellent for *symptoms* ($\alpha > 0.9$), good for *information provided* and for *factors affecting recovery* ($\alpha > 0.8$), and acceptable for *care received* ($\alpha \geq 0.7$). Importantly, none of the scales had poor reliability ($\alpha \leq 0.6$).^[23] Overall, these results presented in Table 3 indicate that the questionnaire was a reliable measure, with good internal consistency.

Further subscales were identified following factor analysis. Symptoms included the following subscales: peripheral nervous system (PNS), central nervous system (CNS), and psychological symptoms. Factors affecting recovery included positive interactions, work support, changes at work, changes at home, physical activity, therapy, and other subscales. Because the subscale, 'other', had a very low reliability, the items constituting it were introduced separately in the regression models. These two items were: immunoglobulin treatment and caring responsibilities. The information provided was divided into two further subscales: provided by specialists including physiotherapists, occupational therapists and by non-specialists including nurses, junior doctors, and general practitioners (GPs). A detailed account of each subscale and the items constituting it can be seen in Table S1 (see Supplementary results). The internal reliability of the new subscales is presented in Table 4.

Table 3 Reliability of each scale and subscale of GBS questionnaire

	Cronbach's alpha	Items	Observations
Scales			
Symptoms	0.94	56	60
Care received	0.70	9	175
Factors affecting recovery	0.80	28	47
Information provided	0.88	11	208
Subscales			
<i>Symptoms</i>			
Initial symptoms	0.89	13	158
Hospital symptoms	0.88	13	156
Residual symptoms	0.86	13	163
Current symptoms	0.89	13	172
<i>Care received</i>			
In hospital	0.65	4	232
After	0.56	4	181

Table 4 Internal consistency of subscales identified following factor analysis

	Cronbach's alpha	Items	Observations
Subscales			
<i>Prompted symptoms</i>			
Peripheral Nervous System	0.85	7	174
Central Nervous System	0.86	3	178
Psychological	0.78	3	180
<i>Hospital symptoms</i>			
Peripheral Nervous System	0.86	7	187
Central Nervous System	0.84	3	181
Psychological	0.75	3	182
<i>Residual symptoms</i>			
Peripheral Nervous System	0.85	7	187
Central Nervous System	0.59	3	179
Psychological	0.79	3	185
<i>Current symptoms</i>			
Peripheral Nervous System	0.91	6	194
Central Nervous System	0.67	3	191
Psychological	0.78	4	185
<i>Factors affecting recovery</i>			
Positive interactions	0.84	6	213
Work support	0.85	4	191
Changes at work	0.65	5	184
Changes at home	0.66	4	202
Physical activity	0.59	3	210
Therapy	0.52	2	209
Other	0.20	3	190
<i>Information provided</i>			
Non-professionals	0.72	4	219
Professionals	0.67	3	223

The reported severity of symptoms for each subscale at different time points (initial before admission to hospital, in hospital, residual and current, i.e. when responders were completing the questionnaire) indicated that symptoms were most severe when responders were in hospital and those affecting the peripheral nervous system (PNS) were most prominent (Figure 1).

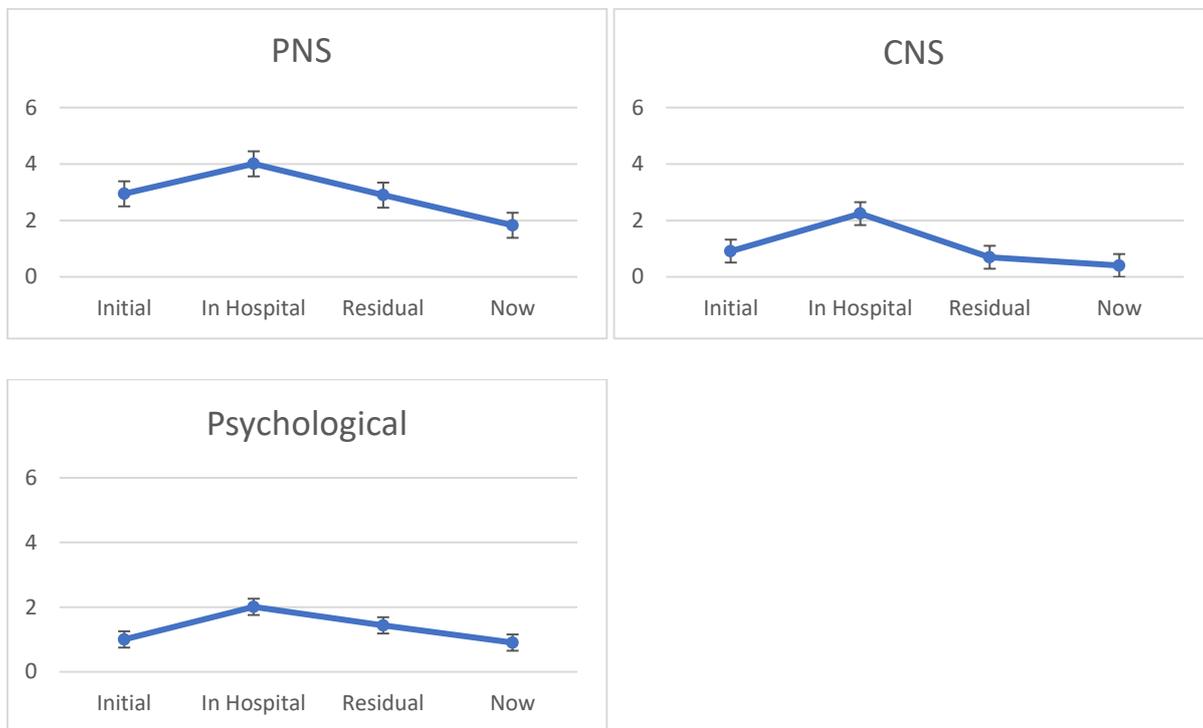


Figure 1 Severity of symptoms for each subscale over time

Responders were more satisfied with the information provided by specialists rather than non-specialists (Figure 2).



Figure 2 Boxplot showing satisfaction with information provided by specialists compared with non-specialists

A combination of physical, psychological and social factors was associated with recovery; these factors were identified following the factor analysis (FA) and an average score was calculated for each factor; details can be seen in supplementary results, Table S3. The factors considered by responders to be most important for recovery were positive social interactions, physical activity including physiotherapy and movement, changes made at home, and immunoglobulin treatment (Figure 3).

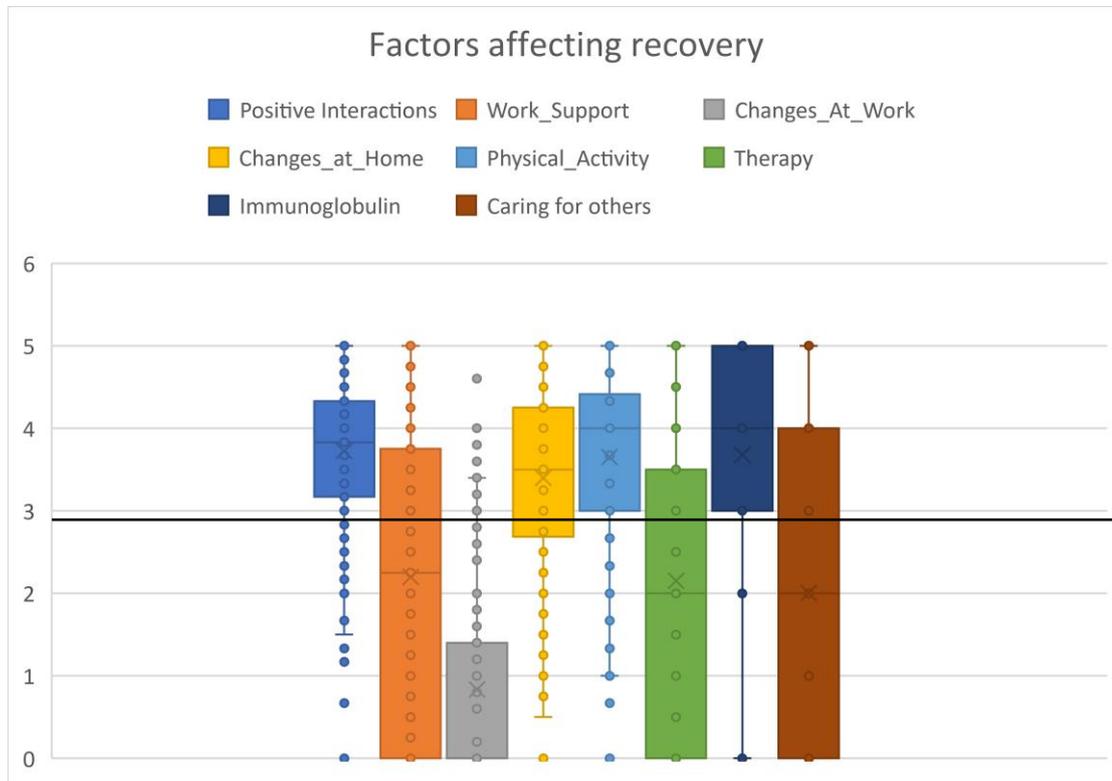


Figure 3 Boxplot of main factors affecting recovery illustrating the distribution of average scores for each factor, three representing the median.

Prediction models

Multivariable regression models were fitted to the data. The predictors used were gender, age, employment status, and living alone or with someone since these variables had been previously shown to influence recovery.[21, 22] In the first regression model, the main scales of the questionnaire (shown in Table 3) were included as predictors. In the second regression model, the main subscales of the questionnaire (shown in Table 4) were included in the model. The results indicated that immunoglobulin and /or plasma exchange treatment was a significant predictor of recovery.

Table 3 Questionnaire scales predicting recovery score

Predictors	Recovery score			
	B	β	95% CI of B	p-value
Gender	-0.33	-0.31	-0.83, 0.20	0.23
Age	-0.29	-0.15	-0.71, 0.14	0.19
Employment	0.50	0.20	-0.07, 1.07	0.09
Household status	0.49	0.16	-0.19, 1.18	0.16
Care received	0.04	0.03	-0.37, 0.44	0.86
Factors affecting recovery	0.36	0.24	-0.03, 0.74	0.07
Information provided	0.22	0.20	-0.10, 0.54	0.17

$R^2 = 0.32, F(7,64) = 4.24, p = 0.001$

Table 4 Questionnaire subscales predicting recovery score

Predictors	Recovery score			
	B	β	95% CI of B	P-value
Gender	0.18	0.09	-0.73, 1.08	0.69
Age	-0.30	-0.19	-0.94, 0.33	0.33
Employment	0.57	0.27	-0.51, 1.65	0.29
Household status	-0.35	-0.12	-1.54, 0.84	0.54
Care received in hospital	-0.18	-0.17	-0.83, 0.47	0.57
Care received after	-0.09	-0.10	-0.48, 0.31	0.66
Information provided by non-specialists	0.09	0.12	-0.26, 0.44	0.59
Information provided by specialists	0.24	0.29	-0.25, 0.73	0.33
Positive social interactions	0.36	0.37	-0.21, 0.92	0.21
Work support	-0.06	-0.11	-0.33, 0.21	0.67
Changes at work	-0.04	-0.06	-0.35, 0.26	0.78
Changes at home	0.04	0.05	-0.42, 0.51	0.85
Physical activity	-0.21	-0.26	-0.73, 0.30	0.40
Therapy	0.10	0.16	-0.17, 0.37	0.44
Immunoglobulin and/or plasmapheresis	0.23	0.38	0.01, 0.45	0.04*
Caring responsibilities	0.01	0.02	-0.21, 0.24	0.91

$R^2=0.55$, $F(16,22)=1.68$, $p=0.13$

* $p<0.05$.

DISCUSSION

Main findings

The high completion rate and low rates of missing data for most questions supported the content and face validity of the questionnaire. The questionnaire showed reliability as excellent for *symptoms*, good for *information provided* and *factors affecting recovery*, and acceptable for *care received* and symptom subscales. Physical, psychological and social factors were associated with recovery and concordance with recent studies [21, 22] supports construct validity. Factors considered by responders to be most important for recovery were positive social interactions, physical activity, changes made at home, and immunoglobulin treatment. Responders were more satisfied with information provided by specialists rather than non-specialists. Multivariable models showed that immunoglobulin and /or plasma exchange treatment were significant predictors of recovery. Being in employment and recovery factors in combination (positive social interactions, support and changes at work support, changes at home, physical activity, and counselling or occupation therapy) were positively associated with recovery but did not reach statistical significance.

Context

Although many people with GBS are told they will recover and some do so completely, many are still affected longer term. Early results from the largest ongoing prospective study, the International GBS Outcome Study [25] (IGOS), have shown that 8% could not walk and 7% had died at one year, with wide international variations in outcome.[26] Previous studies have also shown long term neurological deficits in most patients after a year or beyond. [27, 28] Furthermore, a third had changed work or were affected in function and half had altered their leisure activities.[27] Psychological [29] and social dysfunction [30] often persist longer term, affecting health related quality of life.[19]

Previous research has suggested a wide variation in positive and negative experiences at various stages of treatment and recovery from GBS [21, 22]. We also found wide variations in experience of care from different healthcare professionals during the illness journey, with the most positive experiences of care in hospital, from consultants, followed by nurses and therapists. Consultants followed by physiotherapists were also rated highly for care at follow up, and although in this study physical, psychological and social support were (non-significantly) associated with improvement in symptoms, experience of care and psychosocial support remain important aspects of quality of care.

Rehabilitation studies, involving careful follow-up, show positive benefits of rehabilitation on function [31] and mortality [32] before discharge from hospital, but intensive physiotherapy beyond six months was also

found to improve functional outcomes.[33] Responders in our study valued physiotherapy and perceived this to improve their recovery, but shortfalls in provision for both inpatient and outpatient rehabilitation have been found in previous studies.[34, 35]

Positive social interactions and changes at home were also associated with recovery in this study. Positive social interactions include family or peer support.[22] A systematic review found peer support as a potential intervention for recovery in critical care populations reduced psychological morbidity and improved self-efficacy, although study quality was low.[36]

Strengths and limitations

The number of questionnaires returned was sufficient for the planned analysis, and most participants who began completed the questionnaire. The sample was not intended to be representative of GBS patients as our main aim was to explore the reliability and validity of the questionnaire for measuring responders experience of GBS and its care. The diagnosis of GBS or a variant was based on responder self-identification and we were unable to confirm this from medical records. Some participant characteristics, such as year of and time since diagnosis, time to seek help, number of consultations and delay before diagnosis, place of treatment, length of hospital stay, may have been subject to recall bias.

Implications

The responses to the survey confirmed recent studies suggesting that various physical, psychological and social factors were associated with recovery.[21, 22] Because the survey showed good evidence of face and construct validity and internal consistency, it could be used to assess patient experience and how experience of care and support could be improved in a larger population of people with GBS.

Further research needs to be done to develop Patient Reported Outcome Measures (PROMs) [37] and Patient Reported Experience Measures (PREMs) for GBS beyond traditional disability measures such as the GBS Disability Scale (GDS).[38] The experience scales developed in this survey could be used to develop and evaluate the effect of interventions designed to improve experiences at various stages of treatment and recovery, including in the longer term, including better access to rehabilitation and innovative social interventions, such as peer or employer support.

Conclusion

Our findings showed that the GBS patient experience survey exhibited characteristics of a good measure, with evidence of internal consistency and construct validity. The GBS patient experience questionnaire should be tested more widely to seek further evidence of reliability, construct validity and sensitivity to differences in care and setting.

SUPPLEMENTARY RESULTS

Table S1 Rating of experience and factors affecting recovery

Experience of care or recovery	Missing N (%)	Negative N (%)	Neutral N (%)	Positive N (%)	Total N (%)
How would you rate your experience of care from the following?					
Hospital doctors/consultants	0 (0)	17 (7.1)	29 (12.1)	193 (80.8)	239 (100)
Hospital nursing care	4 (1.7)	18 (7.6)	39 (16.5)	176 (74.3)	237 (100)
Hospital physiotherapy	18 (7.5)	30 (12.6)	25 (10.5)	166 (69.5)	239 (100)
Hospital occupational therapy	48 (20.5)	23 (9.8)	32 (13.7)	131 (56.0)	234 (100)
How would you rate your experience of follow-up treatment following?					
General Practitioner	32 (14.7)	39 (17.9)	46 (21.1)	101 (46.3)	218 (100)
Consultant	18 (8)	26 (11.6)	42 (18.8)	138 (61.6)	224 (100)
Pain clinic	156 (80.4)	13 (6.7)	7 (3.6)	18 (9.3)	194 (100)
Physiotherapy	32 (14.2)	25 (11.1)	33 (14.6)	136 (60.2)	226 (100)
Occupational therapy	91 (43.8)	23 (11.1)	20 (9.6)	74 (35.6)	208 (100)
Which of the following affected your recovery?					
Treatment with Ig and/or plasma exchange	36 (15.5)	6 (2.6)	23 (9.9)	167 (72.0)	232 (100)
Counselling or other psychological treatment	125 (58.7)	17 (8)	25 (11.7)	46 (21.6)	213 (100)
Physiotherapy	25 (10.8)	6 (2.6)	13 (5.6)	187 (81.0)	231 (100)
Occupational therapy	64 (29)	6 (2.7)	23 (10.4)	128 (57.9)	221 (100)
Exercise/activity	15 (6.6)	8 (3.5)	15 (6.6)	188 (83.2)	226 (100)
Self-management	10 (4.5)	3 (1.4)	10 (4.5)	197 (89.5)	220 (100)
Modifications to home	85 (39.5)	6 (2.8)	11 (5.1)	113 (52.6)	215 (100)
Moving from hospital to home	25 (11.3)	18 (8.1)	19 (8.6)	159 (71.9)	221 (100)
Adaptation to the condition	22 (10)	30 (13.6)	23 (10.5)	145 (65.9)	220 (100)
Staying positive about the future	4 (1.7)	19 (8.2)	16 (6.9)	193 (83.2)	232 (100)
Seeing current situation as an opportunity for	27 (12.5)	37 (17.1)	48 (22.2)	104 (48.1)	216 (100)
Responsibilities caring e.g. for spouse, children or	92 (42.4)	28 (12.9)	31 (14.3)	66 (30.4)	217 (100)
Relationships with others	18 (8.1)	39 (17.6)	31 (14)	134 (60.4)	222 (100)
Support from family members	6 (2.6)	11 (4.7)	10 (4.3)	205 (88.4)	232 (100)
Support from friends	6 (2.6)	16 (6.9)	20 (8.7)	189 (81.8)	231 (100)
Engaging in social activities	17 (7.7)	42 (18.9)	31 (14)	132 (59.5)	222 (100)
Others knowing or becoming aware you were ill or	9 (4.1)	34 (15.3)	52 (23.4)	127 (57.2)	222 (100)
Other	55 (68.8)	7 (8.8)	1 (1.3)	17 (21.3)	80 (100)
Which of the following work-related factors affected your recovery?					
Work related benefits	106 (54.1)	10 (5.1)	18 (9.2)	62 (31.6)	196 (100)
Phased return to usual work	96 (49)	9 (4.6)	5 (2.6)	86 (43.9)	196 (100)
Change to part-time	147 (77.8)	2 (1.1)	7 (3.7)	33 (17.5)	189 (100)
Change to job role or duties	137 (71.7)	3 (1.6)	7 (3.7)	44 (23.0)	191 (100)
Change of job	157 (83.1)	3 (1.6)	2 (1.1)	27 (14.3)	189 (100)
Adaptations at work)	146 (77.2)	5 (2.6)	9 (4.8)	29 (15.3)	189 (100)
Support from your employer	84 (43.5)	16 (8.3)	7 (3.6)	86 (44.6)	193 (100)
Support from work colleagues	79 (40.5)	12 (6.2)	10 (5.1)	94 (48.2)	195 (100)
Early retirement	151 (77.4)	3 (1.5)	8 (4.1)	33 (16.9)	195 (100)
Other	99 (92.5)	4 (3.7)	2 (1.9)	2 (1.9)	107 (100)

Table S2 Rating of information provided

Variable	Missing N (%)	Negative N (%)	Neutral N (%)	Positive N (%)	Total N (%)
How would you rate the information provided?					
GP	29	104 (44.8)	39 (16.8)	60 (25.9)	232 (100)
Practice Nurse	81	77 (33.9)	34 (15)	35 (15.4)	227 (100)
Junior Doc	23	65 (28.9)	61 (27.1)	76 (33.8)	225 (100)
-Hospital Nurse)	15 (6.7)	54 (24)	58 (25.8)	98 (43.6)	225 (100)
Consultant	4 (1.7)	22 (9.5)	25 (10.8)	181 (78.0)	232 (100)
Physiotherapist	21 (9.1)	30 (12.9)	26 (11.2)	155 (66.8)	232 (100)
Occupational therapist(s)	58	23 (10.3)	37 (16.5)	106 (47.3)	224 (100)
at the time around diagnosis	0 (0)	66 (28.3)	68 (29.2)	99 (42.5)	233 (100)
during your hospital stay)	0 (0)	50 (21.7)	61 (26.5)	119 (51.7)	230 (100)
at the time discharged from hospital	0 (0)	59 (25.8)	65 (28.4)	105 (45.9)	229 (100)
After you left hospital	0 (0)	76 (33.8)	71 (31.6)	78 (34.7)	225 (100)
Health staff	8 (3.6)	58 (25.9)	67 (29.9)	91 (40.6)	224 (100)
Charities		9 (4)	17 (7.5)	177 (78.0)	227 (100)
Social media	76	7 (3.3)	41 (19.1)	91 (42.3)	215 (100)
Online	77	3 (2)	22 (14.7)	48 (32)	150 (100)
Others	93	1 (0.9)	4 (3.7)	11 (10.1)	109 (100)

Table S3 Items loading on each of the subscales of the questionnaire

Sub-scales	Items
Symptoms	
Peripheral Nervous System symptoms	Reduced mobility Numbness Weakness Paralysis* Tiredness or fatigue Tingling or 'pins and needles' sensations Pain
Central Nervous System symptoms	Difficulty speaking Problems with eyesight or vision*
Psychological symptoms	Difficulty breathing Sleep difficulties Mental health problems (e.g. anxiety, depression, etc.) Difficulty with memory or thinking
Factors affecting recovery	
Positive social interactions	Support from friends Others knowing or becoming aware you were ill or disabled Engaging in social activities Relationships with others Support from family members Staying positive about the future
Work support	Support from your employer Support from your work colleagues Phased return to usual work Work related benefits
Changes at work	Change to job role or duties Change of job Adaptations at work Change to part-time
Changes at home	Modifications to home Moving from hospital to home Adaptation to the condition Self-management
Physical activity	Exercise/ activity Physiotherapy
Therapy	Counselling or other psychological treatment Occupational therapy
Other	Responsibilities caring (e.g. for spouse, children, or an older parent) Treatment with immunoglobulins and/or plasma exchange
Information provided	
Specialists	Physiotherapists Occupational therapists Consultant
Non-specialists	Hospital nurses General practitioners (GPs) Practice nurses Junior hospital doctors

*Paralysis was part of CNS subscale of Present Symptoms; Vision problems were part of Psychological subscale of Present Symptoms.

4. Integration and synthesis

Our study showed that experiences of recovery from GBS vary significantly, many people with the condition still experiencing residual (physical, psychological and social) problems long after their discharge from hospital. This is confirmed in the ongoing International GBS Outcome Study [25] (IGOS), the largest prospective study of the illness with 8% unable to walk and 7% dead at one year.[26] Other studies have also shown residual physical effects including pain, chronic fatigue and, difficulty in walking[2, 5], with long-term neurological deficits, psychological or social impacts at one year or beyond.[27, 28]

Previous studies have shown that older age, severe disability at admission and nadir, ventilator dependence, type of nerve dysfunction and changes in physiological parameters affect long term prognosis.[39, 40] The systematic review pointed to perceptions from people with GBS that recovery varied depending on initial severity, care and support received, and attitudes towards recovery.[21] Interview participants also felt that being younger helped recovery, but they also thought that prior health status and fitness, together with selfcare and lifestyle factors, including a focus on maintaining independence and keeping active, being proactive about maintaining function and adjusting lifestyle to condition, improved the experience of recovery.[22]

The systematic review pointed to wide variations in positive and negative experiences of care and support at various stages of treatment and recovery from GBS.[21] The qualitative study also showed that experiences of care varied widely.[22] Some interviewees experienced positive care practices from healthcare professionals, while others had negative experiences, which they felt adversely affected their progress towards recovery.

Safe and effective inpatient care was reported to improve patients' expectations and strengthen coping strategies or change understanding of their health status and this was perceived to help promote recovery, normal function and perceived better quality of life. The survey suggested that the most positive experiences of care were in hospital, from consultants, followed by nurses and therapists.

Survey responders rated consultants followed by physiotherapists highly for follow up care, and although the survey found that physical, psychological and social support were (non-significantly) associated with improvement in symptoms, experience of care and psychosocial support remain important aspects of quality of care. Rehabilitation was perceived to support recovery post-discharge. Studies involving careful follow-up show positive benefits of rehabilitation on function [31] and mortality [32] before discharge from hospital, but intensive physiotherapy beyond six months has also been found to improve functional outcomes.[33] Longer term rehabilitation post-discharge [33] and exercise[11] help GBS patients to regain

independence by strengthening weakened muscles and enabling them to relearn daily activities.

Interviewees and survey responders valued physiotherapy and perceived this to improve their recovery, but shortfalls in provision for both inpatient and outpatient rehabilitation have been found in previous studies.[34, 35]

Reported psychological symptoms associated with or following GBS include sleep disturbance, anxiety or post-traumatic stress disorder; which can hinder recovery and affect a person's daily life activities, work or social function over many years.[9] A positive attitude to recovery was seen by interviewees as helpful and was consistent with GBS being perceived, at least at the initial stages of the illness, to be a temporary condition with a predictable outcome of recovery. Receiving counselling and being discharged from an inpatient care setting to a home environment were also perceived by interviewees as supporting the recovery experience, as these helped to improve patients' expectations of recovery. In contrast, anxiety, depression or sleep difficulties, at the initial or later stages of the illness, were perceived as hindering recovery.

Social support from family and friends was identified in the systematic review as helping recovery and peer support, enabling people with GBS to see how others with the condition were able to get better and recover successfully, provided positive examples giving them hope for the future.[21] Interviewees in our study also cited social factors as supporting or hindering the recovery experience. Attending faith-based meetings helped some to recover and cope with their illness. Positive social interactions with family or sharing experience of GBS with peers or via social media was said to be helpful, and changes at home also facilitated recovery.[22] Factors perceived to negatively affect experience of recovery included the stigma of being seen in a wheelchair or the adverse effect on marital or other relationships.[22] The illness is known to have a profound psychological impact on family members.[41] A systematic review found peer support as a potential intervention for recovery in critical care populations reduced psychologic morbidity and improved self-efficacy, although study quality was low.[36]

The opportunity to return to a supportive work environment was cited by interview participants as helping to cope and reorder their goals, whereas early retirement due to the illness was perceived to hinder recovery.[22] In previous studies, fewer than half of GBS patients returned to work within two years, with some only managing reduced hours or part-time work due to loss of muscle power, muscle pain, disturbed sensation and fatigue. [42, 43] Unemployment or retirement resulted in financial burdens not alleviated by benefits.[16] Colleagues at work made an important contribution to people with the condition feeling 'normal' again. [22]

The systematic review found that patients struggled with lack of knowledge among healthcare professionals and lack of information they received about GBS, which left them feeling that their needs were not being met.[21] Effective communication is integral to high quality health care, but interviewees in our study expressed concerns about poor communication and information provision.[22] Increasing information provision to the public and professionals about GBS would help to increase understanding of the physical and other residual problems affecting people recovering from GBS and increase knowledge about appropriate support systems for those who need them.

Achieving major milestones also helped participants adjust to and come to terms with their new situation. Being diagnosed with and surviving GBS was a life-changing experience for all participants that often made them search for meaning in their new situation, re-appraise their lives and re-order their priorities. Despite recovering in whole or part, the experience of their illness was profound and persistent.[22]

The survey also supported the findings that physical, psychological and social factors were associated with recovery. Factors considered by responders to be most important for recovery were positive social interactions, physical activity, changes made at home, and immunoglobulin treatment. Responders were more satisfied with information provided by specialists rather than non-specialists. Multivariable models showed that immunoglobulin and /or plasma exchange treatment were significant predictors of recovery. Being in employment and a combination of recovery factors including positive social interactions, support and changes at work support, changes at home, physical activity, and counselling or occupation therapy, were positively associated with recovery (but did not reach statistical significance).

Adverse effects of treatment and lack of longer term follow up care, particularly for patients with persistent residual problems such as pain, chronic fatigue and disability, were perceived to hinder recovery, which points to the importance of providing structured follow up care for GBS patients after hospital discharge and well-designed evaluations of such provision.

Implications

In this study, we have identified the support people need for recovery and the ways that healthcare professionals and services can help facilitate this.

Early detection and treatment of GBS to facilitate recovery and prevent complications is important to patients, but structured follow-up care and rehabilitation, appropriate to the medical and social care needs of individual patients following discharge from hospital, should also be planned and implemented more widely for those with GBS. Rehabilitation has been shown to be effective, but outpatient provision may not

always be available to those who could benefit. [34, 35] Patients' psychological needs were often not met by healthcare services, while maintaining a positive attitude was identified as essential for participants to be able to cope with and successfully recover from GBS. It would be useful, therefore, to add psychological therapies to patients' treatment regimens, if needed and wanted by patients.

An important area that needs to be addressed is the lack of knowledge about GBS among healthcare professionals, and the lack of provision of information to patients and relatives about the condition and its prognosis. More needs to be done to raise awareness among healthcare staff about GBS and provide appropriate training where this is needed. Offering additional training on GBS for healthcare professionals would be an appropriate first step towards improving their knowledge, while providing educational resources and information for the public could be another helpful action.

Charities such as GAIN are currently offering a range of support and advice services, which are valued by patients with GBS, but more could also be done through centres of expertise to provide information and support to both patients, families and staff. Greater knowledge about the condition would benefit GBS patients, their families and informal carers, but signposting to further information and support would also be valuable. Informing patients of available support services (such as financial aid, health and social care services, as well as relevant charities) would further ensure that people receive appropriate and personalised care, facilitate their transition from hospitalisation to returning to their everyday lives and, potentially, aide recovery.

We found that participants also viewed peer support as important in their road to recovery. Peer support could help address both areas discussed earlier, regarding lack of information and emotional support offered to patients with GBS. This could also be an area for future research, exploring how peer support should be provided and that it was clinically beneficial before planning for peer support to be widely available to patients with GBS, potentially through hospitals or GBS charities.

Finally, patients often reported requiring extra support to enable them going back to work. This, according to our results, would include increased awareness of GBS and its sequelae for employers, which in turn would increase understanding of the condition by employers and, therefore, making them more inclined to provide adaptations at work for patients wanting to return to their jobs (e.g. flexible working hours/days, areas for rest, etc.).

The GBS patient experience survey showed evidence of internal consistency and construct validity. The questionnaire should be tested more widely to seek further evidence of reliability, construct validity and sensitivity to differences in care and setting.

REFERENCES

1. Sejvar JJ, Baughman AL, Wise M, Morgan OW: **Population incidence of Guillain-Barre syndrome: a systematic review and meta-analysis.** *Neuroepidemiology* 2011, **36**(2):123-133.
2. Willison HJ, Jacobs BC, van Doorn PA: **Guillain-Barre syndrome.** *Lancet* 2016, **388**(10045):717-727.
3. Van der Meche FG, Van Doorn PA, Meulstee J, Jennekens FG, Centre GB-cgotDNRS: **Diagnostic and classification criteria for the Guillain-Barre syndrome.** *Eur Neurol* 2001, **45**(3):133-139.
4. Webb AJ, Brain SA, Wood R, Rinaldi S, Turner MR: **Seasonal variation in Guillain-Barre syndrome: a systematic review, meta-analysis and Oxfordshire cohort study.** *J Neurol Neurosurg Psychiatry* 2015, **86**(11):1196-1201.
5. Yuki N, Hartung HP: **Guillain-Barre syndrome.** *N Engl J Med* 2012, **366**(24):2294-2304.
6. Hughes RA, Swan AV, van Doorn PA: **Intravenous immunoglobulin for Guillain-Barre syndrome.** *Cochrane Database Syst Rev* 2014(9):CD002063.
7. Hughes RA, Swan AV, Raphael JC, Annane D, van Koningsveld R, van Doorn PA: **Immunotherapy for Guillain-Barre syndrome: a systematic review.** *Brain* 2007, **130**(Pt 9):2245-2257.
8. Hughes RA, Pritchard J, Hadden RD: **Pharmacological treatment other than corticosteroids, intravenous immunoglobulin and plasma exchange for Guillain Barre syndrome.** *Cochrane Database Syst Rev* 2011(3):CD008630.
9. Rajabally YA, Seri S, Cavanna AE: **Neuropsychiatric manifestations in inflammatory neuropathies: A systematic review.** *Muscle Nerve* 2016, **54**(1):1-8.
10. Pena L, Moreno CB, Gutierrez-Alvarez AM: **Pain management in Guillain-Barre syndrome: a systematic review.** *Neurologia* 2015, **30**(7):433-438.
11. Simatos Arsenault N, Vincent PO, Yu BH, Bastien R, Sweeney A: **Influence of exercise on patients with Guillain-Barre Syndrome: a systematic review.** *Physiother Can* 2016, **68**(4):367-376.
12. Vallat JM, Sommer C, Magy L: **Chronic inflammatory demyelinating polyradiculoneuropathy: diagnostic and therapeutic challenges for a treatable condition.** *Lancet Neurol* 2010, **9**(4):402-412.

13. Oaklander AL, Lunn MP, Hughes RA, van Schaik IN, Frost C, Chalk CH: **Treatments for chronic inflammatory demyelinating polyradiculoneuropathy (CIDP): an overview of systematic reviews.** *Cochrane Database Syst Rev* 2017, **1**:CD010369.
14. Forsberg A, Ahlstrom G, Holmqvist LW: **Falling ill with Guillain-Barre syndrome: patients' experiences during the initial phase.** *Scand J Caring Sci* 2008, **22**(2):220-226.
15. Cooke JF, Orb A: **The recovery phase in Guillain-Barre syndrome: moving from dependency to independence.** *Rehabil Nurs* 2003, **28**(4):105-108, 130.
16. Royal E, Reynolds FA, Houlden H: **What are the experiences of adults returning to work following recovery from Guillain-Barre syndrome? An interpretative phenomenological analysis.** *Disabil Rehabil* 2009, **31**(22):1817-1827.
17. Forsberg A, Widen-Holmqvist L, Ahlstrom G: **Balancing everyday life two years after falling ill with Guillain-Barre syndrome: a qualitative study.** *Clin Rehabil* 2015, **29**(6):601-610.
18. Forsberg A, de Pedro-Cuesta J, Widen Holmqvist L: **Use of healthcare, patient satisfaction and burden of care in Guillain-Barre syndrome.** *J Rehabil Med* 2006, **38**(4):230-236.
19. Darweesh SK, Polinder S, Mulder MJ, Baena CP, van Leeuwen N, Franco OH, Jacobs BC, van Doorn PA: **Health-related quality of life in Guillain-Barre syndrome patients: a systematic review.** *J Peripher Nerv Syst* 2014, **19**(1):24-35.
20. Teddlie C, Tashakkori A: **Foundations of mixed methods research : integrating quantitative and qualitative approaches in the social and behavioral sciences.** Los Angeles: SAGE; 2009.
21. Laparidou D, Curtis F, Akanuwe J, Jackson J, Hodgson TL, Siriwardena AN: **Patients' experiences and perceptions of Guillain-Barre syndrome: A systematic review and meta-synthesis of qualitative research.** *PLoS One* 2021, **16**(2):e0245826.
22. Akanuwe JNA, Laparidou D, Curtis F, Jackson J, Hodgson TL, Siriwardena AN: **Exploring the experiences of having Guillain-Barre Syndrome: A qualitative interview study.** *Health Expect* 2020, **23**(5):1338-1349.
23. Cronbach LJ: **Essentials of psychological testing**, 5th ed. edn. New York: HarperCollins; 1990.

24. Field AP: **Discovering statistics using IBM SPSS statistics : and sex and drugs and rock 'n' roll**, 4th ed. edn. London: SAGE; 2013.
25. Jacobs BC, van den Berg B, Verboon C, Chavada G, Cornblath DR, Gorson KC, Harbo T, Hartung HP, Hughes RAC, Kusunoki S *et al*: **International Guillain-Barre Syndrome Outcome Study: protocol of a prospective observational cohort study on clinical and biological predictors of disease course and outcome in Guillain-Barre syndrome**. *J Peripher Nerv Syst* 2017, **22**(2):68-76.
26. Hughes RAC: **Guillain-Barre syndrome: looking back... and forward**. *J Neurol Neurosurg Psychiatry* 2020, **91**(2):111-112.
27. Bernsen RA, de Jager AE, van der Meche FG, Suurmeijer TP: **How Guillain-Barre patients experience their functioning after 1 year**. *Acta Neurol Scand* 2005, **112**(1):51-56.
28. Koeppen S, Kraywinkel K, Wessendorf TE, Ehrenfeld CE, Schurks M, Diener HC, Weimar C: **Long-term outcome of Guillain-Barre syndrome**. *Neurocrit Care* 2006, **5**(3):235-242.
29. Bernsen RA, de Jager AE, Kuijjer W, van der Meche FG, Suurmeijer TP: **Psychosocial dysfunction in the first year after Guillain-Barre syndrome**. *Muscle Nerve* 2010, **41**(4):533-539.
30. Bernsen RA, Jacobs HM, de Jager AE, van der Meche FG: **Residual health status after Guillain-Barre syndrome**. *J Neurol Neurosurg Psychiatry* 1997, **62**(6):637-640.
31. Alexandrescu R, Siegert RJ, Turner-Stokes L: **Functional outcomes and efficiency of rehabilitation in a national cohort of patients with Guillain-Barre syndrome and other inflammatory polyneuropathies**. *PLoS One* 2014, **9**(11):e110532.
32. Inokuchi H, Yasunaga H, Nakahara Y, Horiguchi H, Ogata N, Fujitani J, Matsuda S, Fushimi K, Haga N: **Effect of rehabilitation on mortality of patients with Guillain-Barre Syndrome: a propensity-matched analysis using nationwide database**. *Eur J Phys Rehabil Med* 2014, **50**(4):439-446.
33. Prada V, Massa F, Salerno A, Fregosi D, Beronio A, Serrati C, Mannironi A, Mancardi G, Schenone A, Benedetti L: **Importance of intensive and prolonged rehabilitative treatment on the Guillain-Barre syndrome long-term outcome: a retrospective study**. *Neurol Sci* 2020, **41**(2):321-327.
34. Davidson I, Wilson C, Walton T, Brissenden S: **Physiotherapy and Guillain-Barre syndrome: results of a national survey**. *Physiotherapy* 2009, **95**(3):157-163.

35. Carroll A, McDonnell G, Barnes M: **A review of the management of Guillain-Barre syndrome in a regional neurological rehabilitation unit.** *Int J Rehabil Res* 2003, **26**(4):297-302.
36. Haines KJ, Beesley SJ, Hopkins RO, McPeake J, Quasim T, Ritchie K, Iwashyna TJ: **Peer support in critical care: a systematic review.** *Crit Care Med* 2018, **46**(9):1522-1531.
37. Djordjevic G, Stojanov A, Bozovic I, Berisavac I, Arsenijevic M, Lukic Rajic S, Dominovic Kovacevic A, Jovanovic D, Basta I, Peric S: **Six-month prospective study of quality of life in Guillain-Barre syndrome.** *Acta Neurol Scand* 2020, **141**(3):236-241.
38. van Koningsveld R, Steyerberg EW, Hughes RA, Swan AV, van Doorn PA, Jacobs BC: **A clinical prognostic scoring system for Guillain-Barre syndrome.** *Lancet Neurol* 2007, **6**(7):589-594.
39. Zhang Y, Zhao Y, Wang Y: **Prognostic factors of Guillain-Barre syndrome: a 111-case retrospective review.** *Chin Neurosurg J* 2018, **4**:14.
40. Walgaard C, Lingsma HF, Ruts L, van Doorn PA, Steyerberg EW, Jacobs BC: **Early recognition of poor prognosis in Guillain-Barre syndrome.** *Neurology* 2011, **76**(11):968-975.
41. Bernsen RA, de Jager AE, van der Meche FG, Suurmeijer TP: **The effects of Guillain-Barre syndrome on the close relatives of patients during the first year.** *J Neurol Sci* 2006, **244**(1-2):69-75.
42. Bernsen RA, de Jager AE, Schmitz PI, van der Meche FG: **Long-term impact on work and private life after Guillain-Barre syndrome.** *J Neurol Sci* 2002, **201**(1-2):13-17.
43. Bernsen RA, de Jager AE, Schmitz PI, van der Meche FG: **Residual physical outcome and daily living 3 to 6 years after Guillain-Barre syndrome.** *Neurology* 1999, **53**(2):409-410.