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Critical Review of the Literature (CRL) **7,063 words**

A Systematic Review of the Relationship Between Standardised Assessments of Cognitive Functions and Key Constructs Underpinning the Functional Test in the Mental Capacity Act (2005) in Adults with a Neurodegenerative Condition or Neurological Injury

Service Improvement Project (SIP) **4,995 words**

Understanding Perceived Barriers to the Disclosure of Psychological Difficulties by Young People with a Diagnosis of Coeliac Disease

Main Research Project (MRP) **5,121 words**

Does Severity of Post-Stroke Cognitive Impairment Moderate the Relationship Between Cognitive Change and Emotional Distress in the Months Following Stroke?

Executive Summary **926 words**

Connecting Narrative **976 words**

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Abstracts

CRL

A Systematic Review of the Relationship Between Standardised Assessments of Cognitive Functions and Key Constructs Underpinning the Functional Test in the Mental Capacity Act (2005) in Adults with a Neurodegenerative Condition or Neurological Injury

Objective: To investigate the relationship between standardised assessments of cognitive functioning and the functional test in the 2005 Mental Capacity Act (MCA) in adults with a neurodegenerative condition or neurological injury.

Method: Cochrane Library, MEDLINE, PubMed, CINAHL, PsycINFO, EMBASE, Scopus, and Web of Science databases were searched. Included studies reported outcomes of any cognitive test in adults with a clinically confirmed neurological diagnosis in relation to one or more of the four pillars of the MCA functional test ('understanding', 'retention', 'using and weighing-up information', 'communication'). A narrative synthesis was structured in accordance with each pillar of the functional test.

Results: Of the 3,996 titles screened, 34 studies met inclusion criteria, comprising 40 eligible samples. The most consistent findings related to the pillar of 'understanding', where tests of information processing speed and attention/working memory were strongly correlated to 'understanding' in Alzheimer's Disease (AD) samples and where tests of attention/working memory and executive functioning were strongly correlated to 'understanding' in non-AD samples. No studies explored the relationship between cognitive performance and 'retention' or 'using and weighing-up information'. Eight studies reported associations between performance on cognitive measures and 'communication'. Five studies explored the relationship between cognitive performance and 'overall capacity' in accordance with the functional test and reported inconsistent findings.

Conclusions: The four pillars of the functional test in the MCA are seldom referred to separately. Further research which clearly operationalises the pillars of capacity being assessed is required to elucidate both the utility and limits of cognitive measures in supporting complex functional assessments in line with the MCA.

Keywords: Mental Capacity Assessments; Cognitive Tests; Systematic Review

Systematic review (PROSPERO) registration number: CRD42020168984

SIP

Understanding Perceived Barriers to the Disclosure of Psychological Difficulties by Young People with a Diagnosis of Coeliac Disease

Receiving a diagnosis of coeliac disease as a young person poses developmental challenges and can lead to poor physical and emotional health outcomes. There has been little research in the UK regarding young people's experiences of disclosure of psychological difficulties relating to the condition to healthcare professionals. Seven young people with coeliac disease (aged 11-16 years) were interviewed to explore why young people might feel able or unable to disclose psychological difficulties associated with their condition to clinicians. This study used interpretive phenomenological analysis to generate three superordinate themes from the data: 1) "As soon as food started going away it was like oh *ok* this is what it is": Adjusting to the diagnosis within a developmental context, 2) "It can either be a huge deal or not a deal at all": How others respond to my condition matters to me, and 3) "I mean I wouldn't just speak to the random guy or girl": Who is my doctor to me? Findings were discussed with clinicians in the service alongside recommendations for supporting young people in their adjustment to coeliac disease and disclosure of psychological difficulties to clinicians.

Keywords: Interpretative phenomenological analysis; young people, coeliac disease, disclosure, psychological wellbeing

MRP

Does Severity of Post-Stroke Cognitive Impairment Moderate the Relationship Between Cognitive Change and Emotional Distress in the Months Following Stroke?

This study explores whether the severity of acute post-stroke cognitive impairment moderates the relationship between cognitive change and emotional distress in the first 6-months following stroke. 143 acute stroke patients were assessed within the first 3-weeks (T1) and at 6-months (T2) post stroke. Cognitive impairment was assessed at both time points using the Oxford Cognitive Screen (OCS). At 6-months, awareness of cognitive impairment and emotional distress were assessed using the Cognitive Failures Questionnaire (CFQ) and the Hospital Anxiety and Depression Scale (HADS), respectively. An ANCOVA revealed that awareness of cognitive impairment was significantly lower in the 'moderate-severe cognitive impairment' group ($\geq$1 *SD* from the sample cognitive impairment mean) than the 'mild-no cognitive impairment group' (<1 *SD* from the sample cognitive impairment mean; $p < .001$) at 6-months post stroke. A hierarchical multiple regression analysis indicated that awareness of cognitive impairment and Barthel Index were significant predictors of HADS ($R^2 = .263$; $p < .001$). A subsequent moderation analysis revealed that the effect of cognitive change on HADS was moderated by cognitive impairment at T1 ($R^2 = .391$; $p < .001$). The interaction variable explained 2.2% of variance in the model. Specifically, those with more severe cognitive impairment at T1 reported lower emotional distress as a function of cognitive improvement. Implications of these findings are discussed and recommendations for further research are suggested.

Keywords: Stroke; Depression; Anxiety; Awareness; Cognition

Critical Review of the Literature

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Proposed Journal: This report is intended for submission to *The Clinical Neuropsychologist* and has been written and formatted in accordance with this (see Appendix 1.A). This journal has been selected as it is recognised for both clinically relevant scientific research on brain-behaviour relationships, and professional consideration of matters relating to evidence-based practice. Similar methodologies have been published by this journal.

Abstract

Objective: To investigate the relationship between standardised assessments of cognitive functioning and the functional test in the 2005 Mental Capacity Act (MCA) in adults with a neurodegenerative condition or neurological injury.

Method: Cochrane Library, MEDLINE, PubMed, CINAHL, PsycINFO, EMBASE, Scopus, and Web of Science databases were searched. Included studies reported outcomes of any cognitive test in adults with a clinically confirmed neurological diagnosis in relation to one or more of the four pillars of the MCA functional test ('understanding', 'retention', 'using and weighing-up information', 'communication'). A narrative synthesis was structured in accordance with each pillar of the functional test.

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Keywords: Mental Capacity Assessments; Cognitive Tests; Systematic Review

Systematic review (PROSPERO) registration number: CRD42020168984

Introduction

The Mental Capacity Act[1] (MCA) provides a legislative framework for assessing mental capacity in individuals aged over 16 years in England and Wales. It sets out a two-stage test for assessing capacity. First, there must be an impairment or disturbance of the mind or brain which may impact their ability to make a specific decision[1]. If this stage is met, the assessor should complete a functional test of the individual's ability to: (i) understand information relevant to the decision; (ii) retain the information for long enough to make a decision; (iii) use or weigh-up the information; (iv) communicate a decision using any means[1] (see Appendix 1.B).

The MCA[1] facilitates the protection and empowerment of individuals who lack capacity. However, uncertainties remain around who should conduct such assessments, how assessments should be performed, and what degree of impairment constitutes incapacity[2]. Consequently, capacity assessments have been criticised as largely subjective and inconsistent by patients and carers[3]. This is problematic, as mental capacity decisions directly impact an individual's autonomy over numerous aspects of their lives[2].

It has been suggested that combining standardised interview tools for assessing capacity with cognitive measures may provide a more comprehensive understanding of an individual's ability to make a decision in line with legal standards[2,4]. Current NICE guidelines and British Psychological Society (BPS) guidance suggest that use of cognitive measures that are not primarily designed to assess capacity may yield information relevant to the assessment[5,6].

Cognitive neuropsychological models are increasingly utilised to theorise on the role of specific cognitive domains in the four-component model of 'decisional capacity' set out by Appelbaum and Grisso[7], involving 'understanding', 'appreciation', 'reasoning', and 'expressing a choice'[8,9] (see Appendix 1.C). For example, Palmer and Harmell[8] state that

'reasoning' may draw on working memory and executive functions including abstraction. They argue that by using neurocognitive measures, clinicians can narrow down the reasons why an individual may be experiencing difficulties with decision-making and optimise the individual's role in decision-making by capitalising on their cognitive strengths. However, it remains unclear how performance on cognitive measures is associated with the four pillars of the functional test of mental capacity, as defined by the MCA[1]. This review will aim to address this, through specifically investigating the literature concerning adult participants with neurological injuries or neurodegenerative conditions.

Methods

This review was conducted in accordance with preferred reporting for systematic reviews and meta-analyses guidelines[10], and was registered with PROSPERO (CRD42020168984).

Search Strategy

The following databases were systematically searched in April 2021 using a detailed search strategy: The Cochrane Library, MEDLINE, PubMed, CINAHL, PsycINFO, EMBASE, Scopus, and Web of Science. Search terms related to mental capacity, cognitive assessment, and neurological disease or injury (see Appendix 1.D). Search results were limited to studies in the English language. A hand-search of relevant reference lists was also completed. Titles and abstracts were then screened against the inclusion criteria by the primary researcher. Selected full texts were then screened against the inclusion criteria by the primary and secondary researcher. Inter-rater reliability was calculated using Cohen's kappa statistic to ascertain the scoring agreement between these researchers. A third researcher was consulted if necessary.

Inclusion and Exclusion Criteria

Studies were included if they met the following criteria: peer-reviewed quantitative or mixed method scientific studies; adult participants with a neurological injury or neurodegenerative condition; assessed mental capacity of participants for any decision, in accordance with one or more elements included in the functional test ('understanding'; 'retention'; 'ability to use and weigh-up information'; 'communication') or 'overall' mental capacity, as set out under the MCA[1]; assessing one or more cognitive domains using standardised measures of cognitive functioning; cognitive scores were considered as potential predictors of one or more elements included in the functional test for mental capacity.

Studies were excluded if they involved mixed populations e.g., healthy controls alongside the targeted population where the results are not disaggregated; assessed 'overall' mental capacity of participants using a legal standard that is not in accordance with the functional test set out in the MCA[1], whereby no reasonable conclusion could be drawn relating to any element of the English and Welsh functional test[1]; or did not consider cognitive scores as potential predictors of decisional capacity. Additionally, all case-reports, review papers, theses, and conference abstracts were excluded.

Quality Assessment

Internal validity and bias within studies was assessed using the National Heart, Lung, and Blood Institute Study Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies[11] (NIH; see Appendix 1.E). Study quality was assessed in accordance with the primary aims of the paper and quality rating was carried out independently by two researchers. Inter-rater reliability was calculated using Cohen's kappa statistic to ascertain the scoring agreement ($\kappa = 0.61$).

Data Synthesis

Data extraction was completed with a modified version of the Cochrane Public Health data extraction and assessment template[12] (see Appendix 1.F). Studies reporting the relationship between standardised measures of cognitive functioning and 'overall' decisional capacity in line with the MCA[1], as opposed to reporting findings in relation to each of the four pillars of the functional test, were initially considered in line with cognitive neuropsychological theory and associated models within a section titled 'overall mental capacity'. Where possible, study findings were subsequently grouped in accordance with the four pillars of the functional test set out in section 3 of the MCA[1].

Subsequently, study findings relating to each pillar were synthesised in accordance with established cognitive domains e.g., attention/working memory. Where possible, cognitive measures were categorised by cognitive domain in accordance with original studies. However, due to variability in domain allocation of the same measure across papers, tentative allocations were made based on the majority.

The MCA[1] is a legal framework for England and Wales, however, other countries apply alternative legal standards when assessing mental capacity. Such studies were included if they applied a mental capacity tool akin to at least one of the four pillars of the functional test set out in the MCA[1] e.g. Mac-CAT-T[13]. In these instances, conclusions were drawn regarding aspects of the assessment that are indisputably related to the functional test: 'understanding' and 'expression/communication'. Domain-specific criteria for each mental capacity tool used in included studies is outlined in Appendix 1.G.

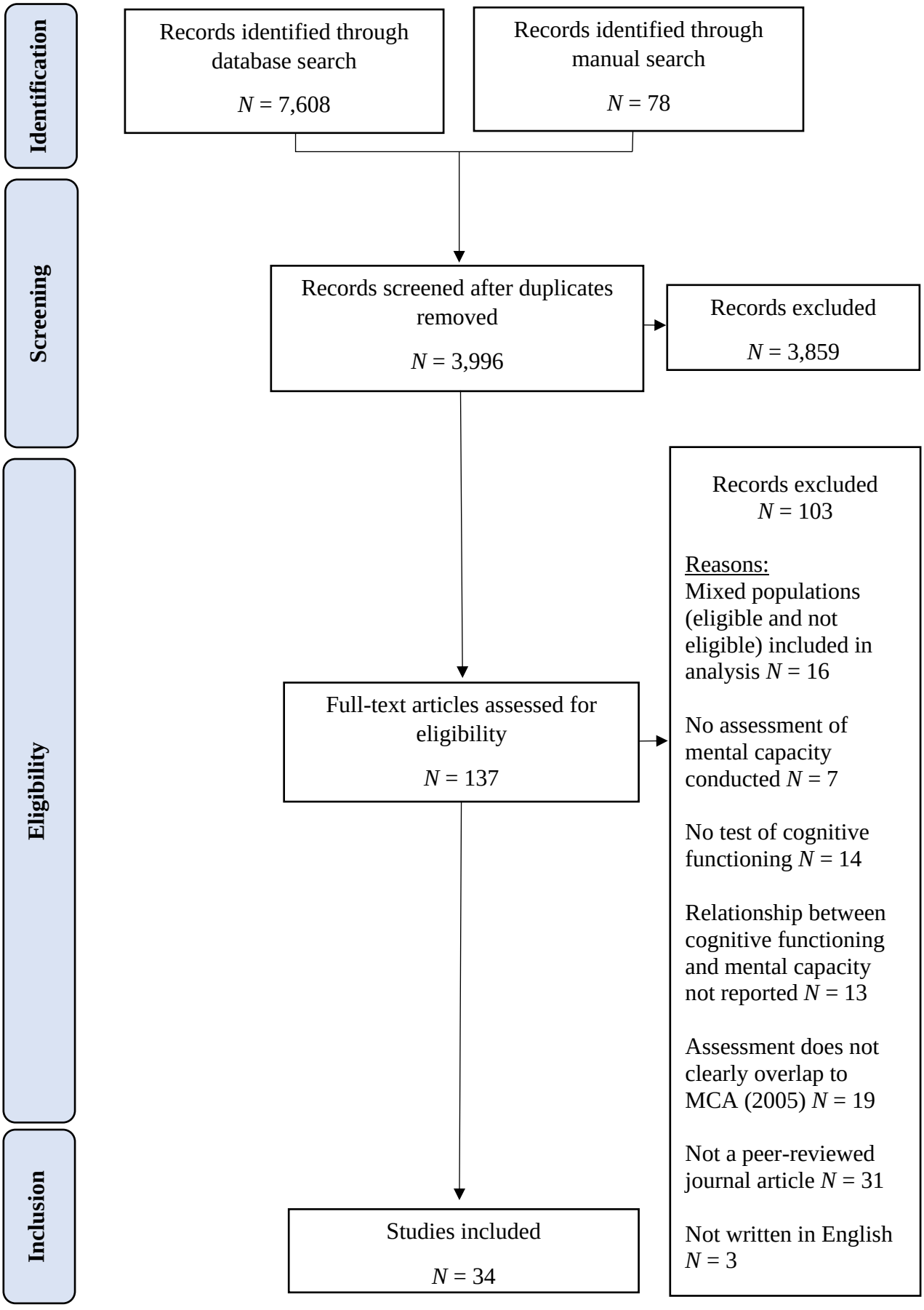
Results

Following the removal of duplicates, 3,996 articles were identified for screening. Titles and abstracts were screened, resulting in 137 articles eligible for full-text screening. Substantial agreement was found between the two researchers ($\kappa = 0.77$). Any

disagreements were discussed and a consensus was reached. Following full-text review, 34 articles were included (see Figure 1).

Figure 1

Flow Chart of Study Screening Against Inclusion and Exclusion Criteria



Study Characteristics

Studies were predominantly cross-sectional, although one study was a single-blind RCT[14] and two studies included longitudinal follow-up[15,16]. Of the 34 studies included, 22 reported analyses of cognitive relationship to capacity as the primary aim[15-36], as opposed to a secondary objective[14,37-47] (see Appendix 1.H). Where studies reported on more than one distinct group of participants or a sample was considered at multiple time-points, results were considered separately[15,28,29,32,42,43]. Consequently, 40 samples were included from 34 studies. Table 1 provides detailed methodological information regarding each study.

Publications spanned 8 countries, although most were carried out in the USA ($n = 20$)[15,17-19,21-24,27,28,30,32,34,36,38-40,45-47], followed by China[25,42-44] and the UK[14,20,26,33] ($n = 4$, respectively). All but three inpatient samples[15,26,32] recruited outpatients or those living in the community. Sample sizes ranged from 13 to 613 participants and included participants with dementia ($n = 13$)[17,20-22,24,25,30,33,35-37,44,45], mild cognitive impairment (MCI; $n = 1$)[23], Parkinson's disease (PD; $n = 2$)[27,38], relapsing-remitting multiple sclerosis (RRMS; $n = 1$)[14], progressive MS ($n = 1$)[40], progressive supranuclear palsy (PSP, $n = 1$)[39], amyotrophic lateral sclerosis (ALS, $n = 1$)[16], stroke ($n = 1$)[26], malignant glioma (MG; $n = 2$)[46,47], brain metastasis (BM; $n = 2$)[19,34], and traumatic or acquired brain injury (TBI/ABI; $n = 2$)[15,29]. Study samples with mixed neurological populations included those with either dementia or MCI[28,31,41-43], dementia or ABI[29], BM or MG[18], or varying degrees of TBI[32]. The findings of these mixed populations were pooled[18,31,41] or reported separately[28,29,32,42,43] within each of these studies.

Twenty-one studies assessed capacity to consent to treatment[14-19,22,25,28,31,32,36-42,44,45,47]. Of these, all but two studies[14,36] used standardised capacity tools. The most frequently used capacity tool was the Capacity to Consent to Treatment Instrument (CCTI)[15,18,19,22,28,32,36,38-40,45,47,48]. Other tools used include the Mac-CAT-T[13,16,22,25,37,41,44], Linguistic Instrument for Medical Decision-Making (LIMD)[31,49], Assessment of Capacity

for Everyday Decision-Making (ACED)[42,50], and Hopkins Competency Assessment Test (HCAT)[17,51]. Nine studies assessed capacity to consent to research[21,23,24,27,30,33-35,46] and six of these used standardised capacity instruments[23,24,27,30,34,46]. The most used tool was the MacArthur Competence Assessment Tool-Research (Mac-CAT-R)[23,24,27,30,52]. Other tools used were the Capacity to Consent to Research Instrument (CCRI)[34,46], and non-standardised tools including: a 17-item Informed Consent Questionnaire (ICQ)[21], an informed consent form[35], and a legally applicable protocol based on the MCA[1,33]. One study[43] assessed financial capacity using the ACED[50]. One study assessed capacity to appoint a lasting power of attorney (PoA)[20], and another assessed capacity to make a discharge decision[26] using a non-standardized, but legally applicable protocol based on the MCA[1]. One study collated findings from a range of capacity decisions using a non-standardised semi-structured capacity interview[29] (see Appendix 1.G for a summary of all capacity tools used).

Many included studies reported correlational analyses exploring the relationship between mental capacity outcome and cognitive performance[14,15,17-19,23,25,27,28,30,34-41,44,46,47]. 'Capacity outcome' was frequently operationalised by scores achieved on each capacity standard assessed by a capacity tool (e.g., 'Understanding'; see Appendix 1.G). Several studies[15,17-19,22,25,28,31,32,34,36,38-40,42,43,45-47] carried out regression analyses to determine whether cognitive performance predicted capacity scores.

Eleven studies categorised capacity dichotomously (e.g. capacity vs no capacity)[16,18-21,24,26,27,29,30,33] and used logistic regression to predict impaired or intact capacity[16,18-21,27,33], ROC analysis to report optimal cut-off scores[18-20,24,29,30], or *t*-tests and non-parametric equivalents to explore whether impaired or intact capacity groups significantly differed in performance on cognitive tests[26]. Two studies also reported predictive equations to indicate incapacity[18,19] (see Appendix 1.H for a summary of analyses and main findings).

Table 1

Study Characteristics

Study	Country	Neurological population	Exclusion criteria	Sample Size; Setting	Average Age (SD); Average Years of Education (SD)	Capacity Decision	Capacity tool used	Cognitive measures used	SQ
Bassett[17]	USA	mild and moderate AD	NR	20; Community	75 (9.87); NR	Treatment	HCAT	Word-list learning (both immediate and delayed recall; CERAD)[53]; WAIS-R digit-span[54]; WAIS-R similarities[54]; TMT[55]	6/13
Bertrand et al.[37]	Brazil	mild and moderate AD	HT, Aph, Alc, Psych, Ep, UMP	71; Outpatient	78 (6.3); 7.5 (3.9)	Treatment	Mac-CAT-T	MMSE[56,57]; ADAS-Cog[58]; WAIS-III digit-span forwards and backwards[59]	8/13
Dreer et al.[15i]	USA	moderate and severe TBI	Psych (not dep), UMP, NeuroHis	34; Inpatient	30 (11.7); 12.2 (2.5)	Treatment	CCTI	GOAT[60]; Token test[61]; WAIS-R digit-span[54]; WAIS-III arithmetic[59]; WAIS-R block design[54]; BVDT[62]; TMT A[55]; SDMT[63]; WMS-R LM 1 and 2[64]; RAVLT[65,66]; 7/24 Spatial Recall Test[67]; EXIT-25[68]; TMT B[55]; WCST[69,70]; Controlled Oral Word Association[62]; category fluency[71]; The Grooved Pegboard[72]	10/14

Dreer et al.[15ii]	USA	moderate and severe TBI	Psych (not dep), UMP, NeuroHis	24; Community	30 (11.7); 12.2 (2.5)	Treatment	CCTI	GOAT[60]; Token test[61]; WAIS-R digit-span[54]; WAIS-III arithmetic[59]; WAIS-R block design[54]; BVDT[62]; TMT A[55]; SDMT[63]; WMS-R LM 1 and 2[64]; RAVLT[65,66]; 7/24 Spatial Recall Test[67]; EXIT-25[68]; TMT B[55]; WCST[69,70]; Controlled Oral Word Association[62]; category fluency[71]; The Grooved Pegboard[72]	10/14
Dymek et al.[38]	USA	Idiopathic PD (mild level cognitive impairment)	NeuroHis, Psych, (not milddep), NeuroleptiHis, NeurosurgeryHis.	20; Community	75 (7.5); 14.3 (3.1)	Treatment	CCTI	WMS-R information and orientation[64]; WMS-R mental control[64]; WMS-R digit-span[64]; BNT[73]; SAC[74]; Token test[61]; DRS[75]; WMS-R LM 1 and 2[64]; EXIT-25[68]; trails A[55]; trails B[55]; COWF[61]; category fluency[76]; WAIS-R similarities[54]; WAIS-R comprehension[54]	7/13
Gerstenecker et al.[19]	USA	BM	Psych, Alc, DAC (not dep)	41; Community	59 (12.4); 13.7 (2.7)	Treatment	CCTI	HVLT-R[77]; WAIS-III digit-span[59]; WAIS-III digit-symbol coding[59]; letter fluency^; COWA[62]; trails A[55]; trails B[55]	8/12

Gerstenecker et al.[40]	USA	Primary and secondary progressive MS	Psych, Alc, DAC (not dep)	22; Community	49 (8.7); 13.6 (2.5)	Treatment	CCTI	WAIS-III digit-span[59]; SRT[78]; WMS-III LM 1 and 2[79]; 7/24 Spatial Recall Test[67]; PASAT[80]; letter fluency[81]; COWA[62]; EXIT-25[68]	9/13
Gerstenecker et al.[18]	USA	BM and MG	Psych (not dep), Alc, DAC, NeuroHis	94; Outpatient	58 (12.1); 14 (2.4)	Treatment	CCTI	HVLT-R[77]; WAIS-III digit-span[59]; WAIS-III digit-symbol coding[59]; letter fluency^; COWA[62]; trails A[55]; trails B[55]	8/13
Gerstenecker et al.[39]	USA	PSP	Psych, Alc, NeuroHis, DAC, Dementia	13; Community	67 (5); 15 (2.6)	Treatment	CCTI	DRS-2[82]; HVLT-R[77]; 10/36 Spatial Recall Test[67]; COWA[62]; oral trails B[83]; Frontal Assessment Battery[84]; oral trails A[83]; oral symbol-digit modalities test[85]	8/13
Gerstenecker et al.[34]	USA	BM	Psych, Alc, NeuroHis, DAC, OMC, Dementia	61; Community	58.8 (11.4); 13.3 (2.6)	Research	CCRI	HVLT-R[77]; WAIS-III digit-span[59]; WAIS-III digit-symbol coding[59]; COWA[86]; trails A[55]; trails B[55]	7/13
Gregory et al.[20]	UK	mild, moderate and severe AD	ESL, Aph	74; Community	80’ (NR); 14’ (NR)	PoA	protocol based on MCA[1]	MMSE[56,57]	7/13
Guarino et al.[21]	USA	mild and moderate AD	NR	613; Community	79 (7); Categorical (highest	Research	ICQ (unstandardized)	MMSE[56,57]; ADAS-Cog[87]	6/12

					high school graduation)				
Gurrera et al.[22]	USA	mild and moderate mixed dementia	Psych	88; Community	75 (6.2); 13.9 (3.1)	Treatment	CCTI, Mac-CAT-T, HCAI (pooled)	WMS-III digit-span forwards and backwards[79]; WMS-III LM 1 and 2[79]; WAIS-III vocabulary[59]; Trails A[88]; Trails B[88]; VSAT[89]; BNT[73]; COWA[62]; mazes test[90]	6/13
Hamann et al.[41]	Germany	amnesic MCI and mild AD	Psych, NeuroHis, DAC	100; Outpatient	72 (8.3); 42% <10 years, 58% >10 years	Treatment	Mac-CAT-T	MMSE[56,57]	7/13
Jefferson et al.[23]	USA	amnesic MCI	Psych, NeuroHis	40; Community	74 (7.5); 15.8 (2.9)	Research	Mac-CAT-CR	BNT[73]; Token test[61]; COWA[62]; WAIS-III similarities[59]; WAIS-III matrix reasoning[59]; trails A[91]; trails B[91]; judgement[92]; CVLT-II (learning; long delay; free recall)[93]	8/13
Kim and Caine[24]	USA	mild and moderate AD	NR	37; Outpatient	79 (5.8); 13.8 (3.2)	Research	Mac-CAT-CR	MMSE[56,57]	5/13
Lui et al.[44]	China	very mild dementia and mild AD	Aph, Psych, NeuroHis, ModDem, SevDem (CDR =2-3), <60	33; Community	78 (5.8); 2.2 (NR)	Treatment	Mac-CAT-T	Cantonese MMSE[94]	8/13

Lui et al.[25]	China	very mild and mild dementia	Aph, Psych, NeuroHis, ModDem, SevDem, <60	50; Community	80 (7); 1.7 (NR)	Treatment	Mac-CAT-T	Cantonese MMSE[94]; Chinese ADAS-Cog[95]; delayed word recall test[96]; category fluency[97]; digit- and visual-spans^^; concept formation^^^	8/13
Lui et al.[42a]	China	mild AD	CDR ≥2, Aph	95; Community	82 (6.6); 1.6 (3.3)	Treatment	ACED	Cantonese MMSE[94]; Chinese ADAS-Cog[95]; delayed word recall test[96]	8/13
Lui et al.[42b]	China	amnesic MCI	CDR ≥2, Aph	99; Community	78 (7); 3.2 (3.5)	Treatment	ACED	Cantonese MMSE[94]; Chinese ADAS-Cog[95]; delayed word recall test[96]	8/13
Lui et al.[43a]	China	mild AD	CDR ≥2, Aph	90; Community	82 (6.6); 1.7 (3.3)	Financial	ACED	Cantonese MMSE[94]; Chinese ADAS-Cog[95]; delayed word recall test[96]; category fluency[97]	8/13
Lui et al.[43b]	China	MCI	CDR ≥2, Aph	92; Community	78 (6.8); 3 (3.2)	Financial	ACED	Cantonese MMSE[94]; Chinese ADAS-Cog[95]; delayed word recall test[96]; category fluency[97]	8/13
Mackenzie et al.[26]	UK	Stroke	None	34; Inpatient	71 (11.47); NR	Discharge	protocol based on MCA[1]	MEAMS remembering pictures, orientation, naming, unusual views[98]; name and address registration and recall[76]; WAIS-III matrix reasoning[59]; WAIS-III digit-	9/12

								span[59]; letter fluency[62]; BADS rule shift[99]	
Marson et al.[36]	USA	mild and moderate AD	NR	29; Outpatient	71 (8.1); 12.5 (2.5)	Treatment	protocol to assess treatment capacity based on legal standards set out by Appelbaum and Grisso[7]	WAIS digit-span[100]; DRS[75]; trails A[55]; trails B[55]; BNT[73]; COWF[61]; category fluency[101]; Token test[102]; auditory comprehension screen[74]; reading comprehension screen[74]; WMS-R LM 1 and 2[64]; WMS-R paired verbal associates 1 and 2[64]; WAIS similarities[100]; WAIS-R comprehension[54]; MMSE[94]	6/13
Marson et al.[45]	USA	mild and moderate AD	NR	72; Outpatient	71 (7.5); 13.5 (2.5)	Treatment	CCTI	DRS[75]; WAIS-R digit-span[54]; Token test[102]; auditory comprehension screen[74]; reading comprehension screen[74]; WMS-R LM 1 and 2[64]; WMS-R verbal paired verbal associates 1 and 2[64]; WAIS-R similarities[54]; WAIS-R comprehension[54]; trails A[55]; trails B[55]	6/13
Marson et al.[46]	USA	MG (Grade 3 and 4)	Psych, Alc, DAC (not dep)	26; Community	53 (12.2); 14.7 (3)	Research	CCRI	HVLT[103]; 7/24 Spatial Recall[67]; WAIS-III digit-span[6]; WAIS-III spatial span[59]; WAIS-III comprehension[59]; trails A[104];	8/13

								trails B[104]; COWA[62]; category fluency[105]; BNT[73]	
Moelter et al.[27]	USA	Idiopathic PD	NR	90; Community	76 (6.2); 15.5 (2.9)	Research	Mac-CAT-CR	DRS-2[82]; MoCA[106]	8/13
Moro et al.[35]	Italy	Very mild, mild, and moderate AD	Und, Beh, HT, Psych, NehroHis	130; Community	77 (5); 7 (3.1)	Research	Unstandardised consent form	Attentional matrix[107]; Raven[108]; Cognitive estimation[109]; verbal judgements[107]; AVLT•; story recall[107]; verbal span[107]; trails A[110]; phonetic fluency[107]; semantic fluency[107]; reversed span[107]; constructive apraxia[109]	6/13
Okonkwo et al.[28a]	USA	mild AD	NR	31; Community	74 (8.59); 13.26 (3.07)	Treatment	CCTI	MMSE[56,57]; DRS-2[82]; WMS-III spatial span[79]; omission and commission errors on the CPT[111]; BNT[73]; category fluency[101]; WMS-III LM 1 and 2[79]; WMS-III visual reproduction[79]; CVLT-II[91]; 10/36 Spatial Recall Test[112]; trails A[55]; trails B[55]; WAIS-III digit-symbol test[59]; CPT hit reaction time[111]; CLOX 1 and 2[113]; cognitive competency test - verbal reasoning subtest[114]; trails 3[55]; WRAT-3 arithmetic[115]	8/13

Okonkwo et al.[28b]	USA	MCI	NR	60; Community	68 (6.77); 14.87 (3.14)	Treatment	CCTI	MMSE[56,57]; DRS-2[82]; WMS-III spatial span[79]; omission and commission errors on the CPT[111]; BNT[73]; category fluency[101]; WMS-III LM 1 and 2[79]; WMS-III visual reproduction[79]; CVLT-II[91]; 10/36 Spatial Recall Test[112]; trails A[55]; trails B[55]; WAIS-III digit-symbol test[59]; CPT hit reaction time[111]; CLOX 1 and 2[113]; cognitive competency test - verbal reasoning subtest[114]; trails 3[55]; WRAT-3 arithmetic[115]	8/13
Pachet et al.[29c]	Canada	Dementia (mixed severity)	ESL, VisHear, Illit	88; Outpatient	73 (13.51); 11.18 (3.61)	Range of Decisions	RCAT Functional Inquires	MMSE[56,57]	6/13
Pachet et al.[29d]	Canada	ABI (mixed severity)	ESL, VisHear, Illit	24; Outpatient	58 (15.81); 12.05 (1.96)	Range of Decisions	RCAT Functional Inquires	MMSE[56,57]	6/13
Palmer et al.[30]	USA	mild and moderate AD	Psych, DAC	30; Outpatient	77 (6.6); 14.6 (2.9)	Research	Mac-CAT-CR	MMSE[56,57]	9/13
Reen et al.[14]	UK	RRMS	MedCh (4 weeks); ConCh (4 weeks);	24; Community	42.58 (8.63); Categorical (highest:	Treatment	Unstandardised protocol (BRIMMS) and	VESPAR series completion[116]; WTAR reading[117]; BICAMS (SDMT, CVLT-II, and BVMTR visual memory)[118]	8/13

			OMC; Psych		postgraduate)		consultation		
Spataro et al.[16]	Italy	ALS	Psych; DAC	102; Community	62’ (54-68); 8’ (5-13)	Treatment	Mac-CAT-T	Phonemic fluency[76]; FrSBe[119]; MMSE[57,120]	11/14
Stormoen et al.[31]	Sweden	AD and MCI	ModDem, SevDem (MMSE <20), OMC, dyslexia, VisHear, Dep (>9 Cornell)	41; Community	AD: 73 (7.6), MCI: 69 (8.8); AD: 10.3 (3.6), MCI: 12.3 (2.9)	Treatment	LIMD	MMSE[56,57]; BNT[73]; word sequence production; sentence repetition; inference; logico-grammatical sentences; reading speed; reading aloud; word fluency; WAIS-R information; WAIS-R similarities; WAIS-R block design; WAIS-R digit-span; WAIS-R digit-symbol coding; corsi block tapping; RAVLT; trails A; trails B; Stroop word task; Stroop colour task[54,57,71,73,121-126]	8/13
Triebel et al.[47]	USA	MG (Grade 3 and 4)	Psych, Alc, DAC (not dep)	26; Community	53 (12.2); 14.7 (3)	Treatment	CCTI	WMS-III spatial span[79]; WMS-III digit-span[79]; HVLT (total and recognition)[77]; WAIS-III comprehension[59]; BNT[73]; category fluency[101]; trails A[55]; trails B[55]; 7/24 Spatial Recall Test[67]	8/13
Triebel et al.[32e]	USA	mild TBI	penetrating TBI, Aph, LD,	49; Inpatient and	40 (15.10); 13.37 (2.40)	Treatment	CCTI	trails A[55]; trails B[55]; WAIS-III digit-symbol coding[59]; WAIS-III symbol search[59];	8/13

			NeuroHis, Alc, Psych	Outpatient (mixed)				WAIS-III arithmetic[59]; WTAR reading[117]; category fluency[81]; CVLT-2 (total recall, short delay free recall, long-delay free recall[91]; WMS-III LM 1 and 2[79]	
Triebel et al.[32f]	USA	moderate and severe TBI	penetrating TBI, Aph, LD, NeuroHis, Alc, Psych	55; Inpatient and Outpatient (mixed)	37 (15.23); 12.58 (2.48)	Treatment	CCTI	trails A[55]; trails B[55]; WAIS-III digit-symbol coding[59]; WAIS-III symbol search[59]; WAIS-III arithmetic[59]; WTAR reading[117]; category fluency[81]; CVLT-2 (total recall, short delay free recall, long-delay free recall[91]; WMS-III LM 1 and 2[79]	7/13
Warner et al.[33]	UK	mild and moderate AD and vascular dementia	NR	176; Community	79 (7.6); 11 (2.7)	Research	protocol based on MCA[1]	MMSE[56,57]; ADAS-Cog[127]	6/13

Note. [i,ii] = same sample tested at two time points (as inpatients then as outpatients) and considered separately; . [a,b,c,d,e,f] = samples disaggregated and considered separately; NR = not reported; ’ = median (inter-quartile range); Exclusion criteria: Alc: history of alcoholism or substance misuse; Aph: aphasia; Beh = behavioural disorder; ConCh (4 weeks) = condition has changed within the last 4 weeks; DAC: disease affecting cognition; Dep: depression (above/below score on scale); Ep: epilepsy; ESL: English second language; Illit: illiterate; LD: diagnosis of a learning disability; MedCh (4 weeks) = medication has changed within the last 4 weeks; ModDem: moderate dementia; NeuroHis: neurodegenerative history; OMC: other medical conditions that affect cognition; Psych: psychotic symptoms; SevDem: severe dementia; UMP: uncontrolled medical problems (e.g. hypertension/diabetes); Und = disorder with verbal comprehension; VisHear: vision or hearing impairment; < n / > n: younger/older than n; Cognitive Measures Used: The consortium to establish a registry for Alzheimer's disease (CERAD) Battery[53]; Wechsler Adult Intelligence Scale-Revised (WAIS-R)[54]; Reitan, Trail Making Test[55]; The Mini Mental State Examination (MMSE)[56,57]; The cognitive subscale of the "Alzhiemer's Disease Assessment scale" (ADAS-Cog)[58]; Wechsler Adult Intelligence Scale 3rd Edn (WAIS-III)[59]; The

Galveston Orientation and Amnesia Test (GOAT)[60]; Token Test from the Multilinguial Aphasia Examination[61]; Controlled Oral Word Fluency from the Multilingual Aphasia Examination (COWF)[61]; Benton Visual Discrimination Test (BVDT)[62]; Controlled Oral Word Association Test from the Multilinguial Aphasia Examination (COWA)[62,86]; Symbol-Digit Modalities Test (SDMT)[63]; Wechsler Memory Scale-Revised (WMS-R)[64]; The Rey Auditory Verbal Learning Test (RAVLT)[65,66]; 7/24 Spatial Recall Test[67]; The Executive Interview (EXIT-25)[68]; Wisconsin Card Sorting Test (WCST)[69,70]; Animal, Vegetable and Clothing Naming (Category Naming)[71]; The Grooved Pegboard[72]; Boston Naming Test (BNT)[73]; Simple Auditory Comprehension Test (SAC)[74]; Dementia Rating Scale (DRS)[75]; Lezak, Animal Fluency (Category Naming)[76]; Lezak, name and address registration and recall[76]; Hopkins Verbal Learning Test-Revised (HVLT-R)[77]; Selective Reminding Test (SRT)[78]; Weschler Memory Scale 3rd Edn (WMS-III)[79] Wechsler Memory Scale 3rd Edn, Logical Memory Tests 1 and 2 (WMS-III; LM 1 and 2)[79]; ^Participants were given 1 minute apiece to name as many words as possible beginning with the letters, “C”, “F”, and “L”; Dementia Rating Scale 2 (DRS-2)[82]; The Paced Auditory Serial Addition Test (PASAT)[80]; Spreen and Strauss, category fluency[81]; Oral Trail Making Test Parts A and B (Oral Trails A and B)[83]; The Frontal Assessment Battery (FAB)[84]; Symbol-Digit Modality Test-Revised (SDMT-R)[85]; The Alzheimer's Disease Assessment Scale (ADAS-Cog)[87]; Army Individual Test Battery, Trails A and Trails B[88]; Visual Search and Attention Test (VSAT)[89]; Mazes Test (Large Print Version)[90]; California Verbal Learning Test (CVLT)[91]; Stern and White, Judgement test[92]; California Verbal Learning Test (CVLT)[93]; Cantonese Mini-Mental State Examination (MMSE)[94]; Chinese Alzheimer’s Disease Assessment Scale (ADAS-Cog)[95]; Unspecified delayed word recall test[96]; Unspecified category fluency[97]; ^^Ascending and descending digit-and visual-spans were used to assess attention and working memory, which are closely related to the integrity of executive function; ^^^Subjects were presented with 3 pairs of objects and asked to explain what each of pair of items had in common. Each comparison was rated at 2 points if it entailed an abstract generalisation, 1 point if a response entailed a specific ‘concrete’ likeness, and 0 points if no similarity was observed. The total score ranged from 0 to 6; The Middlesex Elderly Assessment of Mental State: MEAMS[98]; Behavioural Assessment of the Dysexecutive Syndrome (BADS)[99]; Wechsler Adult Intelligence Scale (WAIS)[100]; Spreen and Straus, Animal Naming (category fluency)[101]; Short form of the Token test[102]; Hopkins Verbal Learning Test (HVLT)[103]; Reitan, Trail Making Test[104]; Gladsjo et al., Category Fluency[105]; The Montreal Cognitive Assessment (MoCA)[106]; Italian standardisation and calibration of neuropsychological tests[107]; Raven’s progressive matrices and vocabulary scales[108]; The Mental Deterioration Battery[109]; Trail Making Test norms[110]; Conners Continuous Performance Test (CCPT)[111]; 10/36 Spatial Recall Test[112]; Executive Clock Drawing Task (CLOX)[113]; Cognitive competency test[114]; The Wide Range Achievement Test 3rd Edn (WRAT-3)[115]; References of tests in Stormoen et al., (2014) are not individually specified and generic references are given[54,57,71,73,121-126]; Verbal and Spatial reasoning task (VESPAR): series completion subtask[116]; Wechsler Test of Adult Reading (WTAR)[117]; The Brief International Cognitive Assessment for Multiple Sclerosis battery (BICAMS) including subtests Symbol-Digit Modalities Tests (SDMT), CVLT-II, and the Brief Visuospatial Memory Tests Revised, recall trials (BVMTR)[118]; The Frontal Systems Behaviour scale (FRsBS)[119]; The Mini-Mental State Examination (MMSE) Italian norms[120]; The Alzheimer's Disease Assessment Scale (ADAS-Cog)[127]; Delis-Kaplan Executive Function System (D-KEFS)[128]; see Appendix 1.I

for details of cognitive assessments. SQ = study quality rating using the NIH Tool[11]. SQ is calculated as Total “Yes” – Total “NA” per paper. This is in line with tool guidance which suggests that “NA” scores should not count negatively towards the quality rating.

'Overall capacity'

'Overall capacity' considers capacity as a unitary concept, as opposed to considering each of the four pillars set out in the functional tests of the MCA[1] independently. Five studies[20,26,29,31,33] explored the relationship between cognitive performance and 'overall capacity' as defined by the MCA[1]. Warner et al[33] found that lower ADAS-Cog score and age were associated with an increased likelihood of 'overall capacity' (*OR* = 0.830; *OR* = 0.916) respectively, but did not find a significant association with MMSE in an AD population. In a further AD sample, Gregory et al[20] reported that increasing MMSE score was associated with an increased likelihood of 'overall capacity' (*OR* = 1.6). These authors reported an optimal MMSE cut-off score of ≤18 for 'overall incapacity'. However, findings from a further study indicate an MMSE cut-off score of ≤22 for 'overall incapacity' in an AD population and a cut-off score of ≤24 in an ABI population[29].

Findings from an acute inpatient stroke population suggest no association between tests assessing a range of cognitive domains and 'overall capacity'[26]. However in a community AD/MCI sample, Stormoen et al[31] found that the WAIS-R 'similarities' and 'information' subtests, along with inference tests, which draw on both executive functioning and receptive language/semantic knowledge, to be the strongest predictors of 'overall capacity' based on the LIMD tool. Their model explained 73% of the variance in 'overall capacity'. Most studies in this review explored the relationship between cognitive measures and distinct capacity standards (e.g., understanding), which will now be explored in more detail.

'Understanding'

Global Measures

Global measures of cognition, such as the MMSE and ADAS-Cog, were commonly used in populations with dementia, although the association between performance and 'understanding' varied (r = 0.28-0.64 MMSE; r = -0.3 - -0.63 ADAS-Cog)[23,25,30,36,37,41,44]. Regression analyses suggest that the MMSE explained small amounts of variance in AD and MCI populations when confounding variables are taken into account (R^2 = 0.27-0.4)[42,43], or 45% variance when a semantic fluency test is also included[25]. A further study suggests that decreasing scores on the MMSE, and increasing scores on the ADAS-Cog were associated with an increased likelihood of impaired 'understanding' (*OR* = 0.87; *OR* = 1.07) in an AD sample[21]. Kim and Caine[24] reported MMSE scores <19 showed high specificity (85%) but low sensitivity (41.2%) for incapacity in 'understanding'. Moreover, the authors found MMSE scores >26 showed high sensitivity (94.1%) but low specificity (30%) for incapacity in 'understanding'. Similarly, Palmer et al[30] reported an optimal MMSE cut-off score of <24 and yielded 61.5% sensitivity and 87% specificity in detecting incapacity in 'understanding'.

Attention/ Working Memory

Multiple studies reported medium-strong positive correlations between 'understanding' and measures of verbal attention and working memory (r = 0.45-0.83)[14,15,17-19,28,35,36,39,40,46,47]. Word learning and recall measures were found to strongly correlate with 'understanding' in populations with MG and BM (r = 0.65-0.83)[18,19,46,47] in all but one study[34]. Tests including the CVLT-2, SRT, a composite memory score derived from an AVLT, story recall, and verbal span also evidenced medium-strong associations with 'understanding' (r = 0.45-0.78) in MCI, AD, MS, and TBI samples[14,15,28,35,40]. Several studies suggest that the WMS-LM-1 measure was strongly associated with 'understanding' in populations including TBI, MCI, AD, and progressive MS (r = 0.55-0.69)[15,28,36,40].

Studies exploring the relationship between digit-span measures and 'understanding' were more variable. Several studies suggested that digit-span measures had a medium-strong association with 'understanding' across TBI, MG, and all but one[34] BM sample (r = 0.45-0.69)[15,18,19,46,47]. However, studies involving participants with dementia diagnoses found small associations between digit-span measures and 'understanding' (r = -0.04-0.27)[17,25,37]. Of these, only the digit-span backwards measure was significant (r = 0.24, $p < 0.05$)[37].

Information Processing Speed

Performance on trails A measures had variable association with 'understanding' across populations and decisions (r = -0.39 - -0.71)[17-19,23,28,36,46]. Two studies found that performance on trails A was predictive of 'understanding' in AD populations (R^2 = 0.5-0.89)[17,28]. Another study[45] reported that an 'executive dysfunction error', whereby an individual experiences a disruption of task awareness and processing, explained 23% variance in 'understanding' in an AD sample and explained 32% variance in trails A. Performance on digit-symbol coding measures had a medium-strong association with 'understanding' in RRMS, BM, and MG samples (r = 0.44-0.70)[14,18,19,34]. Two studies found that performance on combined oral trails A and digit-symbol coding, or digit-symbol coding alone explained 54% variance in 'understanding' when combined with expressive language, attention/working memory, and executive functioning measures in PSP, BM, and MG samples[18,39]. However, five studies did not include trails A in multivariate analyses because it failed to meet the significance level[15,28,34,38,47].

Executive Functioning

Several studies reported a medium-strong association between trails B switching measures and 'understanding' across TBI, AD, MCI, MG, and BM populations (r = -0.42 - -0.79)[15,17-19,23,28,34,46,47] which was supported by subsequent regression analyses[15,18,28,32,34,39]. Reported associations were significant in all but three populations with AD[17,28,36]. Studies

suggested that measures of abstract thinking, such as 'similarities', had a more variable relationship to 'understanding' in dementia, MCI, and PD populations (r = 0.38-0.81)[17,23,25,27,36]. A numerical reasoning measure was strongly associated with 'understanding' in a RRMS population (r = .68)[14]. One study reported that the DRS 'conceptualisation' subdomain explained 70% variance in an AD population[36]. Evidence suggests that executive elements of screening tools such as the EXIT-25, MoCA, and DRS were associated with 'understanding', but with variable effect size (r = 0.29-0.81)[15,27,36,38,40].

Expressive Language

Studies suggested that the expressive language measures most strongly correlated with 'understanding' were those which also assessed semantic language and executive functioning skills, such as category and letter fluency measures. Several studies reported medium-large associations between semantic fluency measures and 'understanding' in AD, TBI, MG, BM, and PSP populations (r = 0.49-0.8)[15,18,19,25,39,46,47]. Four studies found large associations between phonemic fluency measures and 'understanding' (r = 0.48-0.8) in TBI, BM, and MG populations[15,18,19,46]. Increasing scores on a measure of phonemic fluency were associated with an increased likelihood of intact 'understanding' (*OR* = 1.10) in an ALS sample[16]. Further studies found that combined letter and category fluency scores had a medium-large association with 'understanding' (r = 0.47-0.77) in a sample with MCI and progressive MS respectively[23,40]. Subsequent regression analyses suggested that category and letter fluency performance alone predicted between 59% and 71% variance in 'understanding' in samples with progressive MS and MG respectively[40,46]. Two studies reported that a verbal learning/attention measure, such as WMS-LM-1 or HVLT, together with a measure of category fluency predicted between 56% and 72% variance in 'understanding' in TBI and MG populations respectively[15,47]. One study reported performance on both the HVLT and a measure of letter fluency predicted 68% variance in

'understanding' in a sample with BM[19]. Gerstenecker et al[18] extended this by reporting a model which combined performance on phonemic fluency, HVLT, digit-symbol coding, and trails switching, which predicted 54% variance in 'understanding' in a combined sample of individuals with MG and BM. Moro et al[35] reported that combined performance on measures of phonetic fluency, category fluency, trails A, and reversed span had a moderate association with 'understanding' (r = 0.44) in an AD sample. Two further studies including AD, MCI, and PD populations did not include verbal fluency measures in multivariate analyses, because they failed to meet the significance level[28,36,38]. Naming tests, such as the BNT, had variable association with 'understanding' across populations (r = 0.25-0.72)[23,36,46] and explained 11% variance in 'understanding' in a population with mild-moderate AD[36].

Receptive Language

Several studies reported the 'comprehension' subtest from the WAIS to be a good indicator of 'understanding' across MG, PD, and mild-moderate AD populations (r = 0.53-0.72)[36,38,46,47]. The relationship between performance on the less complex comprehension Token test and 'understanding' was more variable. For example, two studies reported large correlations (r = 0.59-0.61) in TBI and AD populations[15,36]. However, two further studies found that this association was not significant in a PD and MCI population respectively[23,38]. A subsequent regression analysis by Dreer et al[15] reported that the Token test, together with the digit-span and trails B switching measures, predicted 71% variance in 'understanding' from baseline to 6 months in a moderate/severe TBI population. This indicates that receptive language measures may be more sensitive in people with more severe or enduring cognitive impairment. The Token test is often used as a measure of executive functioning in AD populations[15]. In line with this, one study found that an executive 'loss of detachment error', whereby an individual presents with an inability to maintain the hypothetical context of a

decision situation, explained 29% variance in 'understanding' and was in part predicted by the Token test (R^2 = 0.43)[45].

Correlational studies suggested that measures assessing reading ability were a poor indicator of 'understanding' (r = -0.1-0.36) in MCI and BM populations[19,23]. However, a proxy pre-morbid IQ measure based on reading ability (WTAR) had a strong association with 'understanding' in a RRMS sample (r = 0.64), perhaps as a measure of cognitive reserve[14].

Long-Term and Overall Memory

WMS-LM-2, a measure of delayed/long-term memory, was strongly associated with 'understanding' and explained 44% variance in an MCI population[28]. Moreover, WMS-LM-2 explained 57.3% variance in 'understanding' when combined with measures of attention/working memory (WMS-LM-1) and expressive language (BNT) in an AD sample[22]. 'Overall memory' was assessed in several studies using a DRS memory subtest[27,28,36,38] and explained between 6% and 68% variance in AD and PD populations, respectively[28,38].

Visuospatial/Constructional Skills

One study reported constructive apraxia had a small, significant association with 'understanding' in an AD sample (r = 0.26)[35]. However, an executive 'non-responsive answers error', whereby an individual provides an answer unrelated to the question presented, explained 15% variance in 'understanding' and was explained by variance in the DRS construction subtest (R^2 = 0.15)[45].

As no studies reported the relationship between cognitive performance and 'retention' or the 'ability to use and weigh-up information' explicitly, the relationship between cognitive performance and 'communication of a decision' (hereon 'communication'; pillar four[1]) will be discussed. Subsequently, how studies addressing 'appreciation' and 'reasoning' may relate to 'weighing-up' (pillar three[1]) will be considered.

'Communication'

Eight studies reported correlations between cognitive measures and 'communication'[25,28,30,36-38,41,44]. Of these, all but one study[38] was in a dementia or mixed MCI/dementia population. Studies reporting the association between performance on measures of global functioning (MMSE or ADAS-Cog) and 'communication' varied (r = 0-0.46 MMSE; r = -0.12 - -0.46 ADAS-Cog)[25,30,37,41,44].

When considering more specific neurocognitive measures, two studies[25,36] reported significant associations between performance on category fluency or the BNT and 'communication', respectively (r = 0.5-0.62). Another study found that an executive 'delegation error', whereby an individual delegates responsibility for decision making to another person, accounted for 33% variance in 'communication' and was associated with variation in the BNT and DRS memory subtest (R^2 = 0.15-0.21, respectively)[45]. This is supported by Dymek et al[38] who reported that the DRS memory subtest accounted for 55% variance in 'communication' in a PD sample and, to a lesser extent Gurrera et al[22], who reported that combining measures of verbal attention/working memory, expressive language, and receptive/semantic language accounted for 10.2% variance in 'communication' in an AD sample.

'Using and Weighing-Up' Information and 'Appreciation'/'Reasoning'

No studies explicitly investigated the relationship between cognitive tests and the ability to 'use and weigh-up' information as most studies adopted the standard of capacity set out by Appelbaum and Grisso[7].

'Appreciation'

The ability to 'appreciate', "requires recognition that one is suffering from a disorder and that the generally accepted risks and benefits of [the decision] apply to one's own

situation"[129]. 'Appreciation' may thus be subsumed under parts of both the first and third pillar of the MCA[1,130] (the ability to 'understand' and 'use/ weigh-up' information). This is supported by some English case law[131].

The association between performance on global measures of cognitive functioning and 'appreciation' was variable (MMSE: r = 0.07-0.67; ADAS-Cog: r = -0.09 - -0.57)[23,25,30,36,37,41-44]. Two studies reported that the MMSE is not a significant predictor of 'appreciation' when controlling for confounding variables[42,43]. Similarly, another study reported that the MMSE was unable to simultaneously show high sensitivity and specificity in predicting 'appreciation'[24].

Findings suggest that performance on cognitive measures relate to 'appreciation' in a similar way to 'understanding'. For example, verbal learning measures appeared to have a moderate-strong association with 'appreciation' (r = 0.35-0.69) across most samples[15,28,34,40,46,47]. Moreover, the digit-span measure appeared to have a weak association with 'appreciation' in samples with dementia[25,37]. Additionally, more complex working-memory measures, such as 'arithmetic' and PASAT, were significantly associated with 'appreciation' in populations with progressive MS and TBI (r = 0.42-0.54)[15,40].

Information processing speed (trails A) and executive switching (trails B) measures were significantly associated with 'appreciation' (r = -0.58 - -0.68; r = -0.41 - -0.65, respectively) in MCI, AD, MG, and BM populations[23,28,34,36,46,47]. Measures involving conceptualisation, judgement, and initiation had small-medium associations with 'appreciation' (r = 0.32-0.54)[23,25,27,28,36].

Category fluency was strongly associated with 'appreciation' (r = 0.6-0.77)[25,46,47], predicting 54%-61% variance in MG samples alone, or combined with the HVLT[46,47]. Phonemic fluency measures were moderately associated with 'appreciation' (r = 0.48-0.49)

in AD and MG populations[36,46]. Performance on verbal recall measures had a varied association with 'appreciation' (r = 0.2-0.71) in AD, MCI, PD, and MG populations[23,25,27,28,47].

Performance on language comprehension measures were most strongly associated with 'appreciation' in AD and MG populations (r = 0.42-0.65)[36,46,47]. Performance on the Token test was moderately associated with 'appreciation' (r = 0.44-0.49) in TBI and AD samples[15,36]. However, it was not significantly associated with 'appreciation' in an MCI population[23]. Performance on the visuospatial, constructional subtest of the DRS-2 had a strong association with 'appreciation' in a population with mild AD (r = 0.53)[28].

'Reasoning'

The ability to 'reason with' relevant information has been argued to have some overlap with engaging in a logical process of weighing options, as both require a judgement of how different information is valued, processed, and related to one another[130]. However, 'reasoning' may overlap with a judgment of 'rationality', which contradicts precedent that 'a person is not to be treated as unable to make a decision merely because he makes an unwise decision'[1,132]. Therefore, it is less clear how 'reasoning' relates to the four pillars of the MCA[1]. However, outcomes of included studies were similar to those relating to 'understanding' and 'appreciation'. Namely, performance on measures of verbal attention/working memory, information processing speed, executive switching, semantic verbal fluency, and delayed verbal memory were associated with 'reasoning'.

Study Quality

No study was excluded from this review of the basis of methodological quality. Study quality, as assessed using the NIH[11] tool can be seen in Table 2. Outcomes were used to assess the internal validity and risk of bias for each study.

Table 2

Study Quality Assessment Table using the NIH Tool[11]

Study	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10	Q11	Q12	Q13	Q14	Overall Quality*
Bassett[17]	•	-	NR	•	-	-	-	•	•	-	•	NR	NA	•	6/13
Bertrand et al.[37]	•	•	NR	•	•	-	-	•	•	-	•	NR	NA	•	8/13
Dreer et al.[15]	•	•	NR	•	-	•	•	•	•	•	•	NR	-	•	10/14
Dymek et al.[38]	•	•	NR	•	-	-	-	•	•	-	•	NR	NA	•	7/13
Gerstenecker et al.[19]	•	•	NR	•	•	-	-	NA	•	-	•	•	NA	•	8/12
Gerstenecker et al.[40]	•	•	NR	•	•	-	-	•	•	-	•	•	NA	•	9/13
Gerstenecker et al.[18]	•	•	NR	•	•	-	-	•	•	-	•	NR	NA	•	8/13
Gerstenecker et al.[39]	•	•	NR	•	•	-	-	•	•	-	•	NR	NA	•	8/13
Gerstenecker et al.[34]	•	•	•	•	-	•	•	•	•	•	•	NR	-	•	7/13
Gregory et al.[20]	•	•	•	•	-	-	-	-	-	-	•	•	NA	•	7/13
Guarino et al.[21]	•	•	NR	•	-	-	-	NA	-	•	•	NR	NA	•	6/12
Gurrera et al.[22]	•	•	-	-	-	-	-	•	•	•	•	NR	NA	-	6/13
Hamann et al.[41]	•	•	•	•	-	-	-	-	•	-	•	NR	NA	•	7/13
Jefferson et al.[23]	•	•	NR	•	-	-	-	•	•	-	•	•	NA	•	8/13

Kim and Caine[24]	•	-	NR	•	-	-	-	•	•	-	•	NR	NA	-	5/13
Lui et al.[44]	•	•	NR	•	-	-	-	•	•	-	•	•	NA	•	8/13
Lui et al.[25]	•	•	NR	•	-	-	-	•	•	-	•	•	NA	•	8/13
Lui et al.[42]	•	•	NR	•	-	-	-	•	•	-	•	•	NA	•	8/13
Lui et al.[43]	•	•	NR	•	-	-	-	•	•	-	•	•	NA	•	8/13
Mackenzie et al.[26]	•	•	•	•	•	-	-	NA	-	•	•	•	NA	•	9/12
Marson et al.[36]	•	-	NR	•	•	-	-	•	•	-	-	NR	NA	•	6/13
Marson et al.[45]	•	•	NR	•	-	-	-	•	•	-	•	NR	NA	-	6/13
Marson et al.[46]	•	•	NR	•	-	-	-	•	•	-	•	•	NA	•	8/13
Moelter et al.[27]	•	•	NR	•	-	-	-	•	•	•	•	•	NA	-	8/13
Moro et al.[35]	•	•	NR	•	-	-	-	•	-	•	•	NR	NA	-	6/13
Okonkwo et al.[28]	•	•	NR	•	•	-	-	•	•	-	•	NR	NA	•	8/13
Pachet et al.[29]	•	•	NR	-	-	-	-	•	•	-	•	NR	NA	•	6/13
Palmer et al.[30]	•	•	NR	•	•	-	-	•	•	-	•	•	NA	•	9/13
Reen et al.[14]	•	•	NR	•	•	-	-	•	•	•	•	-	NA	-	8/13
Spataro et al.[16]	•	•	•	•	-	•	•	•	•	•	•	NR	-	•	11/14
Stormoen et al.[31]	•	•	NR	•	-	-	-	•	•	-	•	•	NA	•	8/13

Triebel et al.[47]	•	•	NR	•	-	-	-	•	•	-	•	•	NA	•	8/13
Triebel et al.[32]	•	•	NR	•	-	-	-	•	•	-	•	•	NA	•	8/13
Warner et al.[33]	•	•	NR	•	-	-	-	•	•	-	-	NR	NA	•	6/13

Note. NA, not applicable; NR, not reported; • denotes "Yes"; - denotes "No"; *'Overall Quality' is calculated as Total "Yes" – Total "NA" per paper. This is in line with tool guidance which suggests that "NA" scores should not count negatively towards the quality rating.

Overall, 24 studies lacked sample size justification and may have been at risk of type-2 error[14-17,20-25,27,29,31-33,35,38,41-47]. Many regression analyses exploring the question set out in this review were not conducted as tests of specific 'a priori' models, but were exploratory in nature, which places further limits on the interpretation of these analyses[14,37-47]. Two studies[22,29] did not disclose participant recruitment methods, therefore sample bias could not be appropriately evaluated. Most studies (N = 31) were cross-sectional in design, which limits conclusions being drawn regarding causal relationships between exposure and outcome variables. Capacity outcome was only assessed once in 76% of studies[17-20,23-25,28-34,36-47], limiting confidence in whether this was correctly classified. Moreover, outcome assessors were only blinded to capacity status of participants in 44% of studies[19,20,23,25-27,30-32,40,42-44,46,47], whereas the remaining studies did not report blinding, which may reflect increased experimenter bias.

All studies had clear research questions. Study inclusion and exclusion criteria were well-defined amongst all but three studies[17,24,36], though quite variable across studies. Participants were selected and recruited from the same or similar populations within all but two studies[22,41]. Both exposure and outcome measures were clearly defined, valid, reliable, and implemented consistently across study participants in most studies[14-19,22-25,27-32,34,37-47]. Key potential confounding variables (e.g., age, severity/duration of diagnosis) were often measured or adjusted statistically for their impact on the relationship between primary exposure and outcome measures[15-21,23,25,26,28-34,36-44,46,47].

Discussion

Associations between cognitive performance and capacity outcomes varied widely between studies. The heterogeneity in samples, capacity measures, and psychometrics included does not allow for clear meta-analysis and thus no confident conclusions can be

drawn regarding the utility of particular cognitive measures in supporting complex assessments of mental capacity. Instead, a broader overview of consistent relationships is provided followed by a discussion of the potential usefulness of neurocognitive measures in supplementing mental capacity assessments.

'Overall capacity'

All five studies which adopted a definition of mental capacity in line with the MCA[1] assessed the relationship between cognitive performance and 'overall mental capacity'[20,26,29,31,33]. Studies exploring the relationship between global measures of cognition, such as the MMSE, and 'overall capacity' suggest they are of limited clinical utility when assessing individuals' capacity in borderline cases and may lead to incorrect assumptions about incapacity.

Two studies used measures of specific cognitive domains and reported opposing findings[26,31]. This may reflect the different populations assessed, or the use of different cognitive and capacity measures. Mackenzie et al.[26] was the only study in this review to find no significant relationship between cognitive performance and capacity outcomes. Notably, in this study all members of the multi-disciplinary team reported low confidence in determining whether a patient had capacity to make a decision, and all unclear cases were not included in their analyses. This study also reported that 50% of acute stroke participants 'failed' tests across multiple domains, suggesting possible floor effects akin to dementia populations[26]. Excluding the most complex capacity cases negates the benefits of including neuropsychological testing in capacity assessments[5,6]. However, given the strengths in sample representation, this is perhaps a true reflection of the limited role of cognitive measures in contributing to capacity assessments in an acute hospital environment. Further research in

clinical samples within the UK, applying assessments in line with the four pillars of the MCA[1] across representative samples is required.

Assessing the relationship between cognitive functioning and 'overall mental capacity' contrasts to the four-pillar model set out in the MCA[1], which implies that an individual may be able to perform differently on one pillar of the functional test than another. Therefore, findings will now be discussed in accordance with each standard of capacity, where possible.

'Understanding'

Measures of global cognitive functioning, such as the MMSE, were primarily used to assess 'understanding' in MCI/AD populations. Findings were variable across studies[17,25,28,30,36,37,41,44] and suggested cut-off scores seldom had concurrent high sensitivity and specificity[24,30]. It is possible that the MMSE is not a consistently good predictor of 'understanding' due to its relative insensitivity in detecting executive function[24].

Studies suggest that tests exploring domains of attention/working memory, and higher-order cognitive abilities, such as executive functioning, information processing speed, and verbal fluency were largely correlated to 'understanding' in >60% of reported associations[14,15,17-19,23,25,27,28,36,38-40,46,47]. However, measures of attention/working memory were often not associated with 'understanding' in studies whereby participants were able to retain and refer to written information during assessment[23,34,46]. Similarly, Reen et al.[14] found that visually presenting information improved MS patient's understanding of treatments when compared to aurally presented information. This suggests that 'understanding' may overlap with the construct of 'retention' in a way that is difficult to separate, as the retained written information may have had the effect of minimising demands on working memory, thereby compensating for this cognitive skill. This is supported by a review of mental capacity

judgments carried out by Keene et al[133], which reported that 'the inability to retain information' was only cited in conjunction with other functional inabilities in dispute cases, as opposed to being cited as the sole functional inability. This finding also emphasises the notable impact that providing information in a mode accessible to the patient (such as written text) can have on increasing their ability to pass this aspect of the functional test. This is consistent with the MCA code of practice, which outlines the importance of taking all possible steps to support the patient in making the decision for themselves.

Findings also suggest emerging patterns of incongruence regarding the association between attention/working memory or executive functioning tests and 'understanding' in dementia samples, compared to other neurological samples. For example, tests such as digit-span and trails B indicated medium-large associations with 'understanding' in most samples, however in dementia samples these associations were often smaller or not reported due to lack of significance. AD has been associated with difficulties understanding, retaining, and organising detailed information regarding a decision[45]. These cognitive impairments may lead to floor effects on memory and executive tests. This poses the risk of minimising the role of attention, working memory, and executive functioning in 'understanding' in this population[25,28,36]. In line with this, several studies suggest that tasks requiring less complex executive functioning skill (e.g., processing speed tasks such as trails A) are strongly associated with understanding, particularly in AD populations, and reflect the importance of internally processing relatively large amounts of information in the process of 'understanding'. A longitudinal study in a TBI population reported that attention and working memory play a key role in 'understanding' initially, but 'understanding' over a 6-month period is primarily mediated by executive functioning and overall frontal lobe function[15]. On one hand, a further study in an ALS sample[16] found that performance in 'understanding' did not significantly change over the course of one year, despite decline in phonemic fluency and

executive function. However, the authors used less comprehensive cognitive measures, with a less cognitively impaired sample which may explain this discrepancy.

Overall, findings suggest global cognitive measures are of limited clinical utility when assessing individuals' 'understanding' which may be due to their insensitivity to executive functioning. However, there are clear associations between domain specific cognitive measures and 'understanding', particularly in studies which perhaps incorporated 'retention' into their definition of 'understanding' by not allowing participants to refer to materials throughout the capacity assessment[134]. In contrast, where participants were able to refer to materials throughout (in accordance with the MCA), the association between tests of attention/working memory and 'understanding' often disappeared. The observed floor effects in AD samples may indicate that domain-specific cognitive tests may have more clinical utility in other neurological samples.

'Communication'

Most studies did not report associations between performance on cognitive measures and 'communication' due to insufficient variability in scores[15,23,28,32,38,40,42,45-47]. Many individuals with cognitive impairment have been shown to communicate a decision equally as well as controls, even if their ability to meet other capacity standards is impaired[22,42]. Of the eight studies which did report associations between cognitive performance and 'communication', seven were carried out in dementia populations. As AD progresses, individuals experience advancing receptive aphasia and characteristic difficulties with processing information which may compromise their ability to communicate[36]. In line with this, studies suggest that performance on expressive language, semantic/receptive language, and working memory measures may be associated with 'communication' in dementia or MCI

populations. However, due to the small number of studies reporting mixed results, it is difficult to draw firm conclusions and further research is required.

'Using and Weighing-Up' Information and 'Appreciation'/ 'Reasoning'

The ability to 'use and weigh-up' information is reported to be at the heart of many legal capacity disputes in England and Wales[133]. Despite this, no studies investigated the relationship between cognitive performance and the ability to 'use and weigh-up information' explicitly. It is possible that the cognitive skills underpinning the construct of 'using and weighing-up' information are difficult to operationalise through cognitive tests. Literature suggests that the process of 'using and weighing-up' information also requires an individual to draw on feedback from their own experiences and different social perspectives[134]. It has been suggested that a more holistic paradigm, which considers emotional and motivational influences of decision making alongside cognitive factors, may provide greater insight into impairments in mental capacity[135]. Specifically, decisions may often be determined on the basis of an often unconscious, somatic signal to the brain representing the practical and emotional consequences of similar past decisions[135]. Worthington[136] suggests that cognitive tests draw on so-called 'cold' executive skills of logical problem solving and are associated with dorsolateral pre-frontal cortex functioning (dlPFC). Despite this, those with ventromedial prefrontal cortex (vmPFC) damage are known to be impulsive[137] and show less sensitivity to the possibility of undesirable outcomes during decision making, which may override intact dlPFC functioning during the 'weighing-up' process[138]. Further research should be carried out to understand the links between social cognition, emotional processing, and decision making, so that Neuropsychologists can gain further insight into impairments in mental capacity in complex cases[136].

Most studies adopted the standard of capacity set out by Appelbaum and Grisso[7]. Some have argued that 'appreciation' may be subsumed under parts of both the first and third pillar of the MCA[1,130] (the ability to 'understand' and 'use' information). However, the ability to 'reason with' relevant information may overlap with a judgment of 'rationality', which contradicts with the MCA[1]. Discussion regarding the legal similarities and differences between capacity standards is beyond the scope of this review, nonetheless findings suggest that performance on cognitive tests relate to the 'appreciation' and 'reasoning' capacity standards in a similar way to 'understanding' which may indicate that they are underpinned by similar, broad, cognitive processes.

Implications

Overall, cognitive performance was often more strongly associated with 'understanding' than for other pillars. On one hand, this suggests that 'understanding' is the most cognitively mediated pillar. However, no studies explicitly explored the relationship between performance on cognitive tests and 'retention' or the ability to 'use and weigh-up information' as set out in the MCA[1]. Moreover, many capacity tools subsumed 'retention' into the measure of 'understanding' and cognitive measures associated with the legal standards 'appreciation' and 'reasoning' were also congruent with those associated with 'understanding'. This suggests that 'understanding', 'retention', 'appreciation', and 'reasoning' may all be sub-served by multiple general domains of cognitive functioning, such as memory and executive functioning[28,34,139].

Findings support the notion that 'understanding' and 'communication' have distinct cognitive predictors, in line with the assumption that they are discrete elements of decision-making capacity[44]. Consequently, studies which adopt a dichotomous approach to capacity may neglect the impact that different cognitive difficulties can have on meeting the

requirements of the functional test. This limits an assessor's insight into how an individual's capacity could be assessed and maximised in more complex and/or borderline cases.

For those without an AD diagnosis, word learning and recall measures (e.g., HVLT, CVLT-2, and the WMS-LM1) combined with verbal fluency measures which draw upon executive functioning and semantic knowledge (e.g., letter and category naming measures) may be particularly useful in assessing 'understanding'. Trails switching measures (e.g., trails B) and working memory/attention measures (e.g., digit-span) may also be useful in supporting an assessment of 'understanding' in non-AD populations. Alternatively, information processing speed measures which utilise executive functioning (e.g., trails A) along with word learning and recall tests (e.g, HVLT, CVLT-2, and WMS-LM1) may contribute to a formulation of 'understanding' capacity in AD populations. Measures associated with 'communication' are less conclusive. However, it appears that measures assessing verbal semantic fluency (e.g., category fluency), along with simple verbal naming tests (e.g., BNT) may provide some insight when assessing individuals with AD. However, it is important to note that several studies excluded individuals with communication difficulties. Thus, future research is needed to identify if cognitive measures may have utility in supporting assessments of capacity in populations with significant communication difficulties. It is important to emphasise that all capacity assessments should be carried out using a range of sources and that clinical judgement also plays a critical role in the contextual and dynamic nature of capacity assessments[140].

Strengths and Limitations

This review involved a comprehensive search of multiple databases spanning over 20 years of studies, and a detailed quality appraisal process. A wide range of cognitive measures were used across included studies. Cognitive measures were categorised by cognitive domain

in accordance with the majority of original studies. However, this may oversimplify the cognitive abilities involved when completing such measures. For example, phonemic fluency measures were categorised as either 'executive functioning' or 'verbal fluency' tasks with approximately equal occurrence, exemplifying that many tests draw on a range of cognitive processes. This makes pinpointing specific cognitive mechanisms associated with standards of capacity complex and, due to the diversity of measures used within each study, it is not possible to draw confident conclusions. Moreover, many studies used different versions of measures e.g., subtests of the WAIS-R or WAIS-III, making the comparison of findings difficult due to psychometric heterogeneity[141]. Additionally, many of the reported measures are no longer used in clinical practice as they have been replaced by more up-to-date versions.

Studies which included similar populations often used different tools to diagnose neurodegenerative conditions or acquired neurological injuries. Additionally, many studies did not distinguish diagnoses subtypes within their samples. Studies also used a range of capacity assessment tools, many of which are reliable and valid, and most studies followed administration instructions. However, some studies adapted the application of capacity tools to reflect how assessments are carried out in clinical practice in an attempt to increase ecological validity[15,23,44,46], but perhaps at the cost of increasing measurement bias.

With regards to publication bias, 13 included studies were carried out by one research group at the University of Alabama. However, these studies were of fair-good quality, with reasonably low levels of bias. The quality assessment chosen, whilst designed for observational studies, may be limited when assessing cross-sectional studies. For example, cross-sectional studies were automatically deemed not to meet criteria for questions regarding follow-up, as these were not applicable (see Appendices E and H). However, this is primarily due to the general lack of available tools to assess the quality of cross-sectional studies.

Conclusions

Existing literature has hypothesised the relationships between the pillars of the functional test in the MCA[1] and associated cognitive functioning at face value[134]. To the author's knowledge, this is the first review empirically exploring the relationship between performance on cognitive measures and the functional test set out in the MCA[1]. Findings indicate the need for further, high powered research exploring the potentially different cognitive profiles underlying incapacity decisions, which clearly operationalises the pillars of capacity being assessed, whilst accounting for confounding variables and social and emotional cognitive factors. This has the potential to inform future, more explicit clinical guidance around the utility of cognitive measures in the context of the functional test set out in the MCA[1].

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Service Improvement Project

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Understanding Perceived Barriers to the Disclosure of Psychological Difficulties by Young People with a Diagnosis of Coeliac Disease

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Proposed Journal: This report is intended for submission to the *British Journal of Health Psychology* and has been written and formatted in accordance with this (see Appendix 2.A). This journal has been selected due to its focus on the emotional and behavioural responses to ill health and the relation of this to health-related behaviours across the lifespan. Research using similar methodology to this paper and relating to both coeliac disease and stigma have been published by this journal.

Abstract

Receiving a diagnosis of coeliac disease as a young person poses developmental challenges and can lead to poor physical and emotional health outcomes. There has been little research in the UK regarding young people's experiences of disclosure of psychological difficulties relating to the condition to healthcare professionals. Seven young people with coeliac disease (aged 11-16 years) were interviewed to explore why young people might feel able or unable to disclose psychological difficulties associated with their condition to clinicians. This study used interpretive phenomenological analysis to generate three superordinate themes from the data: 1) "As soon as food started going away it was like oh *ok* this is what it is": Adjusting to the diagnosis within a developmental context, 2) "It can either be a huge deal or not a deal at all": How others respond to my condition matters to me, and 3) "I mean I wouldn't just speak to the random guy or girl": Who is my doctor to me? Findings were discussed with clinicians in the service alongside recommendations for supporting young people in their adjustment to coeliac disease and disclosure of psychological difficulties to clinicians.

Keywords: Interpretative phenomenological analysis; young people, coeliac disease, disclosure, psychological wellbeing

Introduction

Coeliac disease (CD) is a genetic lifelong autoimmune condition whereby ingesting gluten leads to a range of idiosyncratic symptoms including diarrhoea, abdominal pain, and chronic fatigue (Barratt et al., 2013; Green & Jabri, 2003, 2006). Treatment for CD requires the individual to maintain a strict gluten free diet (GFD), whereby they must exclude foods containing wheat, barley and rye (NICE, 2015).

Research indicates that being diagnosed with CD as a young person, of secondary school age, poses particular challenges when adapting to the new diet and lifestyle (NICE, 2015). Developmentally, this age-range involves increasing autonomy from parents, growing responsibility for one's own wellbeing and food-related activities, and increasing peer-pressure to conform (Ludvigsson et al., 2016). Difficulties adapting to a diagnosis of CD at this developmental stage can lead to non-adherence, poor health-related quality of life, and psychological difficulties (Mayer et al., 1991; Mazzone et al., 2011; Schroeder & Mowen, 2014). A recent systematic review suggests that there is an increased prevalence of anxiety, depression, and somatic complaints in young people with CD, emphasising the importance of identifying young people who may benefit from psychological support (Coburn et al., 2019).

Identification poses a challenge to services, as young people can experience difficulties disclosing psychological difficulties, exacerbated by the process of adjusting to a chronic condition (Rickwood et al., 2007; Rutter, 1995). For instance, research exploring disclosure and support-seeking in young people with heart disease and inflammatory bowel disease (IBD) suggests that only 60% and 62.5% of individuals shared concerns related to their wellbeing during routine consultations, respectively (Ashton et al., 2016; Vatne et al., 2012). Moreover, Vatne et al. (2012) found that young people were more likely to express subtle negative emotional cues during routine consultations, as opposed to clear

verbalisations of distress. A systematic review by Gulliver et al. (2010) suggests that the characteristics of the treating clinician, including credibility, unhelpful responses, and being perceived as ‘too busy’ pose possible perceived barriers to help-seeking in 11-25 year olds.

Identification of young people who are experiencing clinically significant psychological symptoms related to their CD may be further compromised by a lack of integration of psychological support and consultation within multidisciplinary teams (MDT's; Coburn et al., 2020; Crocker et al., 2020). Research in a sample of 800 adults with CD across the UK suggests that emotional well-being was rarely assessed (17.6% cases) by clinicians during follow-up appointments (Crocker et al., 2020). Despite this, a US study reported that 49% of young people attending a paediatric CD clinic displayed clinically significant psychological symptoms (Coburn et al., 2020). Moreover, 68.1% of all families identified mental health consultation as one of their top two priorities when attending medical appointments (Coburn et al., 2020).

Service Context

The stakeholder-service multidisciplinary team (MDT) comprises paediatric gastroenterology Consultants, Junior Doctors, Nurses, a Dietician, and a Clinical Psychologist. This tertiary service diagnoses approximately 30 young people with CD each year and follow-up an additional 75 young people living with the condition. However, only five referrals for young people with CD were made by the gastroenterology team to psychology in 2018. Clinical discussions with gastroenterology Consultants took place during the development of this project. During these discussions, gastroenterology Consultants described a tendency to see CD as a condition which is perhaps ‘easier’ to adjust to and live with than some other gastrointestinal conditions, so do not routinely ask about psychological wellbeing explicitly during consultations. This reflects the current state of services, whereby

psychological consultation has not been routinely integrated nor evaluated in paediatric CD care (Coburn et al., 2020). This raises the question of whether young people with CD perceive that they have had the opportunity to disclose distress associated with CD during routine consultations, or whether they too perceived barriers to disclosure (Gulliver et al., 2010).

We aimed to address these issues systematically by providing a theoretically integrative account of young people's experiences of disclosing or not disclosing psychological difficulties to clinicians in the service. Research objectives were:

1. To understand what it is like for young people to have coeliac disease.
2. To explore young people's experiences of disclosing and not disclosing psychological difficulties associated with CD to clinicians in the paediatric gastroenterology service.
3. To explore young people's perspectives regarding any specific changes that could be made to this service to promote disclosure and improve access to psychological services, if it is felt that this would be helpful for this population.

Subsequently, this study aimed to make recommendations to the service based on findings, as necessary.

Methods

Design and Participants

This study employed an interpretive phenomenological analysis (IPA) methodology in line with its focus on lived experience, with special consideration of the interpersonal and socio-developmental context (Smith et al., 2009). Purposive sampling was used to recruit participants between 11-16 years old who were attending secondary school, to limit sample heterogeneity. Participants were required to have a medical diagnosis of CD, to have been under the care of the stakeholder service for at least 3 months, and have had at least one follow-up appointment. Participants were excluded if they had a co-morbid diagnosed

gastrointestinal condition and/or if they were currently receiving psychological support from any psychological service.

Young people attending CD consultations in the service were screened for eligibility and, if eligible, their parent/guardian was sent a letter by post inviting the young person to participate in an interview. Sixteen-year-olds were sent the invitation letter directly. A further invitation letter was sent three weeks later if they had not responded. Recruitment commenced in March 2020 and demonstrated 25% uptake (28 invited). Interested young people or their parents contacted the primary researcher and were subsequently screened and sent an information sheet. All participants were required to give consent to take part, and parental consent was required for those under the age of 16. All participants completed a demographic questionnaire (see Appendix 2.B for study materials).

Procedure

A semi-structured interview schedule was developed with reference to relevant CD and lifespan research literature, consultation with a young person with CD, and discussions with a Clinical Psychologist (see Appendix 2.C). This was piloted on two potential participants to check for question comprehension and to ensure that the questions elicited in-depth conversations about disclosure of psychological difficulties. Pilot interviews were used in the final analysis as no significant changes were made to the research procedure and interview schedule.

Interviews were either carried out in person at the participants' home, or remotely via video-call software between March 2020 and October 2020. Interviews were audio recorded and lasted between 48 and 85 minutes. Interviews were transcribed verbatim and anonymised. Participants received £5.00 as compensation for their time.

Ethics

This research was approved by the Health Research Authority and Health and Care Research Wales approval Research Ethics Committee, in addition to Oxford University Hospitals Research and Development team (see Appendix 2.D). The study was pre-registered on ClinicalTrials.gov following ethical approval (Identifier: NCT04240340).

Analysis

Analysis comprised five fluid stages, following guidance from Smith et al. (2009; see Table 1 and Appendix 2.E for further details).

Table 1

Stages of Analysis Based on Smith et al. (2009)

Stage	Overview
1	Each interview transcript was repeatedly read and listened to whilst initial, unstructured notes were made denoting powerful recollections of interviews and observations about transcripts.
2	Exploratory descriptive, linguistic, and conceptual codes summarising the experiences described by participants were noted (steps 1 and 2 repeated in an iterative process).
3	Initial notes were reviewed and theme titles that emerged were recorded, considering both specific quotes and overarching narratives for each transcript.
4	Emergent themes were clustered within transcripts to identify cluster labels using methods such as subsumption, abstraction, numeration, polarization, and function. Cluster labels were then combined and reduced into master themes.
5	Patterns in master themes were identified across cases to identify superordinate and subordinate themes.

Credibility Checks

Several validity checks were carried out throughout the research process. Secondary researchers individually reviewed a sample of the transcripts to support the primary researcher in reflecting on the development of themes, and to ensure that they could follow theme development from an idiographic to collective level. Reflexivity was used to identify the influences of personal and professional experiences on the study process (Smith et al., 2009). Peer supervision was employed to facilitate a bracketing interview prior to data collection to elicit preconceptions held by the researcher which may influence the nature of questions asked and the interpretation of data analysis (see Appendix 2.F). A reflective journal was kept throughout the development of the research, and following each interview, to keep a record of personal and professional influences and assumptions (see Appendix 2.G).

Results and Discussion

In line with the flexibility provided by IPA, a decision was made to merge the results and discussion sections by relating superordinate themes to the extant literature (Smith et al., 2009). This enabled the researchers to adequately describe and provide evidence for each superordinate theme within the scope of this paper.

Situating the Sample

The sample consisted of seven white British participants, five females and two males, aged 11-16 (*M* = 14 years, 2 months; *SD* = 1 year, 8 months). All participants had a medical diagnosis of CD, whereby the *M* age of diagnosis was 7 years, 5 months (*SD* = 4 years, 6 months; range 1 year, 6 months – 13 years, 5 months). All participants had at least one family member with CD. One participant had co-morbid ADHD and had previously received psychological input from the service. Remaining participants had no co-morbid diagnoses and were not under the care of other services for their CD. Additional sample characteristics are provided in Table 2.

Table 2

Sample Characteristics

	Pseudonym	Current age (y, m)	Gender	Age when diagnosed (y, m)	Family members with CD
1	Sarah	15y, 4m	F	10y, 0m	Second cousin
2	Maya	14y, 6m	F	9y, 3m	Father; Mother
3	Eve	11y, 10m	F	1y, 6m	Brother
4	Finley	14y, 8m	M	4y, 6m	Sister
5	Sam	14y, 6m	F	3y, 0m	Mother
6	Anna	16y, 4m	F	13y, 5m	Aunt
7	Billy	11y, 11m	M	10y, 9m	Uncle

Note. y = years; m = months; F = female; M = male.

Overview of Themes

Three superordinate themes emerged from the analysis. These were entitled: 1) “As soon as food started going away it was like oh *ok* this is what it is”: Adjusting to the diagnosis within a developmental context, 2) "It can either be a huge deal or not a deal at all": How others respond to my condition matters to me, and 3) "I mean I wouldn’t just speak to the random guy or girl": Who is my doctor to me? These encompassed a range of subordinate themes, as listed in Table 3. Superordinate themes will be discussed in turn using direct quotations to illustrate them (*italics* indicate participant emphasis).

Table 3

Participant Endorsement of Superordinate and Subordinate Themes

Superordinate and Subordinate Themes	Total	Sarah	Maya	Eve	Finley	Sam	Anna	Billy
1. "As soon as food started going away it was like oh *ok* this is what it is": Adjusting to the diagnosis within a developmental context								
1.1 "Over time it just-it feels more normal": Growing with the condition	6		•	•	•	•	•	•
1.2 "Why can't I just have one?...Will it really have an effect on me?": The need for adults to scaffold my understanding over time	7	•	•	•	•	•	•	•
1.3 "My friend group is very good…they always like share what they can like sweets and stuff": the importance of others creating (a safe) space for me	7	•	•	•	•	•	•	•
2. "It can either be a huge deal or not a deal at all": How others respond to my condition matters to me								
2.1 "I don't want to make it serious": The power of labels in mitigating stigma	5	•	•	•		•	•	
2.2 "This has kind of been my life": The problem of others co-creating my identity	5	•	•			•	•	•
2.3 "I didn't want to be different and people to think of me as like this weirdo": Attempts to avoid anticipated stigma	7	•	•	•	•	•	•	•
3. "I mean I wouldn't just speak to the random guy or girl": Who is my doctor to me?	6	•	•		•	•	•	•

Note. • Indicates that the participant endorsed the theme.

1. *"As soon as food started going away it was like oh ok this is what it is": Adjusting to the diagnosis within a developmental context*

This theme encapsulated accounts of participants' journeys in adjusting to their diagnosis. Participants who were diagnosed in early childhood described an easier intrapersonal integration of CD, "It's what I've always known because I was so little I don't- it doesn't really changed how I feel" (Finley, p.9). In contrast, those who were diagnosed from early adolescence described a more complex process of intrapersonal adjustment and acceptance, influenced by feelings of loss, "If I went shopping I'd see like these coco pop bars that I usually love and I'll think oh they used to be my *favourite*" (Billy, p.12). This is in line with existing research which suggests that adjusting to CD compounds the psychosocial challenges associated with the latter stage of development, impacting social relationships with peers and the formation of the young person's identity (Högberg et al., 2003; Olsson et al., 2008; Wagner et al., 2008).

However, the benefits of early diagnosis were challenged by the need for all young people to be provided with developmentally appropriate explanations on an ongoing basis, as they grow with the condition. Participants described a reliance on adults to scaffold their understanding of CD and how to disclose it to others as they age:

> I knew I couldn't have. I couldn't have a certain *piece* of food but… I didn't know what it was so I could say to people that I was gluten free but I couldn't explain to people…what I *actually* had. (Sam, p.9)

As participants developed, they described seeking new information about CD and using this to inform how they managed it e.g., GFD adherence, "I did a whole load of research into it and saw how bad it was and so I was like *I'm not going to do that anymore*" (Sarah, p.8). Provision of developmentally appropriate information relating to the importance of the GFD

and its implications has been evidenced to increase diet adherence in young people (Guandalini & Young, 2014). In particular, young people seek information that is relevant to their daily activities at each stage of their development (Dovey-Pearce et al., 2005).

In line with their growing desire for autonomy, participants described the need for clinicians to adjust their language during appointments so that they could understand and respond to clinicians' questions:

> It's hard for the questions they do ask me. I *understand*, but I don't *get* what they are asking me…when it's passed on to my mum and dad they start the question and they start explaining it and then I'm like, oh I get the question now. (Maya, p.44)

Participants often had limited understanding of their condition, their diet, or the purpose of medical interventions (such as the antibody blood test) because this information had not been updated in accordance with their developmental stage, "there's just blood tests when I go basically...I actually have *no* idea (laughs) like I don't know what they are (laughs)…yeah" (Anna, p.31) and, "[it's] to check how much blood they can get from you without me going pale" (Eve, p.33). This lack of knowledge led to psychological dissonance for some older participants regarding their role in managing their condition. They described feeling both responsible for disclosing and managing their condition, whilst simultaneously feeling unskilled or, in one case, embarrassed about their lack of understanding and ability to describe the condition to others or understand what their medical consultation entailed. As young people developmentally progress through paediatric services, they need to be supported by clinicians to acquire developmentally appropriate skills, knowledge, and motivation to become well informed and committed partners in their healthcare (Dovey-Pearce et al., 2005). Participants who had experienced this reported feeling better able to explain their diagnosis to others, "They told me…my stomach is like an un-blown-up balloon

and then when I eat gluten, the balloon blows up…and that's probably the simplest way, I say the exact same thing every time" (Maya, p.17).

The importance of having an informed understanding of the condition was reinforced by the need for young people to build up a peer-based network of 'supporters', who helped them navigate social situations involving food, playing a key role in their acceptance and adjustment to the condition, "If I'm going to a sleep-over I'm not just there with like a bowl of milk or something...it makes me feel quite good" (Finley, p.21-22), and "I feel like having that person which you just really talk about it…to have conversations like "how's your gluten free" y'know "what's going on" and erm about everything else, more of like (pause) yeah. A supporter" (Sarah, p.23). Absence of this understanding led to difficulties correcting others' misunderstandings, which created difficulties in cultivating support. For example, by leading to peers diminishing the importance of the GFD, "They're like "oh can't you try just a little bit just to see what it tastes like" and it's always. It's *no*. because it's not…It's not that simple" (Sam, p.19). Evidence suggests young people who are able to share appropriate knowledge about CD and the GFD with their peers experience reduced stigma around the condition and, in particular, the GFD (Olsson et al., 2009). Hosting group spaces for young people with CD to share information and coping strategies with each other is also associated with better psychosocial adjustment and reduced feelings of alienation and perceived stigmatization (Shani et al., 2020).

2. *"It can either be a huge deal or not a deal at all": How others respond to my condition matters to me*

This theme also stems from a consideration of developmental context, in that it captures the importance young people place on how others, particularly peers, respond to their condition. When it comes to disclosing the condition, participants hoped to strike a

balance between others taking it seriously and providing safe foods, without drawing unwanted attention from others, which they perceived as highlighting difference:

> Cus I mean you can be like at two ends of the scale like sometimes when people completely overreact like, "OH MY GOD SOMEONE'S GLUTEN FREE LETS ALL DO THIS BIG THING FOR THEM" and then the other side there's like I'm in *this situation* and there's *nothing* gluten free and *no one* cares at all. (Sarah, p.10)

In striking this balance, participants described facing a conundrum with regards to the label they used to disclose their condition. The label 'disease' often drew unwanted attention and could lead to perceived or actual stigma from peers, "If I say coeliac disease they're like "OHMYGOD you've got a DISEASE" (Sarah, p.21) and, "coeliac disease sounds a lot worse than gluten free...because it's *disease*…It makes it sound as though like, you're gunna *die* from it when really you're not" (Eve, p.27). Participants' choice of label could result in others misunderstanding the condition and co-creating the their identity in a way that felt problematic, "A few people responded in like "*oh that's so rubbish*" or "that's *terrible*"...then I have in my mind always, you know in the back of my mind that it is gunna be *rubbish*" (Billy, p.4), and:

> If my friends say "Oh Anna is gluten free" I (laughs) *often* correct them like no I'm actually *coeliac* because it's like not a *choice* and I feel like places like restaurants or pubs they kind of don't *get* that that much. (Anna, p.7)

This challenge speaks to their developmental context, whereby young people are both yearning to carve out their personal identity whilst also consolidating their social identity based on their membership of certain social groups (Arnone & Fitzsimons, 2012). This poses the challenge of trying to navigate the complexities of disclosure to avoid taking on an unwanted identity and the perceived or actual stigma resulting from real or imagined differences (Erikson, 1950; Schroeder & Mowen, 2014).

Participants often anticipated stigma through disclosure to peers, in line with the developmental need to 'fit in' and avoid difference, "I just felt like I was just like the odd one out like the *one* friend who couldn't have that specific thing" (Sam, p.36), and "obviously in primary school everyone wants to be the same…So it's like are they gunna think I'm *weird*" (Maya, p.21). This was sometimes based on experiences of others' stigmatising reactions to disclosure in situations involving food, "He still finds it *shocking*. That I can't eat KFC" (Sam, p.12) and "they'll be like oh Maya with her gluten free, she can't eat anything" (Maya, p.32). CD and the GFD are not always well understood by others, suggesting young people with CD may be more likely to internalise others' views about their condition being undesirable, abnormal, and something that should be concealed through avoidance (Copelton & Valle, 2009; Schroeder & Mowen, 2014), a coping strategy utilised by some young people in this study, "I didn't tell my friends for ages and I can *remember* that. Like not telling them" (Sarah, p.11) and, "on the rare occasions when I have been asked to go out to places to eat I just say no…I'm like very *careful* about it" (Anna, p.5).

Alternative coping strategies included reframing the condition as something that others were missing out on, "when I was younger, like when people asked me why I couldn't eat things I'd say because I'm *special*" (Eve, p.9) and, "I sort of say quite a lot of gluten free food is *better* than food with gluten in it" (Billy, p.17). By reframing the condition and the GFD as a strength or assert, it reduces the likelihood that others will perceive these as weaknesses (Kaushansky et al., 2017). Alternative strategies used to incorporate CD into their identity in a stigma-reducing way included educating their friends and family about the condition to 'bring others onto their side' and curate self-acceptance, "They're like oh that's fine and, accepting that I can't eat it and, it's not personal" (Maya, p.4), and:

> They may have heard the term gluten-free but I don't think they've ever like *asked* like what it *actually* was. So they just never really *learn*…they're just like "oh *what's* that?" and then they *understand* and then they take it seriously. (Sam, p.21)

This is similar to the 'neutralising motive' set out by Sharp (2009), whereby disclosure of and education about a condition was found to minimise the effects of stigma. This approach has been evidenced to help young people maintain a positive social identity and can result in greater understanding and solidarity in their social groups (Schroeder & Mowen, 2014).

3. "I mean I wouldn't just speak to the random guy or girl": Who is my doctor to me?

This theme encapsulates the barriers that young people identified to disclosing psychological difficulties to clinicians. Identified barriers were in line with previous research, suggesting that they are not specific to young people with CD (Beresford & Sloper, 2003; Gulliver et al., 2010; Radez et al., 2020). Some young people held stereotypical beliefs about doctors as serious and strict, "They may think that if they tell them like they may be angry about it because they don't have that like gentle face, they have a very serious face" (Sam, p.34). Some ascribed doctors with a high status and positioned them as 'knowing everything'. This contrasted with a sense of their own inferiority, which could generate feelings of anxiety:

> I definitely myself get nervous…if I'm talking to doctors…it's very intimidating when someone's sitting there and asking you is *this* wrong with you...you're sitting there talking to an adult who knows everything, and you don't know anything yourself. That's the scariest part of it. (Maya, p.43-52)

Many held beliefs about what doctors are and are not interested in hearing about and this often led them to anticipate that medical consultations would not be an appropriate place to disclose emotional difficulties:

> These people are here to help me for medical reasons… if someone was struggling I don't think they'd even *think* about a doctor like being an outlet to explain their emotional like problems with it. (Anna, p.44)

There is evidence that young people who have established and trusted relationships with clinicians are more likely to seek help in the future (Gulliver et al., 2010; Rickwood et al., 2007). Thus, there is a need for clinicians to actively break down barriers. For example, by adapting their body language to communicate safety and time:

> They don't just ask you a question and then do something else but they'll pay attention to it…they usually like turn around on their chair and push out from their computer and that just shows that they're not gunna be doing anything on the computer. (Billy, p.27)

Young people were often aware of the stress on the wider hospital system and either felt that the way they were feeling was not important enough to take up the clinician's time or that they would be seen by the clinician as a timewaster, "I didn't want people to think I'm – not weak, but I didn't want people to think I was like *struggling* with something which is supposedly y'know outlook and seen as so just *easy*" (Sarah, p.36) and, "They're like doctors and they just wanna like- they've got a lot of people to see they just wanna do the session and move on" (Anna, p.36).

In line with existing research, few participants recalled being explicitly asked about their emotional adjustment during consultations and suggested that it would be helpful for clinicians to provide clear permission to disclose psychological difficulties, so that they could recognise and respond to this accordingly (Crocker et al., 2020; Radez et al., 2020):

> I feel like when the doctors are *speaking* maybe there should be like an extra question like "how are you feeling?"...every time I think they say "how are you going with it?" I always assume they're asking me like how am I finding alternatives…I don't

> actually think they're asking me like *emotionally* like "how do you feel?". (Anna, p.42)

Young people reported an assumption that if they were not explicitly asked about their emotional wellbeing, then this must be because they should not be experiencing difficult feelings, clinicians are not interested in this, or that their appointment is not the place to disclose this, "I haven't really spoken about how I feel about being gluten free...I don't really feel it's necessary...I mean it's not something they really *need* to know" (Finley, p.27-28), and:

> [the doctor] has always just sort of been like so "how's your stomach feeling?"…it's never been like "how are *you* feeling?" so like I didn't feel like it was the right *place* for me to talk about my feelings to them. Um or that I *should* be feeling those feelings about something so (pause) *small*. (Sarah, p.37)

Existing literature suggests that young people hold beliefs that they should be able to manage their own emotional difficulties, and may perceive help-seeking as a sign of weakness (Radez et al., 2020; Rickwood et al., 2007). Thus, permission-giving is equally important when young people are doing well with adhering to the diet, to normalise that they may still experience difficult emotions relating to the condition and that this is not a reflection of them not managing well and can be discussed.

Recommendations and Service Feedback

The primary researcher met with the Psychologist in the service to discuss findings and subsequently met with two gastrointestinal Consultants and one Registrar in March 2021 to present the findings and discuss how recommendations may be implemented (see Table 4). A lay summary of results has been sent to the participants who opted to receive this (see Appendix 2.H).

Table 4

Summary of Recommendations, Rationale, and Clinician Responses

Recommendations and rationale relating to the provision of developmentally appropriate information	
Recommendation	Rationale
Provide information in a leaflet that goes out to patients ahead of appointments explaining what the appointment (and blood test) is for in accordance with their developmental stage. During the appointment it can be helpful to normalise forgetting e.g. "Sometimes patients say that they can't remember what the blood test results mean and it can be helpful for me to explain this again – would you find this helpful too?"	Young people described the need for information to be updated and repeated at each appointment in accordance with their developmental stage. A proactive and sensitive approach to providing updated information during appointments is required as young people can feel embarrassed to ask for this information themselves.
Check that young people have the language to explain the condition to their peers and adults.	Young people described how the lack of an age-appropriate understanding of their condition can lead to avoidance of disclosure to peers and adults and, occasionally, non-adherence to the diet. Having the words to talk about the condition may mitigate this risk.
The paediatric diabetes service in the Trust run education classes – where young people and their parents/ a peer come along to discuss the challenges that could come up at different developmental stages. These could be run for young people with CD too.	A young person suggested developing an educational video, or resources for teaching about the condition in schools as a way of making it easier to talk about how the condition makes them feel. By facilitating education classes, the service can equip young people with the knowledge and language to share this information with their peers.
Clinician Responses	
Clinicians questioned why information about the diagnosis and diet already provided at the point of diagnosis, along with signposting to charities such as Coeliac UK, is not sufficient. Through discussion, they recognised that findings suggest that young people need this information to be updated and repeated over time. They described feeling constrained by the limited duration of consultations to provide updated information themselves. As young people do not routinely see other members of the MDT, this provides context for why they may not have sufficient understanding and language of CD and the GFD. This also validates young people's perceptions that they should understand their condition without this information being repeated over time. Clinicians felt that the development of a leaflet to send to	

young people ahead of their appointments would be the most efficient way of providing information. The MDT Psychologist is motivated to continue discussions with the MDT to find a way to develop age-appropriate leaflets and integrate information into appointments themselves. The MDT Psychologist is also motivated to develop education days to take place at different developmental stages e.g., for young people in year 5/6 ahead of transitioning to secondary school, in collaboration with the wider MDT.

Recommendations and rationale regarding how to actively break down perceived barriers to disclosure	
Asking an extra question about how young people are feeling emotionally can be helpful in opening-up a discussion e.g. "Sometimes young people tell me that they worry about eating out with their friends, is this something you worry about sometimes too?" This could be followed-up by use of a screening tool if mental health difficulties are indicated.	Young people reported an assumption that if they were not asked about their emotional wellbeing, then this must be because it should not be spoken about. Thus, even when young people are doing well with adhering to the diet, it is helpful to normalise that they may still experience difficult emotions relating to the condition and that this is still important and can be talked about.
It is necessary for clinicians to take active steps to break down stereotypes to reduce the power-dynamic and create safety in appointments. For example, by putting up an 'emotion poster' or something similar in the consultation room, whereby the clinician could ask the young person which picture represents the way they are feeling that day and share the emotion that they are feeling too. It can also be helpful to reflect on how body language may communicate how interested you are, how much time you have, and how empathetic or safe you are.	Young people commonly disclosed experiencing a strong power-dynamic in their appointments. Showing the young person that the clinician has space and time for discussion, understanding, and empathy of emotions could help redress this power-balance and break down stereotypes. Young people commonly described the importance of the clinicians' body language in developing their perception of how 'safe' and interested they were.
Give information to young people in advance to explain that the hospital is a place where they can go to talk about how they feel, to bring this into the young person's awareness before they arrive at a hospital for their appointment.	Most young people identified that it would be helpful for clinicians to provide clear permission to disclose psychological difficulties, in line with evidence reinforcing the need for disclosure to be scaffolded by others (Crocker et al., 2020; Radez et al., 2020).
Clinician Responses	

Clinicians shared concerns that opening-up conversations about emotions would lead to requests for referrals at a rate that could not be managed by existing psychological provision and that they felt they should be prioritising access to psychology for young people with conditions such as IBD. This reflects very real limitations in service provision, but may also validate participants experiences of feeling that their condition is perhaps ‘less important’ than other gastrointestinal conditions. Through discussion, clinicians were made aware that most young people would likely not need psychology input but would benefit from having their feelings validated and normalised and perhaps being directed towards self-help resources. This would be in line with a stepped care design, which would consider the most efficacious use of resources in a busy paediatric hospital setting (Hamall et al., 2014; NICE, 2019). The MDT Psychologist is motivated to continue to work alongside the MDT to further support MDT recognition of the interdependence among patient’s biological, social, and psychological needs in line with the 10-year plan (NHS England, 2019).

Conclusions

This is the first UK study to directly explore the barriers perceived by young people with CD to disclosing psychological difficulties to clinicians during routine consultations. Findings highlight the importance of clinicians providing developmentally appropriate information to young people and actively breaking down barriers through body language and use of clear questions regarding emotional experiences. Feedback from clinicians in the team suggests that gaps in the provision of developmentally appropriate information and barriers to disclosure of psychological difficulties are perpetuated by resource constraints and a need for improved person-centred, MDT integrated working. Future research could explore this more systematically through interviews with clinicians.

Findings demonstrate an alignment in experience between the seven participants interviewed, suggesting reliability and generalisability. However, the sample comprised of a small number of white British participants receiving support from a single service, which may limit the cultural and geographical generalisability of findings. Several validation methods and credibility checks were used, however the analysis presented is the researchers' interpretation of the data and other interpretations are possible.

This study emphasises the value of in-depth, qualitative feedback from patients in identifying areas for service improvement. Although the provided recommendations are made based on young people's experiences of the stakeholder service, it is hoped they may positively benefit patient experience and improve access to psychological support in other services.

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Main Research Project

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Does Severity of Post-Stroke Cognitive Impairment Moderate the Relationship Between Cognitive Change and Emotional Distress in the Months Following Stroke?

Word Count: 5,121 words (max 5000 words; excluding References and Appendices)

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Proposed Journal: This report is intended for submission to *Neuropsychological Rehabilitation* and has been written and formatted in accordance with this (see Appendix 3.A). This journal has been selected due to its focus on research related to rehabilitation, recovery of function, and brain plasticity. The authors intend to submit this paper to a special issue on the topic of 'the self after brain injury', following a call for papers exploring the relationship between self-awareness and outcomes after brain injury. Research similar to this paper on the topics of stroke and self-awareness, with similar methodologies, have been published by this journal.

Abstract

This study explores whether the severity of acute post-stroke cognitive impairment moderates the relationship between cognitive change and emotional distress in the first 6-months following stroke. 143 acute stroke patients were assessed within the first 3-weeks (T1) and at 6-months (T2) post stroke. Cognitive impairment was assessed at both time points using the Oxford Cognitive Screen (OCS). At 6-months, awareness of cognitive impairment and emotional distress were assessed using the Cognitive Failures Questionnaire (CFQ) and the Hospital Anxiety and Depression Scale (HADS), respectively. An ANCOVA revealed that awareness of cognitive impairment was significantly lower in the 'moderate-severe cognitive impairment' group (≥1 *SD* from the sample cognitive impairment mean) than the 'mild-no cognitive impairment group' (<1 *SD* from the sample cognitive impairment mean; $p < .001$) at 6-months post stroke. A hierarchical multiple regression analysis indicated that awareness of cognitive impairment and Barthel Index were significant predictors of HADS ($R^2 = .263$; $p < .001$). A subsequent moderation analysis revealed that the effect of cognitive change on HADS was moderated by cognitive impairment at T1 ($R^2 = .391$; $p < .001$). The interaction variable explained 2.2% of variance in the model. Specifically, those with more severe cognitive impairment at T1 reported lower emotional distress as a function of cognitive improvement. Implications of these findings are discussed and recommendations for further research are suggested.

Keywords: Stroke; Depression; Anxiety; Awareness; Cognition

Introduction

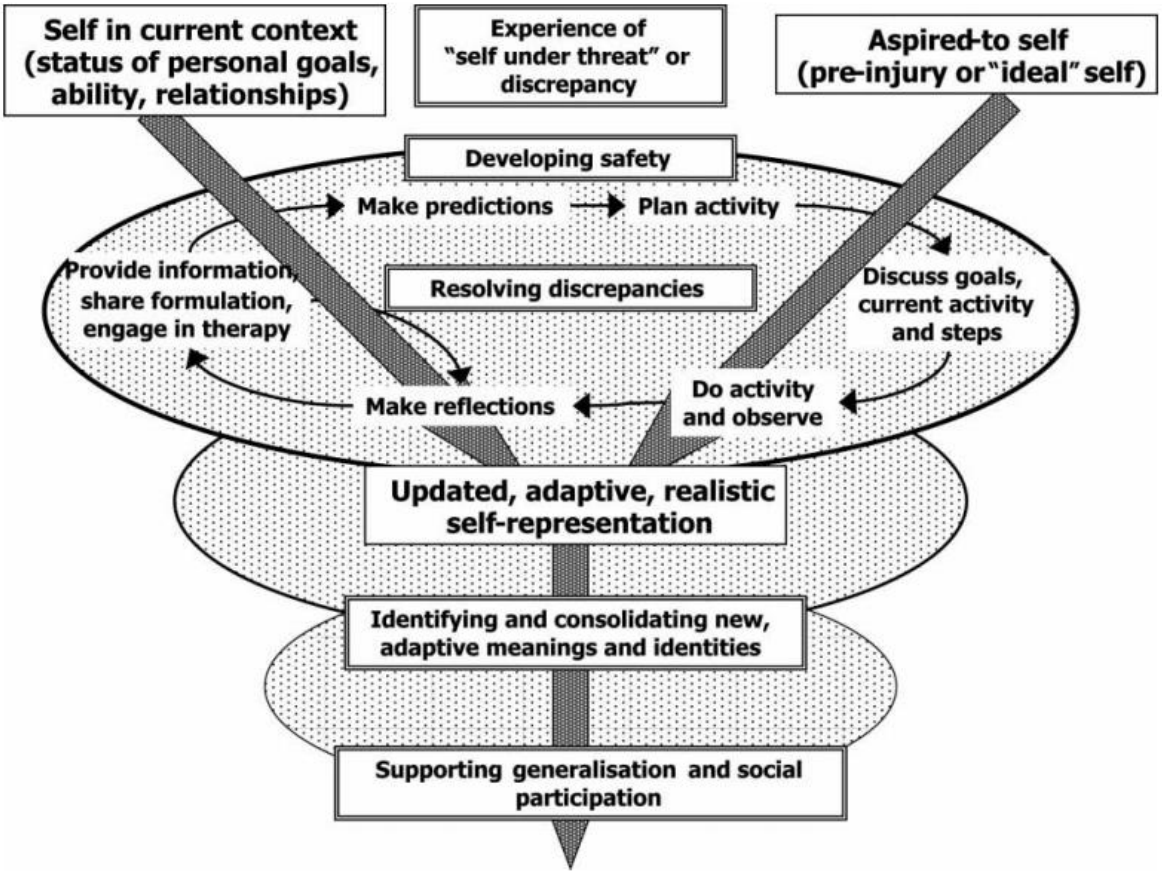
Stroke is the fourth most common cause of death and the leading cause of complex disability in the UK (Adamson et al., 2004; Office for National Statistics, 2018). Post-stroke depression (PSD) and anxiety (PSA) are common psychological consequences of stroke, with prevalence estimated to be between 15% and 40%, and 18% and 24% respectively (Knapp et al., 2020; Mitchell et al., 2017; Wu et al., 2019). Long-term effects of emotional distress have been shown to impact quality of life, physical recovery, and mortality (Ayerbe et al., 2013; Cai et al., 2019; Donnellan et al., 2010; Ezema et al., 2019; Kim et al., 2018). The number of individuals living with the long-term physical and psychological effects of stroke are predicted to increase by >60% by 2035, with the cost of social care for stroke survivors projected to increase by 250% in this period (Wittenberg & King, 2020). Consequently, it is imperative to identify patients at risk of adverse emotional and functional outcomes after stroke to optimise support from services through early detection and treatment.

The "Y-shaped" process model (subsequently referred to as the Y-shaped model; Gracey et al., 2009) indicates how biological, psychological, and social factors can interact to determine outcomes following acquired brain injury (ABI). The model suggests that following ABI, individuals undergo a process of adaptation and reintegration into society, which initially involves them developing awareness, understanding, and adaptive resolution of social and psychological discrepancies in their sense of self (see Figure 1). Gracey et al. (2009) set out three key discrepancies that survivors may experience following ABI: social, interpersonal, and personal discrepancies. It is proposed that when a stroke survivor is aware of such discrepancies, they may experience psychological threat regarding feared and actual catastrophic meanings associated with their post-injury situation. This may lead them to experience automatic emotional reactions and to adopt safety-seeking behaviours to cope with such threats. The model hypothesises that safety-seeking behaviours can reduce threat-

to-self in the short term. However, over time this can prevent the individual from resolving discrepancies, increasing their risk of experiencing poor long-term mental health and psychosocial outcomes.

Figure 1

The "Y-Shaped" Process Model (Gracey et al., 2009)



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The Y-shaped model draws on evidence regarding an individual's 'awareness' of changes to their sense of self post injury. 'Awareness' in the context of disability is understood as one's ability to recognise deficits, understand the functional implications of these deficits, and set realistic goals accordingly (Ownsworth et al., 2007). Awareness can relate to an individual's insight into a specific deficit in their sensory, motor, perceptual, behavioural, or cognitive functioning (Orfei et al., 2009). Awareness typically increases in the years following brain injury, with awareness of physical changes typically preceding

awareness of cognitive and behavioural changes (Hart et al., 2009; Hart et al., 2004). In the literature, ‘awareness’ in the context of brain injury is commonly operationalised as the difference between an individual’s subjective account of their deficits and either an objective test of their deficits, or a second rater of that individual’s deficits (Boosman et al., 2014; Hoofien et al., 2004; Ownsworth et al., 2007). However, there is currently no gold standard measure of awareness (Bogod et al., 2003; Geytenbeek et al., 2017).

Evidence suggests that awareness deficits arise from an interaction between neurological, psychological, and socio-environmental factors (Gracey et al., 2009; Ownsworth et al., 2007). Literature exploring the epidemiology of awareness of cognitive impairment indicates that under-reporting cognitive impairment following stroke may be due to organic changes in the brain which inhibits one’s ability to understand the extent of one’s cognitive impairment (Boosman et al., 2014; Ownsworth et al., 2007). In contrast, subjectively over-reporting the extent of one’s cognitive impairment may be due to psychological mechanisms such as attentional bias, which may lead to hyper-awareness (Boosman et al., 2014; Ownsworth et al., 2007). Evidence suggests that those who are hyper-aware of cognitive impairment are more likely to experience clinically significant psychological distress (Boosman et al., 2014; Carroll & Coetzer, 2011; Geytenbeek et al., 2017; Morton & Barker, 2010; Ownsworth et al., 2007). In line with this, literature indicates that awareness of cognitive impairment influences emotional wellbeing in individuals with long-term disabilities, such as stroke, more than objective cognitive impairment (OCI) alone (Mitrushina & Tomaszewski, 2020; Van Rijsbergen et al., 2019).

Based on the Y-shaped model, one can hypothesise that the emotional consequences of cognitive change in the first 6-months following stroke may differ depending on the level of acute cognitive impairment. Specifically, one may expect that individuals with less acute cognitive impairment and higher levels of awareness may report lower levels of distress following cognitive improvement, due to a perceived reduction in self-discrepancy.

However, one may expect that individuals with greater cognitive impairment, and thus lower levels of awareness immediately post stroke, may report higher levels of distress as a function of cognitive improvement. Specifically, they may gain access to the cognitive skills which enable them to process their situation, potentially leading to greater psychological threat.

This study thus aims to answer the following research questions:

Preliminary Research Questions and Hypotheses

1. What is the relationship between cognitive impairment at 6-months post stroke and awareness of cognitive impairment at 6-months post stroke?

H1: There will be a significant difference in awareness of cognitive impairment between the moderately-severely cognitive impairment and mild-no cognitive impairment groups at 6-months post-stroke.

2. What is the relationship between awareness of cognitive impairment at 6-months post stroke and emotional distress at 6-months post stroke?

H2: There will be a significant association between awareness of cognitive impairment and emotional distress at 6-months post-stroke.

Primary Research Question and Hypothesis

1. Does severity of acute post-stroke cognitive impairment moderate the relationship between cognitive change and emotional distress in the months following stroke?

H1: The effect of cognitive change between subacute and 6-months post-stroke on mood at 6-months post-stroke will be moderated by subacute cognitive impairment.

Findings may have important implications for psychological intervention. For instance, through supporting resource-based decision-making with regards to targeted cognitive-behavioural interventions for stroke-survivors at risk of long-term adverse mood outcomes.

Materials and Methods

Design and Participants

The study used a cross-sectional and longitudinal design. A secondary data analysis was conducted on a subset of data originally collected from acute stroke units at the John Radcliffe Hospital between February 2015 and February 2020 as part of two studies of the Oxford Cognitive Screen (OCS; Demeyere et al., 2015). Namely the OCS-Tablet and OCS-Recovery (NHS Research Ethics Committee references: 14/LO/0648 and 18/SC/0550; see Appendix 3.B). Participants were included in these larger studies if they were adults within 3-weeks of confirmed ischemic or haemorrhagic stroke, able to give informed consent, and able to concentrate for the duration of OCS administration.

Participants were included in the current study if they had completed at least 9 of out 12 subtests of the OCS (Demeyere et al., 2015) within 3-weeks of confirmed ischemic or haemorrhagic stroke (T1) and again approximately 6-months later (T2). Participants were required to have complete data for: (i) demographic information including age, sex, and years of education, (ii) stroke hemisphere, (iii) the National Institutes of Health stroke scale (NIHSS; Lyden et al., 1994), (iv) the Cognitive Failures Questionnaire (CFQ; Broadbent et al., 1982), (v) the Hospital Anxiety and Depression scale (HADS; Snaith & Zigmond, 1986), and (vi) the Barthel Index (BI; Mahoney & Barthel, 1965) at T2. Data for 253 participants were screened in line with these criteria and 143 participants were included in the current analysis (see Appendix 3.C).

Measures

OCI was measured using the OCS, a short cognitive screen designed for use with stroke patients (Demeyere et al., 2015). Domain specific scores relating to apraxia, unilateral neglect, memory, language, executive function, and number abilities are generated. Impairments on the OCS are indicated by a score lower, or in some cases a score higher, than the normative 'cut-off' (5^{th} or 95^{th} centile respectively). It has good test-retest reliability, content validity, specificity, and high sensitivity (Demeyere et al., 2015).

Subjective cognitive impairment was assessed with the CFQ, a 25-item self-report questionnaire whereby subjects rate the frequency of everyday cognitive errors on a 5-point Likert scale ranging from 0 (never) to 4 (very often; Broadbent et al., 1982). This measure is frequently used in stroke samples and has good psychometric properties (Barker-Collo et al., 2010; Bridger et al., 2013; Westerberg et al., 2007).

Emotional distress was assessed with the HADS, a 14-item questionnaire examining symptoms of anxiety and depression on a 4-point scale, whereby higher scores indicate greater symptomatology (Snaith & Zigmond, 1986). Caseness is defined by a score of >8 on both subscales (Bjelland et al., 2002). It has been used with stroke survivors and reports good internal consistency (Cronbach $\alpha = 0.8$), with high specificity and sensitivity in this population (Aben et al., 2002). Total HADS score was used as a measure of emotional distress in the main analyses.

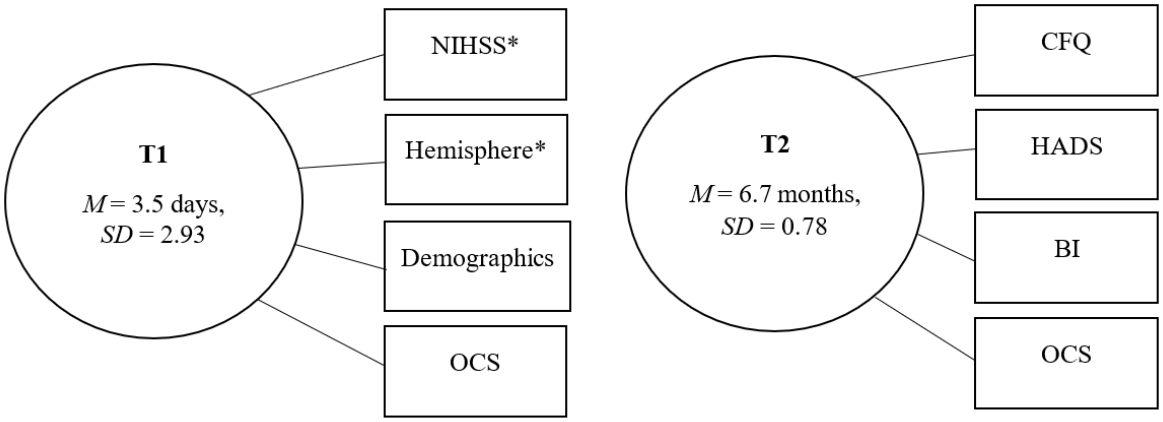
Participant age, sex, and years of education were recorded. The NIHSS, a proxy measure of stroke severity (Lyden et al., 1994), and stroke hemisphere were extracted from participant medical records. Participants also completed the BI, a measure of disability relating to activities of daily living (ADL; Mahoney & Barthel, 1965).

Procedure

Participants were initially assessed with the OCS during admission to an acute stroke unit, up to three weeks after their stroke (T1, *M* = 3.5 days, *SD* = 2.93). A six-month follow-up assessment was planned (T2), although due to delays in contacting and reaching participants following discharge, the actual time interval differed across participants (5 – 9 months, *M* = 6.7 months, *SD* = 0.78). Questionnaires were administered at T2 in addition to the OCS. Stroke characteristic data were extracted from medical records (NIHSS; hemisphere). Assessments took place at the hospital (T1) and at the patient's home (T2). Figure 2 illustrates which assessments were administered at each timepoint.

Figure 2

Summary of Assessments Administered at T1 and T2



Note. Demographics data include age, sex, and years of education; *Data was extracted from patient medical records at T1. NIHSS: the National Institutes of Health stroke scale; OCS: Oxford Cognitive Screen; CFQ: the Cognitive Failures Questionnaire; HADS: the Hospital Anxiety and Depression scale; BI: the Barthel Index.

Data Analyses

Statistical analyses were conducted using the SPSS 27.0 statistical package. Power analyses in GPower 3.1 determined the power for a sample size of 143 participants in each

analysis (see Appendix 3.D; Faul et al., 2009). All hypotheses, methods, and the analysis plan were pre-registered on OSF prior to data analysis (https://osf.io/87kjv).

Computing Variables

Objective Cognitive Impairment (OCI). Raw scores on the OCS at T1 and T2 were used to compute OCI scores at T1 and T2, respectively. These scores were quantified by summing the number of subtests on which a participant scored outside the norm-based cut-off values on the OCS (Demeyere et al., 2015), following the approach of others (e.g. Van Rijsbergen et al., 2020). As a small number of subtests were missing for some participants, the percentage of tests failed was calculated from the number of attempted subtests at each time-point (range 0-12 subtests). The percentage of tests 'failed' comprised the OCI scores (range 0-100%) at T1 and T2, whereby higher scores indicate greater cognitive impairment (see Appendix E).

For analysis one, participants were grouped into the 'moderate-severe cognitive impairment' group (defined as those with a T2 OCI score $\geq$1 *SD* from the sample OCI mean) and the 'mild-no cognitive impairment group' (defined as those with a T2 OCI score <1 *SD* from the sample OCI mean).

Cognitive Change. Existing studies use a range of criteria for indicating cognitive change in stroke samples, thus there is no widely agreed 'gold standard' for calculating this (Brainin et al., 2015). Here, cognitive change was quantified as the difference between the patient's OCI scores over time (T2 - T1, range -100% to 100%, positive scores indicate cognitive decline; see Appendix 3.E).

Awareness. Awareness scores were generated to represent participants' awareness of their cognitive impairment, following a similar procedure used by Mitrushina and Tomaszewski (2020). First, psychometric *Z*-scores were computed from OCI scores at T2. Secondly, psychometric subjective cognitive impairment *Z*-scores were computed for each

participant using raw total scores on the CFQ. Then, OCI *Z*-scores were subtracted from the CFQ *Z*-scores ($Z_{CFQ} - Z_{OCI_T2}$). Awareness scores were computed by transforming the answer into a *T*-score (see Appendix 3.E for further details). This created a distribution of awareness scores centred around a mean of 50 (accurately reporting level of cognitive impairment), with lower scores representing lower levels of awareness (under-estimating cognitive impairment) and higher scores representing hyper-awareness (over-estimating cognitive impairment).

Minimising Bias

To determine whether any selection bias occurred between participants who were re-examined at follow-up and those who were not, and between those who were excluded due to missing data and those who were not, a series of binary logistic regression analyses were carried out whilst correcting for multiple comparisons using the Benjamini and Hochberg (1995) false discovery rate method. Demographic variables, stroke characteristics, and OCI at T1 were predictor variables.

Main Analyses

Prior to the main analyses, data were examined to check whether necessary statistical assumptions had been violated. To identify possible covariates, a series of univariate correlations for continuous predictors and *t*-tests/ANOVAs for categorical predictors were carried out to explore the association between demographic/stroke characteristic variables and the dependent variable (DV) in each analysis (see Appendix 3.G).

To investigate whether there was a difference in awareness between the 'moderate-severe cognitive impairment' group and the 'mild-no cognitive impairment group', an ANCOVA was carried out whilst controlling for BI. To explore the relationship between awareness and emotional distress, a hierarchical multiple linear regression analysis was performed with emotional distress as the DV. BI was entered into the first step as a covariate

(Model 1) and awareness was entered into step two (Model 2). To investigate whether OCI at T1 has a moderating effect on the relationship between cognitive change and emotional distress, the PROCESS plug-in (Hayes, 2017) was used to conduct a moderation analysis. Emotional distress was entered as the DV, cognitive change was entered as the predictor, OCI at T1 was entered as the moderator, and awareness and BI were entered as covariates.

Follow-Up Exploratory Analyses

Exploratory logistic regression follow-up analyses were conducted to ascertain the effects of BI, awareness, and OCI at T2 on the likelihood that participants would report clinically significant symptoms of anxiety and depression.

Results

One-hundred and forty-three participants met criteria for inclusion. Descriptive statistics for clinical and demographic variables for participants are shown in Table 1. Binary logistic regression analyses indicated that completion of follow-up (yes/no) and exclusion from the present study (yes/no) were not predicted by demographic variables, stroke characteristics, or OCI at T1 at $p < .1$ (see Appendix 3.F).

Table 1

Clinical and Demographic Characteristics of Included and Excluded Participants

	Included participants (*N* = 143)				Excluded participants (*N* = 110)			
Variable	*M (SD)*	*Mdn*	Range	*n* (%)	*M (SD)*	*Mdn*	Range	*n* (%)
Age	73.33 (13.73)	75	18 - 93	143 (100)	73.35 (13.17)	75	31 - 95	109 (99)
Sex				143 (100)				110 (100)
Male				74 (52)				61 (55)
Female				69 (48)				49 (45)
Education (years)	12.45 (3.34)	11	6 - 23	143 (100)	12.58 (3.42)	11	9 - 27	53 (48)
Stroke Hemisphere				143 (100)				73 (66)
Right				66 (46)				38 (35)

Left				64 (45)				31 (28)
Both				13 (9)				4 (3)
NIHSS Total	7.70 (7.91)	5	0 - 42	143 (100)	5.71 (4.87)	5	0 - 17	31 (28)
BI Total	16.42 (4.44)	18	1 - 20	143 (100)	16.71 (4.50)	19	3 - 20	90 (82)
HADS-A	5.49 (4.08)	5	0 - 19	143 (100)	5.58 (4.23)	5	0 - 20	101 (92)
HADS-D	4.77 (3.98)	4	0 - 19	143 (100)	5.28 (4.14)	5	0 - 19	101 (92)
HADS Total	10.26 (7.18)	9	0 - 34	143 (100)	10.86 (7.61)	10	0 - 36	101 (92)
CFQ Total	29.39 (17.56)	27	1 - 92	143 (100)	32.58 (18.88)	31	0 - 93	99 (90)
OCI (%) T1	29.75 (25.35)	20	0 - 92	143 (100)	31.46 (28.18)	25	0 - 100	110 (100)
OCI (%) T2	23.50 (16.23)	17	8 - 83	143 (100)	26.56 (17.52)	18	0 - 83	97 (88)

Note. See main body for definitions of variables; *n* = sample total.

Main Analyses

1. *What is the Relationship Between Cognitive Impairment at 6-Months Post Stroke and Awareness of Cognitive Impairment at 6-Months Post Stroke?*

A series of univariate analyses identified that BI was significantly associated with awareness ($r_s(141) = .14$, $p = .090$; see Appendix 3.G), so was included as a covariate. A robust ANCOVA was carried out as the assumption of homogeneity of regression slopes was not met (see Appendix 3.H; Mair & Wilcox, 2020). This produced the same findings as an ANCOVA, therefore unadjusted data are reported.

A one-way ANCOVA was carried out to explore whether there were any significant differences in awareness score between the 'moderate-severe cognitive impairment' group and the 'mild-no cognitive impairment group', whilst controlling for BI (see Table 2 for adjusted descriptive statistics). BI was not significantly related to awareness ($F(1, 140) = 0.58$, $p = .449$). There was a significant effect of severity of OCI at T2 on awareness after controlling for the effect of BI, ($F(1, 140) = 104.81$, $p < .001$, partial $\eta^2 = .43$) suggesting that awareness was significantly lower in the 'moderate-severe cognitive impairment' group (see Figure 3).

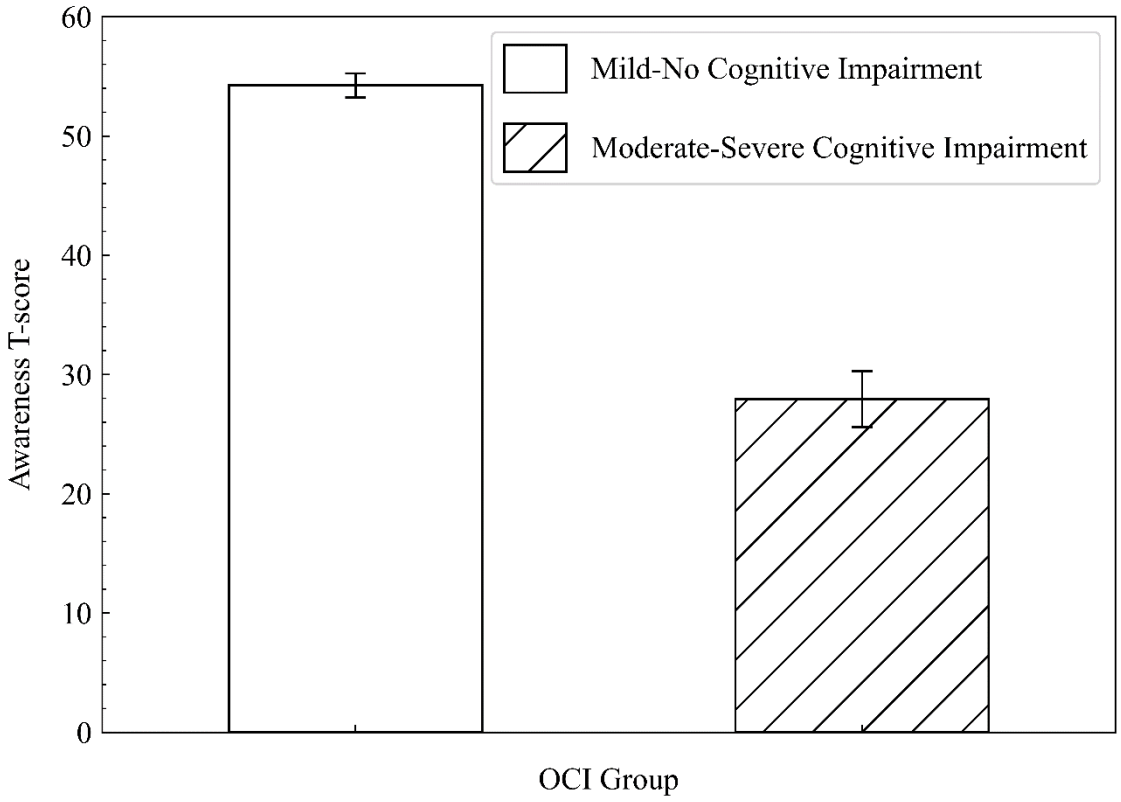
Table 2

Unadjusted and Covariate Adjusted Descriptive Statistics for Awareness

OCI Group	BI			Awareness (Unadjusted)		Awareness (Adjusted)	
	N	*M*	*SE*	*M*	*SE*	*M*	*SE*
Mild-no impairment	120	16.78	0.38	54.17	1.01	54.23	1.02
Moderate-severe impairment	23	14.52	1.15	28.23	2.30	27.92	2.35

Figure 3

The Difference in Mean Awareness T-Scores Between the 'Mild-No Impairment' and 'Moderate-Severe Impairment' Groups at 6-Months Post Stroke



Note. Error bars represent standard errors.

2. What is the Relationship Between Awareness of Cognitive Impairment at 6-Months Post Stroke and Emotional Distress at 6-Months Post Stroke?

In the sample, 28% and 22% of participants met the clinical threshold for anxiety and depression on the HADS, respectively. A series of univariate correlations identified that the only statistically significant relationship between demographic or stroke-related variables and HADS was the relationship between BI and HADS ($r_s(141) = -.37$, $p < .001$; see Appendix 3.G). Therefore, BI was included as a covariate in this analysis. The assumptions of multiple regression were tested and met accordingly (see Appendix 3.I).

A hierarchical multiple regression analysis was carried out whereby potential predictors of HADS Total were entered in blocks. BI was entered first as a covariate, then awareness was added in the second block. The final model indicated that BI was a negative predictor (b = -.63, CI [-.86, -.40], t = -5.33, p < .001), and awareness was a positive predictor (b = .18, CI [.11, .25], t = 5.03, p < .001), together explaining 25.2% variance in the model ($R^2_{Adjusted}$ = .252, F(2, 140) = 24.98, p < .001), with a large effect size of f^2 = .34.

3. Does Severity of Acute Post Stroke Cognitive Impairment Moderate the Relationship Between Cognitive Change and Emotional Distress in the Months Following Stroke?

All assumptions of the moderation analysis were met (see Appendix 3.J). In line with the third hypothesis, the model indicates that cognitive change was a positive predictor of HADS, whereby a reduction in impairment over time was associated with a lower HADS score (b = .19, 95% CI [.09, .30], t = 3.57, p < .01), and this was moderated by OCI at T1 (b = .0020, 95% CI [.0002, .0038], t = 2.21, p < .05). However, the interaction variable only explained 2.2% of variance in the model (R^2 Change = .022, F(1, 137) = 4.88, p < .05). Together, predictors explained 36.9% variance in the model ($R^2_{Adjusted}$ = .369, F(5, 137) = 17.57, p < .001; see Table 3), with a large effect size of f^2 = .58.

The Johnson-Neyman (1936) method was used to explore the directionality of the moderation effect (see Appendix 3.K). Figure 4 shows how the relationship between cognitive change and HADS changes at different values of OCI at T1. This indicates that those with a higher OCI score at T1 are more sensitive to the effects of cognitive change on HADS. Specifically, those with more severe cognitive impairment at T1 reported lower emotional distress at T2 as a function of cognitive improvement.

Table 3

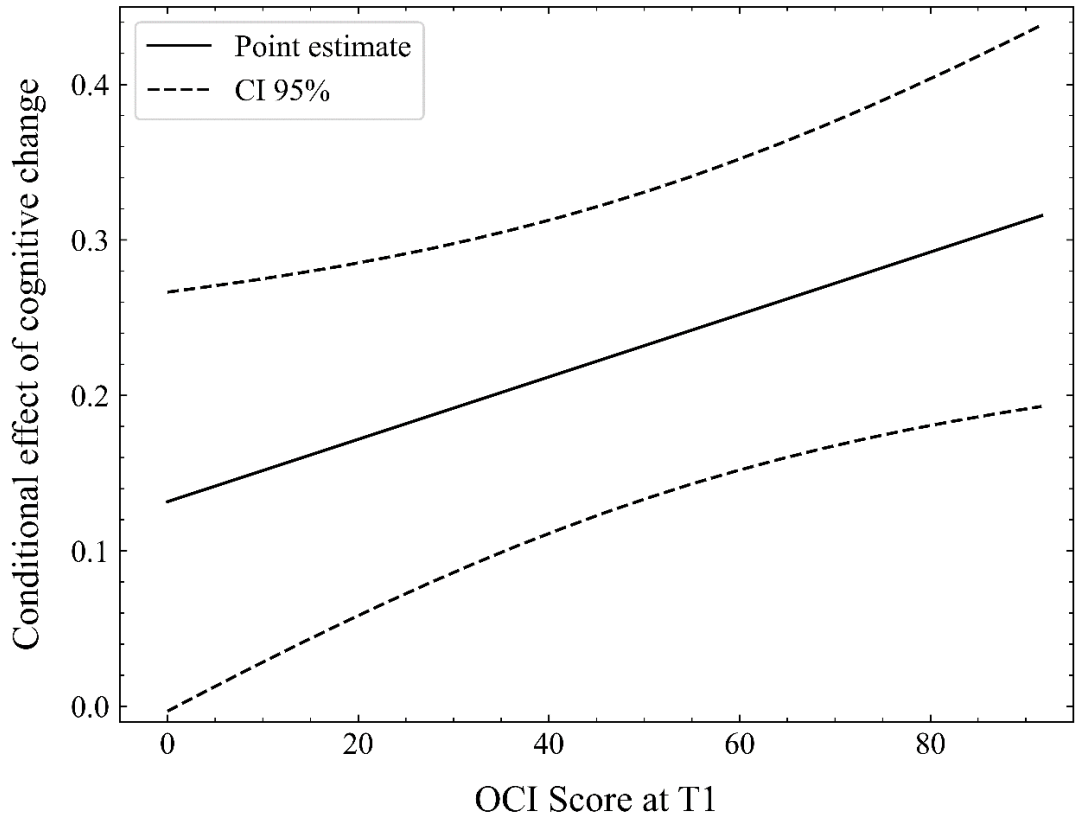
Hierarchical Multiple Regression Analyses of OCI at T1 as a Moderator of the Relationship Between Cognitive Change and Emotional Distress (HADS Total Score) Controlling for the Effect of BI and Awareness

	b (CI)	*SE b*	Beta	*t*	*p*
Constant	-0.49 (-7.27, 6.29)	3.43		-0.14	.886
Cognitive change	0.19 (0.09, 0.30)	0.05	.59	3.57	.001
OCI at T1	0.22 (-0.13, 0.31)	0.05	.79	4.83	.000
Interaction	0.0020 (0.0002, 0.004)	0.0009	.21	2.21	.029
BI	-0.42 (-0.65, -0.19)	0.12	-.26	-3.64	.000
Awareness	0.37 (0.27, 0.47)	0.05	.75	7.49	.000

Note. R^2 = .391; 95% confidence intervals reported in parentheses. Cognitive change and OCI at T1 are centred variables. The interaction variable is defined as cognitive change × OCI at T1.

Figure 4

The Conditional Effect of Cognitive Change on HADS Score as a Function of OCI Score at T1



Note. Conditional effect of cognitive change represents the gradient between cognitive change and HADS score for a given OCI score at T1. Those with a higher OCI score at T1 are more sensitive to the effects of cognitive change on HADS. Upper and lower 95% confidence intervals are indicated by the dashed lines.

Exploratory Analyses

Further exploratory analyses were conducted to examine whether BI, awareness, and OCI at T2 were differentially related to anxiety and depression symptomatology. Two forced-entry logistic regression analyses were performed with BI and awareness entered as predictors in the first, and BI, awareness, and OCI at T2 entered as predictors in the second.

OCI at T2 did not significantly improve the anxiety model so was excluded. The assumptions of the logistic regression were met for both models (see Appendix 3.L).

Increasing awareness was associated with an increased likelihood of reporting clinically significant anxiety (b = .046, 95% CI [.012, .087], p < .01, OR = 1.05, 95% CI [1.01, 1.08]) and depression symptomatology (b = .097, 95% CI [.054, .154], p < .001, OR = 1.10, 95% CI [1.05, 1.20]). Higher BI scores were associated with a reduction in the likelihood of clinically significant anxiety (b = -.147, 95% CI [-.252, -.051], p < .01, OR = 0.863, 95% CI [0.79, 0.95]) and depression symptomatology (b = -.150, 95% CI [-.261, -.063], p < .01, OR = 0.861, 95% CI [0.78, 0.96]). Higher OCI T2 scores were associated with an increase in the likelihood of clinically significant depression symptomatology (b = .07, 95% CI [.02, .12], p < .01, OR = 1.07, 95% CI [1.02, 1.12]); see Appendix 3.M for further details).

Discussion

To the authors knowledge, this is the first study to empirically test the Y-Shaped model. This study explored whether well-established relationships between severity of cognitive impairment and awareness of cognitive impairment, and between awareness of cognitive impairment and emotional distress could be demonstrated in this stroke sample. This study then explored whether the effect of cognitive change over the first 6-months post stroke on mood is moderated by the severity of acute cognitive impairment.

Main Findings

Findings suggest a strong relationship between awareness of cognitive deficits and cognitive functioning, with participants reporting accurate awareness when only mildly cognitively impaired and participants with more severe or global cognitive impairment underestimating their cognitive deficits. This finding supports evidence that poor awareness of cognitive deficits is associated with organic changes sustained by brain injury through

stroke (Boosman et al., 2014; Ownsworth et al., 2007). In line with the Y-shaped model, those with greater cognitive impairment may have reduced capacity to process, retain, or understand the cognitive consequences of their stroke (Gracey et al., 2009).

With regards to the second hypothesis, findings suggest that greater awareness of cognitive impairment is strongly associated with greater distress, in line with previous work in various ABI samples (Boosman et al., 2014; Geytenbeek et al., 2017; Mitrushina & Tomaszewski, 2020; Morton & Barker, 2010). Moreover, findings suggest that BI is a negative predictor of emotional distress in accordance with previous research (Huang et al., 2014; Park et al., 2012). These findings may be explained by the relationship between 'threat-to-self', 'self-discrepancy', and emotional distress prescribed by the Y-shaped model. Specifically, an individual with greater awareness of their cognitive deficits and higher levels of disability in carrying out ADLs following stroke may experience a threatened pre-injury identity, accompanied by an increase in emotional distress (Gracey et al., 2009). Alternatively, it is possible that increased emotional distress heightens one's awareness of self-discrepancy posed by cognitive change post stroke. This should be investigated in a future longitudinal study which measures the dynamic influence of variables at multiple time-points.

The final analysis explored the effect of cognitive change on emotional distress, and whether this was moderated by severity of OCI at T1. Findings suggest that cognitive improvement in the months following stroke is significantly associated with lower reported emotional distress. In line with the Y-shaped model, improvement in cognitive performance may reduce the discrepancy between pre- and post-injury cognitive ability, enabling the individual to progress through a process of post stroke adjustment, associated with decreased distress (Gracey et al., 2009). However, the causal mechanism cannot be established in current findings due to the absence of a measure of threat-to-self and adjustment.

The moderation model indicates that the effect of cognitive change was moderated by acute cognitive impairment (see Figure 4). To the best of the authors' knowledge, this is the first observation that emotional outcomes in the months following stroke depend, partially, on the interaction between cognitive change and the severity of acute post-stroke cognitive impairment. Specifically, those with more severe acute cognitive impairment reported lower emotional distress as a function of cognitive improvement. This finding appears incongruent with the awareness hypothesis (Boosman et al., 2014; Ownsworth et al., 2007). One may expect that an individual with a greater acute cognitive impairment would have lower levels of awareness, and thus low emotional distress. If an under-aware individual's cognitive function were subsequently to improve, one may expect that they may gain the capacity to become aware of any discrepancy in their pre- and post-stroke self, which may then lead to increased emotional distress. This incongruence would suggest that awareness of cognitive impairment is not the dominant factor in determining the relationship between acute cognition and anxiety and depression symptomatology at 6-months, which is supported by the $R^2 = .39$ of the model. Improvements in cognition may enable more cognitively impaired individuals to implement behavioural compensatory strategies, which may have a notable impact on their everyday functioning and mood (Nijboer et al., 2013). As the mechanisms and direction of the association between change in cognition and mood cannot be established in this study, this should form the basis for future research.

Exploratory Findings

Findings suggest that awareness and BI are significant predictors of clinically significant anxiety and depression symptomatology. These findings differ from those of Geytenbeek et al. (2017), who reported that depression was not associated with awareness in a TBI sample. This may be a consequence of methodological differences including sample characteristics and measures used. Current findings also suggest that cognitive impairment is a significant predictor of depression symptomatology. This is in line with previous research,

which suggests that deficits in attention, memory, processing speed, and executive functioning, are commonly experienced by stroke survivors and are also associated with depression symptomatology (Broomfield et al., 2011; Nakling et al., 2017; Nys et al., 2006). This may be due to stroke induced damage to the frontal subcortical circuits, which are responsible for both executive skills and emotional regulation (Alexopoulos et al., 1997; Flaster et al., 2013). Moreover, changes to executive skills may interact with psychological mechanisms to further maintain low mood, for example, through exacerbating difficulties with flexible thinking and problem solving (Broomfield et al., 2011). However, due to the exploratory nature of these findings, future studies should use a hypothesis-driven approach to explore this further.

Clinical Implications

Findings emphasise the importance of adopting a biopsychosocial approach to neurorehabilitation, including monitoring stroke patients' cognitive deficits and their capacity to be self-aware when assessing and formulating long-term post-stroke distress and adjustment (Mitrushina & Tomaszewski, 2020; Van Rijsbergen et al., 2019). Findings suggest that patients with more severe or global cognitive impairment 6-months post stroke are less likely to understand, retain, or accept post-stroke cognitive change. Although low awareness appears to be protective against emotional distress at 6-months, present findings suggest that cognitive improvement over the first 6-months post stroke is associated with lower reported emotional distress. Moreover, this effect is largest for those most cognitively impaired in the days following stroke. This has possible implications for the provision of psychological intervention in this subgroup. Namely, targeting this subgroup of patients with psychoeducation and feedback regarding cognitive change and cognitive rehabilitation interventions, alongside emotional support in the early stages post stroke may have a positive impact on their long-term emotional adjustment. In contrast, cognitive improvement

over the first 6-months appears to have a much smaller impact on the emotional wellbeing of those with less cognitive impairment in the weeks following stroke. Psychological interventions such as cognitive behavioural therapy may be better suited to this group, to support them with the process of personal reconciliation.

Strengths and Limitations

A methodological advantage of this study is that participants' self-evaluations of their cognitive abilities were compared to their neuropsychological performance, as opposed to scores based on evaluations provided by family members or clinicians, thus yielding a more objective measure of awareness (Hoofien et al., 2004). A further strength of this study is the large sample size and inclusion of participants with communication difficulties, which makes findings more generalisable to clinical populations.

Some limitations of this study should be noted. Firstly, the measure of awareness used in this study perhaps reflects intellectual awareness, in that participants were asked to rate the implications of cognitive changes post injury. However, 'online awareness', defined as the ability to self-monitor and modify behaviour during performance, was not measured (Toglia & Kirk, 2000). Additional research is required to determine a gold-standard measure for detecting multiple aspects of impaired self-awareness, according to normalised cut-off scores. Secondly, emotional distress was considered as an outcome in this study, however, it could also be a potential risk factor for awareness and cognitive impairment, and this should be explored in future studies. Thirdly, the study sample consisted of primarily patients with a mild stroke (median NIHSS score 5 out of 42) and patients with high levels of functional independence in personal care and mobility (median BI score 18 out of 20), which makes generalisability of the findings to the entire stroke population difficult. Fourthly, data was not collected regarding premorbid psychological history, despite evidence that this variable

is associated with self-awareness, PSD, and psychosocial stroke outcomes (Hurst et al., 2020; Taylor-Rowan et al., 2019).

Conclusion

This study suggests that emotional outcomes in the months following stroke depend partially on the interaction between cognitive change and the severity of stroke-induced cognitive impairment. Specifically, those with more severe acute cognitive impairment saw greater reduction in their emotional distress as a function of cognitive improvement. These results have potential implications for improving long-term emotional adjustment in this sub-group of patients through psychoeducation and cognitive rehabilitation interventions, alongside emotional support.

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Executive Summary

Background

A stroke is a brain attack caused when the blood supply to the brain is cut-off, killing brain cells. More people are surviving stroke than ever before, although many are left with physical disabilities, cognitive impairment, and report poor mood outcomes.

The "Y-Shaped" process model suggests that as stroke survivors develop an awareness and understanding of the impact of these post-stroke changes on their functioning, they may experience a discrepancy between their 'pre-stroke' and 'post-stroke' identities, which may cause heightened distress. In line with this, evidence has suggested that those with greater post-stroke cognitive impairment had reduced awareness of these changes and reported 'normal' levels of anxiety and low mood. However, a subgroup of stroke survivors with mild post-stroke cognitive impairment had 'hyper-awareness' of these changes and reported high levels of anxiety and low mood.

Impairments in cognition often improve in the months following stroke, as the brain recovers. The "Y-Shaped" process model may suggest that cognitive improvement could lead those with more severe cognitive impairment to develop a greater awareness of the discrepancy between their 'pre-stroke' and 'post-stroke' identities, which may be associated with greater emotional distress. This study thus aims to investigate whether the extent of cognitive impairment in the days following stroke impacts the relationship between cognitive change and emotional distress at 6-months post stroke.

Research Questions

1. What is the relationship between cognitive impairment at 6-months post stroke and awareness of cognitive impairment at 6-months post stroke?
2. What is the relationship between awareness of cognitive impairment at 6-months post stroke and emotional distress at 6-months post stroke?

3. Does severity of post-stroke cognitive impairment in the days following stroke moderate the relationship between cognitive change and emotional distress in the months following stroke?

Methods

Participants were recruited from an inpatient stroke ward. One-hundred and forty-three stroke patients (74 men and 69 women) aged between 18 and 93 years (M = 73.33, SD = 13.73) were assessed within the first 3-weeks (T1) and at 6-months (T2) post stroke. Participants' cognition was assessed using the Oxford Cognitive Screen (OCS) at both T1 and T2. At T2, participants were also asked to complete the Hospital Anxiety and Depression Scale (HADS; a measure of emotional distress), the Cognitive Failures Questionnaire (CFQ; a measure of subjective cognitive impairment), and the Barthel Index (BI; a measure of disability relating to activities of daily living (ADL)). NIHSS score (a proxy of stroke severity) and stroke hemisphere were extracted from participant medical records. The difference between participants' objective and subjective cognitive impairment scores constituted their awareness score.

Participants were divided into a 'moderate-severe cognitive impairment' group and a 'mild-no cognitive impairment' group according to their scores on the OCS at T1, then mean awareness scores were compared between groups. Participants were assessed as a pooled sample for subsequent analyses.

Key Findings

In line with previous research, the 'moderate-severe cognitive impairment' group had significantly lower awareness of their cognitive impairments than the 'mild-no cognitive impairment' group at T2, when controlling for disability relating to ADL. Greater awareness of cognitive impairment and lower levels of disability relating to ADL were significant predictors of greater emotional distress at T2.

Greater awareness was associated with an increased likelihood of reporting clinically significant symptoms of anxiety and depression. Reduced levels of disability relating to ADL was associated with a reduction in the likelihood of reporting clinically significant symptoms of anxiety and depression.

Findings suggest that cognitive improvement over the first 6-months post stroke is associated with lower reported emotional distress. Moreover, this effect is largest for those most cognitively impaired in the days following stroke. On one hand, this is not in line with what we expected to find based on the "Y-Shaped" Process model. However, this finding may suggest that awareness of cognitive impairment is not the most important factor in determining emotional distress in the months following stroke.

Research Implications

To the best of the authors' knowledge, this is the first observation that emotional outcomes in the months following stroke depend, partially, on the interaction between cognitive change and the severity of cognitive impairment in the days following stroke. Future research could investigate the causal mechanisms underlying this relationship, by exploring the role of 'psychological threat', coping, and adjustment. Repeating this study with a sample of patients with moderate-severe strokes (as scored by the NIHSS) would also be warranted to determine whether findings can be generalised to other stroke samples. Investigating additional factors, including access to neurorehabilitation, pre-morbid mental health, and social support may further understanding of the factors that contribute to emotional distress in the months following stroke.

Clinical Implications

Findings emphasise the importance of clinicians considering the relationship between biological, psychological, and social factors following stroke, including monitoring stroke patients' cognitive deficits and their capacity to be self-aware. Although reduced

awareness appears to be protective against emotional distress at 6-months, our findings suggest that cognitive improvement over the first 6-months post stroke is associated with lower reported emotional distress, particularly for those most cognitively impaired in the days following stroke. Targeting this subgroup of patients with psychoeducation regarding cognitive changes whilst providing strategies to manage these, alongside emotional support in the early stages post stroke may have a positive impact on their long-term emotional adjustment. In contrast, cognitive improvement over the first 6-months appears to have a much smaller impact on the emotional wellbeing of those less severely cognitively impaired in the days following stroke. Psychological interventions such as cognitive behavioural therapy may be better suited to this group, to support them with long-term adjustment to stroke.

Connecting Narrative

CRL

I developed an interest in the Mental Capacity Act (MCA, 2005) during my undergraduate degree in psychology and law, whereby I developed critical thinking skills through considering the application of the law within the wider context of society. This fostered my curiosity regarding the translation of MCA (2005) principles into clinical practice. As my interest in neuropsychology developed through pre-training clinical experience, I became increasingly curious as to how cognitive tests could support this from a more objective position in complex cases.

A brief scoping search highlighted few papers directly referencing the MCA (2005) but several regarding how performance on cognitive tests related to capacity standards within the functional test (MCA, 2005; e.g., the ability to understand information pertaining to a decision). I initially chose to focus on literature regarding treatment decisions only. However, the research team suggested that I broaden the scope of the review to include all decisions to optimise its clinical utility. It was helpful to have early discussions around how to best structure the narrative synthesis as there were several possibilities (e.g., structure according to nature of decision, clinical population, capacity standard etc.). I decided to structure in accordance with capacity standard as I felt this would be more accessible for a busy clinician looking to use cognitive testing to support their assessment of a certain area of capacity when applying the functional test.

For me, carrying out the synthesis was one of the most challenging aspects of this project as I was confronted with significant diversity in findings. Supervision was helpful in reminding me that transparency and providing a clear rationale for my approach was acceptable. I feel that this process has also enabled me to learn how to critically appraise research and theory-practice links.

MRP

In line with my CRL interests, I was motivated to develop a main research project in neuropsychology. I was keen to collaborate with the Translational Neuropsychology research group at the University to develop my research skills within a well-established group. I initially developed a project exploring the relationship between cognitive appraisals and satisfaction with health and functioning, as a domain of quality of life (QoL), in long-term stroke survivors, based on Leventhal's Self-Regulation model. This was approved by the course and I had subsequently selected validated stroke specific measures, contributed towards the group's NHS ethics application, and started recruitment when the pandemic hit, and I was unable to continue with the project due to lockdowns and restrictions placed on research activity within the Trust. This came with a great sense of sadness as I had spent a significant amount of time and resource developing this project and was passionate about seeing it through. However, uncertainty around the duration of restrictions and concerns regarding resuming data collection with a population classified as 'extremely vulnerable' led me back to the drawing board. From here, I developed another project within this population that could be completed within the remaining duration of the course.

I was incredibly grateful for the flexibility offered to me in developing a new project with pre-existing data. However, this initially posed challenges in coming up with a novel project grounded in theory. Thankfully, I began my specialist placement in inpatient neurorehabilitation around this time and came across the Y-shaped process model which led me to explore literature around awareness. I was able to collaboratively develop my present project by consolidating this learning through discussions with Clinical Neuropsychologists on placement and on the course team. My experiences in completing this project have led to a newfound appreciation of the flexibility and resilience required to push forwards when faced with a multitude of other competing demands on your time, particularly during a pandemic.

SIP

My SIP was also impacted by the pandemic due to restrictions placed on all research activity within the Trust. As I had gone through a very long and iterative process of ethics and recruited over 50% of participants by March 2020, I was determined to find a way to complete this project, rather than having to start two afresh. Moreover, I felt that I really owed it to the young people who had already given up their time to take part. Thankfully, it was easy to move participant interviews online and I was given permission to restart recruitment 6-months after the restrictions were first introduced.

At the start of training, I did not intend to carry out research in paediatrics or coeliac disease but had a longstanding interest in health psychology. Following the research fayre held by the course and teaching on the topic of stigma, I contacted my external supervisor who had previously conducted stigma research in paediatric gastroenterology. During this project's consultation process, I became curious about the views held by some medical professionals regarding coeliac disease, being perhaps 'easier' to live with than other gastrointestinal conditions and their reasoning that those with coeliac disease are therefore less vulnerable to poor emotional outcomes. Following a literature search indicating that this was not the case, I felt motivated to develop a project which enabled these young people's voices to be heard. Because of this, I felt it was important to have someone with personal experience support with the development of the aims and interview schedule and found this input invaluable to the project.

I chose to do an entirely qualitative project for my SIP as I felt it would be an ideal opportunity to develop new skills. The process of coming to terms with IPA was daunting but incredibly rewarding. I found that gathering and analysing qualitative data brought meaning to the data in a way that I have not experienced before. Through doing this project, I have noticed that I have become more curious and observant in clinical placements with

regards to opportunities for service improvement and am motivated to pursue future SIP opportunities in qualified roles.

Acknowledgements

I would firstly like to acknowledge and thank those who took the time to take part in my research, without whom this thesis would not be possible. In particular, I would like to thank the inspiring young people who willingly took some of their time to meet with me and share their experiences in my SIP – I hope this research goes a small way towards improving access to psychological services.

I am very grateful to my research supervisors, Dr Matthew Knight, Dr Nigel King, Dr Louise Johns, Professor Nele Demeyere, Dr Owen Williams, Dr Annabel David, and Dr Juliet Kennedy for their advice, enthusiasm, solutions, and words of encouragement over the last three years. I am particularly grateful for their pragmatic and containing support when facing multiple hurdles with projects during the pandemic. I am also grateful to Dr Georgie Boothroyd and Dr Aonghus Ryan for taking the time to think through different ideas with me for my MRP and Elise Milosevich for her valued guidance on conducting a high quality CRL. Thank you too to my Course Tutor, Dr Helen Jenkins, for your approachable, compassionate, and reliable presence over the last three years, and for always reminding me that I was doing well even when I felt like I wasn't.

Thank you to all of the clients I have had the privilege to work with. I have been profoundly influenced by your stories, strength, and commitment. You have been my best teachers and have undoubtably made me a better Psychologist. Thank you too to all of my placement supervisors for sharing your wisdom and skills and the warm and welcoming teams I have had the pleasure of working alongside.

I would like to give a big thank you to my family and friends, who have continuously shown me that there is life outside of training, have encouraged my self-care, supported me through pandemic-related research nightmares, madly celebrated my achievements and validated my experiences when faced with challenges. Thank you for being so

understanding when I have been less available, I am so looking forward to spending more time with you again!

Another big thank you to my 16 fellow ‘Girls of the Oxford DClinPsy’. I am so grateful to have journeyed through the ups and downs of training alongside a group of such intelligent, strong, and passionate women. Thank you for the belly-aching laughs, the baking triumphs, the secret Santa’s, the outstanding musical performances, the appreciation awards, the zoom chats, and for making me feel I was never alone when things got tough. I cannot wait to see the places you’ll go. Thank you especially to Frankie, for (very literally) journeying alongside me and always bringing a fresh perspective to things.

Finally, I would like to thank my partner, Ryan. You did not sign up for what the last three years have entailed but have forever been by my side. Thank you for prising me away from my computer at the weekends, constantly refuting my self-doubt, proof-reading my countless drafts, dancing around the room to my achievements, and holding my hand through the challenges. This thesis marks the end of this chapter of our lives and the start of the next (hopefully) less stressful one. Bring it on!

Appendices

Appendix 1.A

CRL Instructions for Authors

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Appendix 1.B

Principles of the Mental Capacity Act 2005

The five key principles of the Mental Capacity Act 2005:

- Capacity is presumed unless proven otherwise;
- All practical steps to help a person to make a decision must be taken;
- An irrational decision does not equate to the absence of capacity;
- If a person lacks capacity, any decisions made must be in their best interests;
- Any decision for an adult lacking capacity made must be the least restrictive option available for their basic rights and freedoms.

Criteria for determining a lack of capacity:

- They cannot understand information relevant to the decision;
- They cannot retain information for the time required to make the decision;
- They are not able to appreciate the relevance of the information or to use and weigh that information as part of the process of making the decision;
- They are not able to communicate a decision (whether by talking, using sign language or any other means).

Taken from: Department of Health (2005). Mental Capacity Act. London, HMSO. Retrieved from https://www.legislation.gov.uk/ukpga/2005/9

Appendix 1.C

Alternative Definition of Decision-Making Capacity

As identified by Appelbaum and Grisso[7], the term "decisional capacity" is generally defined in the literature in terms of four functional tasks or abilities:

(1) Understanding - the ability to comprehend the information being disclosed relevant to the choice presented;

(2) Appreciation - the ability to apply the information to one's own situation or condition;

(3) Reasoning - the ability to consider and compare potential consequences of the proposed treatment or research protocol relative to alternatives; and

(4) Expression of a choice - the ability to communicate a stable choice.

Appendix 1.D

Search Strategy - Example from PubMed Search

PubMed Search Strategy:			
Population	**Test**	**Cognitive Function**	**Mental Capacity**
"Neurocognitive Disorders"[Mesh] OR Cognitive Disorder* OR Cognitive Impair* OR Cognitive Dysfunction* OR Cognitive Deficit* OR Cognition Disorder* OR Cognition Impair* OR Cognition Dysfunction* OR Cognition Deficit* OR Cognitively Impair* OR Neurocognitive Disorder* OR Neurocognitive Impair* OR Neurocognitive Dysfunction* OR Neurocognitive Deficit* OR *"Brain Diseases"[Mesh]* OR "TBI" OR "ABI" OR "HI" OR "Head Injury" OR "Brain Injury" OR "stroke" OR "Transient ischaemic attack" OR "TIA" OR "CVA" OR "Cerebrovascular accident"	*"Neuropsychological Tests"[Mesh]* OR Neuropsychological Test* OR Neuropsychological Batter* OR Neuropsychological Screen* OR Neuropsychological Evaluat* OR Neuropsychological Score* OR Neuropsychological Measure* OR Neuropsychological Exam* OR Neuropsychological Assess* OR Neuropsychological Perform* OR Neuropsychology Test* OR Neuropsychology Batter* OR Neuropsychology Evaluat* OR Cognitive Test* OR Cognitive Batter* OR Cognitive Screen* OR Cognitive Evaluat* OR Cognitive Score* OR Cognitive Measure* OR Cognitive Exam* OR Cognitive Assess* OR Cognitive Perform* OR Cognition Test* OR Cognition Batter* OR Cognition Screen* OR	*"Comprehension"[Mesh]* OR Comprehen* OR *"Executive Function"[Mesh]* OR Executive function* OR Executive dysfunction* OR "Executive control" OR "Executive performance" OR "Dysexecutive" OR "Cognitive flexibility" OR *"Memory"[Mesh]* OR "Retain" AND memor* OR "Retention" AND memor* OR "Visual memory" OR "Verbal memory" OR "Visuospatial memory" OR "Spatial memory" OR "STM" OR "Short-term memory" OR "Short term	*"Decision Making"[Mesh]* OR "Decision-making ability" OR "Decision making ability" OR "Decision-making capacity" OR "Decision making capacity" OR *"Clinical Decision-Making"[Mesh]* OR "Capacity" AND "consent" OR Decision* AND Autonom* OR "Consent" AND Autonom* OR "Incapacity" AND Decision* OR

OR “Parkinson’s Disease” OR “PD” OR “Motor Neurone Disease” OR “MND” OR “Huntington’s disease” OR “Huntingtons” OR “Epilepsy” OR “Epileptic” OR “hydrocephalus” OR “encephalitis” OR *“Neurodegenerative Diseases”[Mesh]* OR “dementia” OR “Alzheimer’s disease” OR “Vascular dementia” OR “Dementia with lewy bodies” OR “Mixed dementia” OR “Frontotemporal dementia” OR “Posterior cortical atrophy” OR “Cerebral atrophy” OR “mild cognitive impairment” OR “MCI” OR “Primary progressive aphasia” OR *“Memory Disorders”[Mesh]* OR “Korsakoff Syndrome” OR “Amnesia” OR *“Brain Neoplasms”[Mesh]* OR “Brain Tumour” OR “Brain Tumor” OR abscess* AND “brain” OR	Cognition Evaluat* OR Cognition Score* OR Cognition Measure* OR Cognition Exam* OR Cognition Assess* OR Cognition Perform* OR Neurocognitive Test* OR Neurocognitive Batter* OR Neurocognitive Screen* OR Neurocognitive Evaluat* OR Neurocognitive Score* OR Neurocognitive Measure* OR Neurocognitive Exam* OR Neurocognitive Assess* OR Neurocognitive Perform* OR *“Aptitude Tests”[Mesh]* OR *“Psychometrics”[Mesh]* OR “MMSE” OR “Mini-mental state examination” OR CAMDEX OR CAMCOG OR “MOCA” OR “Montreal Cognitive Assessment” OR WAIS OR WMS OR “Wechsler” OR “RBANS” OR “Repeatable battery for the assessment of neuropsychological status” OR “DKEFS” OR “D-KEFS” OR “Delis-Kaplan executive function system” OR “stroop” “Hayling” AND “Brixton” OR	memory” OR “Working memory” OR “LTM” OR “Long-term memory” OR “Long term memory” OR “Retrospective memory” OR *“Problem Solving”[Mesh]* OR “Weighing-up” OR “weighing up” OR “Weigh-up” OR “weigh up” OR *“Spatial Learning”[Mesh]* OR *“Verbal Learning”[Mesh]* OR “Verbal Fluency” OR *“Communication”[Mesh:NoExp]* OR “Communicat*” OR *“Speech Intelligibility”[Mesh]* OR “Aphasia” OR “Processing Speed” OR “Speed of Processing” OR *“Attention”[Mesh:NoExp]* OR “Arithmetic” OR Cognitive Domain* OR Neurocognitive Domain* OR Neuropsychological Domain* OR Cognitive	“Incapacity” AND “Consent” OR “mental capacity” OR Treatment decision* OR Discharge decision* OR “Financial capacity” OR “Driving capacity” OR *“Informed Consent”[Mesh:NoExp]* “Informed consent” OR “consent” AND sexual relationship* OR “capacity” AND sexual relationship* OR *“Mental Competency”[Mesh]* OR Mental competenc* OR Financial competenc* OR “Competency to stand trial” OR “diminished capacity” OR “Power of attorney”

"Multiple Sclerosis"[Mesh] OR "Multiple Sclerosis" OR "MS" AND	"Trail-making test" OR "verbal fluency test" OR "Luria-Nebraska" OR "Halstead-Reitan" AND	function* OR Neurocognitive function* OR *"Orientation"[Mesh]* AND	

Appendix 1.E

Quality Assessment Tool

National Heart, Lung and Blood Institute Study Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies[11]

Criteria	Yes	No	Other (CD, NR, NA)*
1. Was the research question or objective in this paper clearly stated?			
2. Was the study population clearly specified and defined?			
3. Was the participation rate of eligible persons at least 50%?			
4. Were all the subjects selected or recruited from the same or similar populations (including the same time period)? Were inclusion and exclusion criteria for being in the study pre-specified and applied uniformly to all participants?			
5. Was a sample size justification, power description, or variance and effect estimates provided?			
6. For the analyses in this paper, were the exposure(s) of interest measured prior to the outcome(s) being measured?			
7. Was the timeframe sufficient so that one could reasonably expect to see an association between exposure and outcome if it existed?			
8. For exposures that can vary in amount or level, did the study examine different levels of the exposure as related to the outcome (e.g., categories of exposure, or exposure measured as continuous variable)?			
9. Were the exposure measures (independent variables) clearly defined, valid, reliable, and implemented consistently across all study participants?			
10. Was the exposure(s) assessed more than once over time?			
11. Were the outcome measures (dependent variables) clearly defined, valid, reliable, and implemented consistently across all study participants?			
12. Were the outcome assessors blinded to the exposure status of participants?			
13. Was loss to follow-up after baseline 20% or less?			
14. Were key potential confounding variables measured and adjusted statistically for their impact on the relationship between exposure(s) and outcome(s)?			

*CD, cannot determine; NA, not applicable; NR, not reported

Guidance for Assessing the Quality of Observational Cohort and Cross-Sectional Studies

The guidance document below is organized by question number from the tool for quality assessment of observational cohort and cross-sectional studies.

Question 1. Research question

Did the authors describe their goal in conducting this research? Is it easy to understand what they were looking to find? This issue is important for any scientific paper of any type. Higher quality scientific research explicitly defines a research question.

Questions 2 and 3. Study population

Did the authors describe the group of people from which the study participants were selected or recruited, using demographics, location, and time period? If you were to conduct this study again, would you know who to recruit, from where, and from what time period? Is the cohort population free of the outcomes of interest at the time they were recruited?

An example would be men over 40 years old with type 2 diabetes who began seeking medical care at Phoenix Good Samaritan Hospital between January 1, 1990 and December 31, 1994. In this example, the population is clearly described as: (1) who (men over 40 years old with type 2 diabetes); (2) where (Phoenix Good Samaritan Hospital); and (3) when (between January 1, 1990 and December 31, 1994). Another example is women ages 34 to 59 years of age in 1980 who were in the nursing profession and had no known coronary disease, stroke, cancer, hypercholesterolemia, or diabetes, and were recruited from the 11 most populous States, with contact information obtained from State nursing boards.

In cohort studies, it is crucial that the population at baseline is free of the outcome of interest. For example, the nurses' population above would be an appropriate group in which to study incident coronary disease. This information is usually found either in descriptions of population recruitment, definitions of variables, or inclusion/exclusion criteria.

You may need to look at prior papers on methods in order to make the assessment for this question. Those papers are usually in the reference list.

If fewer than 50% of eligible persons participated in the study, then there is concern that the study population does not adequately represent the target population. This increases the risk of bias.

Question 4. Groups recruited from the same population and uniform eligibility criteria

Were the inclusion and exclusion criteria developed prior to recruitment or selection of the study population? Were the same underlying criteria used for all of the subjects involved? This issue is related to the description of the study population, above, and you may find the information for both of these questions in the same section of the paper.

Most cohort studies begin with the selection of the cohort; participants in this cohort are then measured or evaluated to determine their exposure status. However, some cohort studies may recruit or select exposed participants in a different time or place than unexposed participants, especially retrospective cohort studies–which is when data are obtained from the past (retrospectively), but the analysis examines exposures prior to outcomes. For example, one research question could be whether diabetic men with clinical depression are at higher risk for cardiovascular disease than those without clinical depression. So, diabetic men with depression might be selected from a mental health clinic, while diabetic men without depression might be selected from an internal medicine or endocrinology clinic.

This study recruits groups from different clinic populations, so this example would get a "no."

However, the women nurses described in the question above were selected based on the same inclusion/exclusion criteria, so that example would get a "yes."

Question 5. Sample size justification

Did the authors present their reasons for selecting or recruiting the number of people included or analyzed? Do they note or discuss the statistical power of the study? This question is about whether or not the study had enough participants to detect an association if one truly existed.

A paragraph in the methods section of the article may explain the sample size needed to detect a hypothesized difference in outcomes. You may also find a discussion of power in the discussion section (such as the study had 85 percent power to detect a 20 percent increase in the rate of an outcome of interest, with a 2-sided alpha of 0.05). Sometimes estimates of variance and/or estimates of effect size are given, instead of sample size calculations. In any of these cases, the answer would be "yes."

However, observational cohort studies often do not report anything about power or sample sizes because the analyses are exploratory in nature. In this case, the answer would be "no." This is not a "fatal flaw." It just may indicate that attention was not paid to whether the study was sufficiently sized to answer a prespecified question–i.e., it may have been an exploratory, hypothesis-generating study.

Question 6. Exposure assessed prior to outcome measurement

This question is important because, in order to determine whether an exposure causes an outcome, the exposure must come before the outcome.

For some prospective cohort studies, the investigator enrolls the cohort and then determines the exposure status of various members of the cohort (large epidemiological studies like Framingham used this approach). However, for other cohort studies, the cohort is selected based on its exposure status, as in the example above of depressed diabetic men (the exposure being depression). Other examples include a cohort identified by its exposure to fluoridated drinking water and then compared to a cohort living in an area without fluoridated water, or a cohort of military personnel exposed to combat in the Gulf War compared to a cohort of military personnel not deployed in a combat zone.

With either of these types of cohort studies, the cohort is followed forward in time (i.e., prospectively) to assess the outcomes that occurred in the exposed members compared to nonexposed members of the cohort. Therefore, you begin the study in the present by looking at groups that were exposed (or not) to some biological or behavioral factor, intervention, etc., and then you follow them forward in time to examine outcomes. If a cohort study is conducted properly, the answer to this question should be "yes," since the exposure status of members of the cohort was determined at the beginning of the study before the outcomes occurred.

For retrospective cohort studies, the same principal applies. The difference is that, rather than identifying a cohort in the present and following them forward in time, the investigators go back in time (i.e., retrospectively) and select a cohort based on their exposure status in the past and then follow them forward to assess the outcomes that occurred in the exposed

and nonexposed cohort members. Because in retrospective cohort studies the exposure and outcomes may have already occurred (it depends on how long they follow the cohort), it is important to make sure that the exposure preceded the outcome.

Sometimes cross-sectional studies are conducted (or cross-sectional analyses of cohort-study data), where the exposures and outcomes are measured during the same timeframe. As a result, cross-sectional analyses provide weaker evidence than regular cohort studies regarding a potential causal relationship between exposures and outcomes. For cross-sectional analyses, the answer to Question 6 should be "no."

Question 7. Sufficient timeframe to see an effect

Did the study allow enough time for a sufficient number of outcomes to occur or be observed, or enough time for an exposure to have a biological effect on an outcome? In the examples given above, if clinical depression has a biological effect on increasing risk for CVD, such an effect may take years. In the other example, if higher dietary sodium increases BP, a short timeframe may be sufficient to assess its association with BP, but a longer timeframe would be needed to examine its association with heart attacks.

The issue of timeframe is important to enable meaningful analysis of the relationships between exposures and outcomes to be conducted. This often requires at least several years, especially when looking at health outcomes, but it depends on the research question and outcomes being examined.

Cross-sectional analyses allow no time to see an effect, since the exposures and outcomes are assessed at the same time, so those would get a "no" response.

Question 8. Different levels of the exposure of interest

If the exposure can be defined as a range (examples: drug dosage, amount of physical activity, amount of sodium consumed), were multiple categories of that exposure assessed? (for example, for drugs: not on the medication, on a low dose, medium dose, high dose; for dietary sodium, higher than average U.S. consumption, lower than recommended consumption, between the two). Sometimes discrete categories of exposure are not used, but instead exposures are measured as continuous variables (for example, mg/day of dietary sodium or BP values).

In any case, studying different levels of exposure (where possible) enables investigators to assess trends or dose-response relationships between exposures and outcomes–e.g., the higher the exposure, the greater the rate of the health outcome. The presence of trends or dose-response relationships lends credibility to the hypothesis of causality between exposure and outcome.

For some exposures, however, this question may not be applicable (e.g., the exposure may be a dichotomous variable like living in a rural setting versus an urban setting, or vaccinated/not vaccinated with a one-time vaccine). If there are only two possible exposures (yes/no), then this question should be given an "NA," and it should not count negatively towards the quality rating.

Question 9. Exposure measures and assessment

Were the exposure measures defined in detail? Were the tools or methods used to measure exposure accurate and reliable–for example, have they been validated or are they objective?

This issue is important as it influences confidence in the reported exposures. When exposures are measured with less accuracy or validity, it is harder to see an association between exposure and outcome even if one exists. Also as important is whether the exposures were assessed in the same manner within groups and between groups; if not, bias may result.

For example, retrospective self-report of dietary salt intake is not as valid and reliable as prospectively using a standardized dietary log plus testing participants' urine for sodium content. Another example is measurement of BP, where there may be quite a difference between usual care, where clinicians measure BP however it is done in their practice setting (which can vary considerably), and use of trained BP assessors using standardized equipment (e.g., the same BP device which has been tested and calibrated) and a standardized protocol (e.g., patient is seated for 5 minutes with feet flat on the floor, BP is taken twice in each arm, and all four measurements are averaged). In each of these cases, the former would get a "no" and the latter a "yes."

Here is a final example that illustrates the point about why it is important to assess exposures consistently across all groups: If people with higher BP (exposed cohort) are seen by their providers more frequently than those without elevated BP (nonexposed group), it also increases the chances of detecting and documenting changes in health outcomes, including CVD-related events. Therefore, it may lead to the conclusion that higher BP leads to more CVD events. This may be true, but it could also be due to the fact that the subjects with higher BP were seen more often; thus, more CVD-related events were detected and documented simply because they had more encounters with the health care system. Thus, it could bias the results and lead to an erroneous conclusion.

Question 10. Repeated exposure assessment

Was the exposure for each person measured more than once during the course of the study period? Multiple measurements with the same result increase our confidence that the exposure status was correctly classified. Also, multiple measurements enable investigators to look at changes in exposure over time, for example, people who ate high dietary sodium throughout the followup period, compared to those who started out high then reduced their intake, compared to those who ate low sodium throughout. Once again, this may not be applicable in all cases. In many older studies, exposure was measured only at baseline. However, multiple exposure measurements do result in a stronger study design.

Question 11. Outcome measures

Were the outcomes defined in detail? Were the tools or methods for measuring outcomes accurate and reliable–for example, have they been validated or are they objective? This issue is important because it influences confidence in the validity of study results. Also important is whether the outcomes were assessed in the same manner within groups and between groups.

An example of an outcome measure that is objective, accurate, and reliable is death–the outcome measured with more accuracy than any other. But even with a measure as objective as death, there can be differences in the accuracy and reliability of how death was assessed by the investigators. Did they base it on an autopsy report, death certificate, death registry, or report from a family member? Another example is a study of whether dietary fat intake is related to blood cholesterol level (cholesterol level being the outcome), and the cholesterol level is measured from fasting blood samples that are all sent to the same laboratory. These examples would get a "yes." An example of a "no" would be self-report by subjects that they

had a heart attack, or self-report of how much they weigh (if body weight is the outcome of interest).

Similar to the example in Question 9, results may be biased if one group (e.g., people with high BP) is seen more frequently than another group (people with normal BP) because more frequent encounters with the health care system increases the chances of outcomes being detected and documented.

Question 12. Blinding of outcome assessors

Blinding means that outcome assessors did not know whether the participant was exposed or unexposed. It is also sometimes called "masking." The objective is to look for evidence in the article that the person(s) assessing the outcome(s) for the study (for example, examining medical records to determine the outcomes that occurred in the exposed and comparison groups) is masked to the exposure status of the participant. Sometimes the person measuring the exposure is the same person conducting the outcome assessment. In this case, the outcome assessor would most likely not be blinded to exposure status because they also took measurements of exposures. If so, make a note of that in the comments section.

As you assess this criterion, think about whether it is likely that the person(s) doing the outcome assessment would know (or be able to figure out) the exposure status of the study participants. If the answer is no, then blinding is adequate. An example of adequate blinding of the outcome assessors is to create a separate committee, whose members were not involved in the care of the patient and had no information about the study participants' exposure status. The committee would then be provided with copies of participants' medical records, which had been stripped of any potential exposure information or personally identifiable information. The committee would then review the records for prespecified outcomes according to the study protocol. If blinding was not possible, which is sometimes the case, mark "NA" and explain the potential for bias.

Question 13. Followup rate

Higher overall followup rates are always better than lower followup rates, even though higher rates are expected in shorter studies, whereas lower overall followup rates are often seen in studies of longer duration. Usually, an acceptable overall followup rate is considered 80 percent or more of participants whose exposures were measured at baseline. However, this is just a general guideline. For example, a 6-month cohort study examining the relationship between dietary sodium intake and BP level may have over 90 percent followup, but a 20-year cohort study examining effects of sodium intake on stroke may have only a 65 percent followup rate.

Question 14. Statistical analyses

Were key potential confounding variables measured and adjusted for, such as by statistical adjustment for baseline differences? Logistic regression or other regression methods are often used to account for the influence of variables not of interest.

This is a key issue in cohort studies, because statistical analyses need to control for potential confounders, in contrast to an RCT, where the randomization process controls for potential confounders. All key factors that may be associated both with the exposure of interest and the outcome–that are not of interest to the research question–should be controlled for in the analyses.

For example, in a study of the relationship between cardiorespiratory fitness and CVD events (heart attacks and strokes), the study should control for age, BP, blood cholesterol, and body weight, because all of these factors are associated both with low fitness and with CVD events. Well-done cohort studies control for multiple potential confounders.

Some general guidance for determining the overall quality rating of observational cohort and cross-sectional studies

The questions on the form are designed to help you focus on the key concepts for evaluating the internal validity of a study. They are not intended to create a list that you simply tally up to arrive at a summary judgment of quality.

Internal validity for cohort studies is the extent to which the results reported in the study can truly be attributed to the exposure being evaluated and not to flaws in the design or conduct of the study–in other words, the ability of the study to draw associative conclusions about the effects of the exposures being studied on outcomes. Any such flaws can increase the risk of bias.

Critical appraisal involves considering the risk of potential for selection bias, information bias, measurement bias, or confounding (the mixture of exposures that one cannot tease out from each other). Examples of confounding include co-interventions, differences at baseline in patient characteristics, and other issues throughout the questions above. High risk of bias translates to a rating of poor quality. Low risk of bias translates to a rating of good quality. (Thus, the greater the risk of bias, the lower the quality rating of the study.)

In addition, the more attention in the study design to issues that can help determine whether there is a causal relationship between the exposure and outcome, the higher quality the study. These include exposures occurring prior to outcomes, evaluation of a dose-response gradient, accuracy of measurement of both exposure and outcome, sufficient timeframe to see an effect, and appropriate control for confounding–all concepts reflected in the tool.

Generally, when you evaluate a study, you will not see a "fatal flaw," but you will find some risk of bias. By focusing on the concepts underlying the questions in the quality assessment tool, you should ask yourself about the potential for bias in the study you are critically appraising. For any box where you check "no" you should ask, "What is the potential risk of bias resulting from this flaw in study design or execution?" That is, does this factor cause you to doubt the results that are reported in the study or doubt the ability of the study to accurately assess an association between exposure and outcome?

The best approach is to think about the questions in the tool and how each one tells you something about the potential for bias in a study. The more you familiarize yourself with the key concepts, the more comfortable you will be with critical appraisal. Examples of studies rated good, fair, and poor are useful, but each study must be assessed on its own based on the details that are reported and consideration of the concepts for minimizing bias.

Appendix 1.F

Data Extraction Tool

General Information		
Citation		
Location		
Study Details		
Study Intention	Aims of study	
	Was the primary aim related to the aim of this review?	
Methods		
Participants	Number of Participants	
	Age	
	Gender	
	Ethnicity	
	Years of Education	
	Neuro condition, reported severity and duration	
	Diagnostic criteria used	
	Co-morbidity	
	Control Group (Y/N)	
	Characteristics of control group	
Recruitment	Setting (e.g. inpatient/community)	
	Participant uptake for all who were eligible to take part	
	Inclusion criteria	
	Exclusion criteria	
	Was the study population clearly specified and defined?	
	How representative is the sample?	
Measures and outcomes	What is the capacity decision assessed?	
	What capacity tool was used?	
	Is this tool validated (and used as validated?)	
	What cognitive tests were used?	
	Was the researcher carrying out the tests blind to capacity status/ vice-versa?	
	Was capacity assessment carried out more than once or inter-rated?	
	Did the researchers look at different standards of capacity (e.g. ‘understanding’, ‘communication of a decision’)?	
Results	Data analysis	

	Was a significant association found between capacity and cognition (Y/N)?	
	Was the power reported? If so, was it adequate?	
	Were confounding variables considered? Which ones?	
	Key results	
Other relevant information	Author's conclusions	
	Additional notes	

Appendix 1.G

Capacity Tools and Assessments Used in Included Studies

Table A1

Capacity Assessments used in Studies Aligning to the Definition of Capacity set out by Appelbaum and Grisso[7]

Tool	Descriptor	Details
		Treatment Decisions
CCTI[48]	Description	A standardized psychometric instrument designed to assess the treatment consent capacity of adults. Involves presenting the individual with two clinical vignettes (a neoplasm condition and a cardiac condition). Each vignette presents a hypothetical medical problem and symptoms and two treatment alternatives with associated risks and benefits. Vignettes are presented orally and in writing at a "5th - 6th grade reading level". After reading and hearing the vignettes, the individual answers questions designed to test competency under five legal standards[7].
	Inter-rater reliability	$r = 0.83$ for interval scales; and $r > 0.96$ for categorical scales[49].

Definition of 'Understanding'	Assesses the capacity to understand the treatment situation and choices.
Definition of 'Appreciation'	Assesses the capacity to appreciate the emotional and cognitive consequences of a treatment choice (this standard emphasizes the individual's awareness of the consequences of a treatment decision, i.e., its emotional impact, rational requirements, and future consequences).
Definition of 'Reasoning'	Assesses the capacity to provide rational reasons for a treatment choice (this standard tests the capacity to use logical processes to compare the benefits and risks of various treatment options and weigh this information to reach a decision). This measure also has a separate 'reasoning' standard which assesses the capacity to make the reasonable treatment choice. This standard emphasizes outcome rather than the mere fact of a decision or how it has been reached (the individual who fails to make a decision that is roughly congruent with the decision that a reasonable person in like circumstances would make is viewed as incompetent). This standard is not used to calculate overall capacity.

	Definition of 'Communication of choice'	Assesses the capacity simply to evidence a treatment choice (this standard focuses on the presence or absence of a decision, and not on the quality of the decision).
	Scoring/Determination of capacity outcome	'Capacity performance' is the quantitative score received for each legal standard (understanding, appreciation, reasoning, communication of choice). Scores are summed across vignettes to create a composite score for each standard. There is no CCTI total score. Capacity status is also calculated (capable, marginally capable, or incapable of consenting to treatment) for each standard. Capacity status is operationalised using either predetermined cut-off scores or psychometric cut-off scores derived from a cognitively intact control group.
Mac-CAT-T[13]	Description	An instrument designed to assess decision-making capacity. Utilizes a semi-structured interview to guide the clinician through an assessment of capacity to make an actual treatment decision. After disclosing information, individuals are asked to repeat what they have been told in their own words.
	Inter-rater reliability	$r > 0.87$[22]

Definition of 'Understanding'	Assesses comprehension of the nature of their illness, recommended treatment, alternatives, and risks and benefits.
Definition of 'Appreciation'	Assesses acknowledgment of illness and the potential value of treatment.
Definition of 'Reasoning'	Evaluates an individual's problem-solving ability when faced with a specific treatment choice.
Definition of 'Communication of choice'	Determines whether an individual can make a selection consistent with their reasoning.
Scoring/Determination of capacity outcome	Each dimension yields respective summary score. 'Understanding' is measured by 13 items. 'Appreciation' is measured by two items, 'Reasoning' is assessed by eight items, 'Communicating a choice' is assessed using one item. No Mac-CAT-T total score is calculated because significant enough deficits on even one dimension may result in incompetence, even when performance on other dimensions are intact. Scores must be considered along with other information, such as the clinical diagnostic assessment and other relevant factors. It does not provide a final assessment of capacity to consent to treatment. However, some cut-off scores have been given[129]. Namely, capacity to consent is

		given if an individual scores >4 in 'Understanding', >0 in 'Appreciation, >5 in 'Reasoning' and >0 in 'Communicating a decision'.
Chinese version of the ACED[50]	Description	Developed for the assessment of everyday decision-making in a Western population of older adults with very mild to moderate late-life cognitive impairments and translated into Chinese with back-translation. The Chinese ACED, like the original, has three versions for different activities of daily living. These include preparing meals, managing medications, and managing money. The interviewer collects information from caregivers regarding functional deficits to allow the ACED instrument content to be tailored to each patient. For standardized content, participants receive a copy of information about the functional problem, the options to address it, and the risks/benefits of these options, to minimize the influence of short-term memory. The interviewer uses the structured format of the ACED to discuss a patient's everyday medication management and the possible solutions to managing their problems and has a standardised rating criteria for subjects' responses.
	Inter-rater reliability	$r = 0.65 – 0.72$[142]

	Definition of 'Understanding'	Assesses the ability to comprehend basic information about a problem, its potential solutions, and the risks and benefits associated with those solutions.
	Definition of 'Appreciation'	Assesses the ability of a person to recognize how a problem or solution pertains to his or her specific situation.
	Definition of 'Reasoning'	Assesses the ability to process the information in a logical fashion toward a decision.
	Definition of 'Communication of choice'	Assesses ability to communicate a choice for the decision.
	Scoring/Determination of capacity outcome	The interviewer scores responses using a three- point scale: 0 = inadequate, 1 = marginal, or 2 = adequate. Scores for each standard are totalled. Higher scores indicate better performance: understanding (0–10), appreciation (0–8), reasoning (0–10), and expressing a choice (0–2). An ACED total score is not calculated, as significant deficits on any single dimension may result in mental incompetence, even when performance on other dimensions is intact.
HCAT[51]	Description	A brief screening measure for assessing a patient's capacity to provide informed consent or prepare advance directives regarding medical treatments. Consists of a short description

	(essay) of the informed consent process and the durable power of attorney, followed by six questions. These questions evaluate the patient's comprehension of the information disclosed. Versions of the essay at the "13th-grade, eighth-grade, and sixth-grade reading levels".
Inter-rater reliability	$r = 0.05$[51]
Definition of 'Understanding'	Assess comprehension of information disclosed (e.g., What are four things a doctor must tell a patient before beginning a procedure?).
Definition of 'Appreciation'	N/A
Definition of 'Reasoning'	N/A
Definition of 'Communication of choice'	N/A
Scoring/Determination of capacity outcome	The questionnaire includes true/false and sentence-completion questions, some of which have more than one part. Individual's score 1 point for each correct answer. Possible scores on the questionnaire range from 0 to 10, with scores of 3 or lower signifying inadequate comprehension.

HCAI[143]	Description	A tool to assess medical decision making. Consists of two clinical vignettes (treatment of an eye infection and administration of cardiopulmonary resuscitation) that are presented after general concepts of choice, risk, and benefit have been reviewed. Probe questions are then asked to score each decisional ability (understanding, reasoning, appreciation and expression of choice).
	Inter-rater reliability	Pilot data (n = 17): r = 0.93[22]
	Definition of 'Understanding'	Assesses understanding of relevant information.
	Definition of 'Appreciation'	Assesses appreciation of consequences of alternatives.
	Definition of 'Reasoning'	Assesses reasoning or weighing or costs and benefits.
	Definition of 'Communication of choice'	Assesses an individual's ability to make a choice.
	Scoring/Determination of capacity outcome	Items are scored 0, 1, or 2 using a detailed scoring manual. There is no specific guidance provided for mapping scores onto dichotomous ratings, and clinicians must incorporate scores into a broader assessment that considers each patient separately. However, in most

		research studies, researcher use statistically defined thresholds e.g., 'impaired' capacity is defined as 2.5 standard deviations below the healthy control groups mean performance.
Protocol to assess treatment capacity based on legal standards set out by Appelbaum and Grisso[7]	Description	Marson et al[36] developed two specialized clinical vignettes (A- neoplasm and B-cardiac) designed to test the competency of AD patients. Each vignette presents a hypothetical medical problem and symptoms and two treatment alternatives with associated risks and benefits. The vignettes were written at a "fifth- to sixth-grade" reading level. The vignettes are simultaneously presented orally and in written form to subjects in an uninterrupted disclosure format. Subjects then answer questions designed to test competency under the legal standards set out by Appelbaum and Grisso[7].
	Inter-rater reliability	NR
	Definition of 'Understanding'	Assesses memory for words, phrases, ideas, sequences of information, and comprehension of the fundamental meaning of information about treatment. Requires broad comprehension of a vignette's medical problem, related symptoms, and treatment choices and respective risks and benefits.

	Definition of 'Appreciation'	Assesses awareness of a treatment decision: its emotional impact, rational requirements, and future consequence. This requires the individual to go beyond the vignette information about a medical problem and treatment choices and to "appreciate," or identify, three types of consequences of their treatment decisions: personal emotional consequences, short-term practical consequences, and longer term (1 year) life consequences.
	Definition of 'Reasoning'	N/A
	Definition of 'Communication of choice'	Assesses the presence or absence of a decision and not on the quality or reasoning behind the decision.
	Scoring/Determination of capacity outcome	Subject scores on the three LSs are summed across vignettes to create composite legal standard variables. All individuals are assigned a competency status on LS3 and LS5 (competent, marginally competent, incompetent) based on cut-off scores for marginal competency statistically derived from control performance.
Unstandardised BRIMMS protocol and	Description	The BRIMMS protocol was designed by Reen and Langdon, and was based on previous work on optimal methods to present treatment risks and benefits to MS patients[144], as well as a comprehensive review of the literature. Specifically, all quantitative probabilities of

'consultation as usual'[14]	risks and benefits were presented using N-in-NxX frequencies (e.g. 25 in 1000, 450 in 1000). Horizontal bar charts with values presented quantitative information. Only the bar charts were shown visually to patients in the BRIMMS aural consultation style. Treatment benefits of the treatment group versus a placebo group from a hypothetical clinical trial was presented using absolute numbers and with baseline information included (i.e. the total number of people with a treatment benefit in both the treatment and placebo group). Consultation: treatment information was provided as usual in a clinical consultation, developed in collaboration with MS nursing staff on two NHS sites and based on treatment information in existing educational booklets provided to MS patients in the UK. Treatment risks and benefits were presented using verbal terms and 1-in-X frequencies. Treatment benefits between the treatment and placebo group from a hypothetical clinical trial was presented using a relative format. No visual graphs were provided with the quantitative information. there were four possible consultation styles in this study: BRIMMS aural, BRIMMS visual, usual consultation aural, usual consultation visual. Each style of consultation took

	approximately 20 minutes to administer. All four consultation styles were presented to patients in one session, with 15 minute breaks provided between each new consultation style.
Inter-rater reliability	NR
Definition of 'Understanding'	Patients' understanding of treatment information was assessed using comprehension questions immediately after presenting a treatment risk or benefit. Patients had to state the total number of people who would experience the minor risk, adverse risk or benefits of the hypothetical treatment across the three time periods (1 year/2 years/5 years). For treatment benefits, patients were also asked to report the differences in the treatment group and placebo group from a hypothetical clinical trial, using a multiple-choice format. Therefore, for each drug, patients were assessed on six questions for benefits, three questions each for the two minor risks, and three questions for an adverse risk of taking the treatment (i.e., a total of 15 questions per drug or a total of 30 questions per treatment set).
Definition of 'Appreciation'	N/A
Definition of 'Reasoning'	N/A

	Definition of 'Communication of choice'	N/A
	Scoring/Determination of capacity outcome	Answers were marked as correct if patients stated the precise value or a value within 10%. Incorrect or missing answers were scored as 0. Understanding was measured continuously (range 0-30). Treatment understanding scores were collapsed across aural and visual forms of each consultation style. That is, understanding scores for BRIMMS and usual consultation were computed in the correlation.
	Research Decisions	
Mac-CAT-CR[52]	Description	A semi-structured assessment format for evaluating abilities related to the decisional capacity of subjects in clinical research. Four component abilities of a decisional capacity standard are assessed: understanding, appreciation, reasoning, and choice. Administration of the instrument begins with the disclosure of selected information about a given research project, on the basis of which these abilities are measured. Following disclosure of discrete informational elements, subjects' understanding is queried by asking them to paraphrase the disclosure about specific items.

Inter-rater reliability	$r = 0.87$[145]
Definition of 'Understanding'	Individuals, at a minimum, must have the ability to understand the basic elements of disclosure including the purpose of the research project, the procedures involved, material risks, possible benefits, and alternatives to research participation.
Definition of 'Appreciation'	Individuals must apply the disclosed information to their own situations, including their recognition that the research project is aimed at generalizable knowledge, not at optimizing the treatment they receive, and that they are truly free to decline to participate without penalty.
Definition of 'Reasoning'	Individuals must be able to manipulate the information disclosed to them, comparing, and weighing the consequences of the alternatives before them.
Definition of 'Communication of choice'	Individuals must select the desired option and to sustain a consistent decision.
Scoring/Determination of capacity outcome	Responses to the Mac-CAT–CR items are scored in a structured fashion, with scores aggregated on scales for each of the four abilities being assessed. 'Understanding' has a total score of 26, 'Appreciation' a total score of six, 'Reasoning' a total score of eight and

'Expression of choice' a total score of two. Each item is scored 0-2 and higher scores indicate better performance. However, composite scores across all four scales are not generated, because poor performance on any one of the relevant abilities may indicate significantly impaired capacity, rendering a composite score meaningless. Nor does the Mac-CAT–CR provide a fixed cut-off above which subjects are deemed competent to consent. The degree of capacity required for participation in a given study should reflect the characteristics of that study, including its complexity and risks. Moreover, the capacity demanded of subjects at risk for decisional impairment (e.g., persons with mental illnesses, developmental disabilities, dementia, and serious medical disorders) ought not to exceed the abilities demonstrated by the general population. Finally, decisions about capacity should take into account clinical data that may not be reflected in assessment scores. Although investigators or institutional review boards can set Mac-CAT–CR cut-offs for particular studies, the instrument is designed to be used as an aid to clinical judgment, with investigators retaining the discretion to determine whether subjects are capable of consenting.

CCRI[46]	Description	A structured, hypothetical, vignette-based interview designed to evaluate an individual's ability to consent to research participation. Consent standards are derived from four legal standards described by Appelbaum and Grisso[7]. The participant assumes the role of a potential research candidate and is asked to consider his or her involvement in a hypothetical drug trial for the treatment of memory loss. The CCRI includes a written consent document modelled on routine informed consenting procedures used in human participant research protocols. Specifically, the informed consent document includes (a) the purpose and plan of study, (b) description of study procedures, (c) study evaluations, (d) early treatment discontinuation, (e) risks, (f) benefits, (g) alternative medications and treatments, (h) new research findings, (i) confidentiality, (j) payment for participation, (k) payment for research related injuries, (l) voluntary participation, and (m) other information. The consent document is provided in writing and reviewed orally with the participant. After the simulated consenting process, the participant responds verbally to a series of 36 questions designed to evaluate understanding and comprehension of the informed consent

	material. The participant is allowed to refer to the written consent form when responding to questions.
Inter-rater reliability	The CCRI shows high inter-rater reliability across all four standards[46].
Definition of 'Understanding'	Assesses ability to demonstrate understanding/comprehension of the information presented in the simulated consenting procedure, its application to the participant, its purpose and goals, procedures and risks and benefits.
Definition of 'Appreciation'	Assesses ability to appreciate the personal consequences associated with that choice including: 1. Short-term practical consequences and 2. Long-term life consequences.
Definition of 'Reasoning'	Assesses ability to provide reasons why he/she would/would not participate in the hypothetical study, based on the information presented in the consent form.
Definition of 'Communication of choice'	Assesses ability to express a choice to participate or not participate (quality of decisional process not considered here).
Scoring/Determination of capacity outcome	Participant responses are recorded in writing and scored by the examiner according to predefined criteria (item responses are scored as 0, 1, or 2 points). Points are totalled for each of the four consenting standards. A total of 91 points can be awarded on the

		'Understanding' standard, a maximum of four points can be awarded on the 'Appreciation' standard, a maximum of six points can be awarded on the 'Reasoning' standard and 'Communication of choice' has a total score of one (yes/no). Capacity classification (no impairment, mild/moderate impairment, severe impairment) can be determined using psychometric cut-off scores derived from a control group. e.g., a 'no impairment rating' can be defined as a score higher than 1.5 SD below the control group mean, a mild/moderate impairment rating can be defined as a score 1.5-2.5 SD below the control group mean, and a severe rating can be defined as a score lower than 2.5 SD below the control group mean.
ICQ (unstandardized)[21]	Description	Guarino et al.[21] developed a 17-item unstandardized capacity assessment instrument that focused on understanding of factual information about an AD clinical trial and the patient's perceived understanding (participant's subjective beliefs about understanding). The assessment is not designed to address other important components of capacity such as appreciation of the significance of the decision, reasoning, or expression of choice. During administration, authors described the study, its purpose and procedures, and the risks and benefits of participation. Patients and caregivers were given the opportunity to take home

	the informed consent document and ask questions. Patients were subsequently asked to complete a questionnaire without the support of their caregivers. Questionnaire responses were reviewed with the patient, and all items answered incorrectly were clarified. After this, the questionnaire was re-administered.
Inter-rater reliability	NR
Definition of 'Understanding'	The first 14 items of the ICQ test 'understanding of the important elements of the study', including potential benefits and risks. Items 15-17 assessed participants' 'perceived understanding' and whether they felt the benefits and risks were adequately explained (items-15-17 are not included in the overall assessment of capacity).
Definition of 'Appreciation'	N/A
Definition of 'Reasoning'	N/A
Definition of 'Communication of choice'	N/A
Scoring/Determination of capacity outcome	If the participant was able to adequately understand the study on the first or second administration and/or the investigator believed that the participant had adequate decisional

		capacity and was making a voluntary decision, they were judged to have capacity to consent. To be judged to have capacity as measured by the ICQ, the participant had to answer 70% of questions 1-10 and 100% of questions 11-14 correctly on the first or second administration. Whilst the ICQ was used as a tool to help determine capacity, the final decision as ultimately based on the primary site investigator's clinical judgement.
Unstandardized consent form[35]	Description	The consent form was extracted from a real text of consent used in a research protocol regarding genetics (unreferenced). The consent form was simplified with respect to the original and its lexico-syntactic readability was improved by reducing the length of the words and the sentences used. Eight multiple-choice questions with three alternative responses (one correct) were administered. Ambiguous alternatives were avoided. Participants were asked to read each text with the help of an examiner when necessary. Immediately after, they addressed the questions. People were allowed to consult the text any time they felt it was necessary.
	Inter-rater reliability	NR

Definition of 'Understanding'	Ability of the participant to select correct multiple-choice options in response to questions pertaining to the content of the consent form.
Definition of 'Appreciation'	N/A
Definition of 'Reasoning'	N/A
Definition of 'Communication of choice'	N/A
Scoring/Determination of capacity outcome	One point was assigned for each correct response (range score, 0–8). To calculate the percentage of patients who failed to comprehend the consent form, a receiver operating characteristic (ROC) curve analysis was computed for each text. The scores with the best sensitivity (true positive rate) and specificity (100, false-positive rate) combination were selected as cut-offs for capacity. A cut-off of 6.5 was chosen, with 79% sensitivity and 83% specificity (area under the curve = 88%).

Note. NR = Not Reported; N/A = Not Applicable; CCTI: the Capacity to Consent to Treatment Instrument[48]; Mac-CAT-T: the MacArthur Competence Assessment Tool for Treatment[13]; Chinese version of the ACED: the Assessment for Capacity for Everyday Decision-Making[50]; HCAT: the Hopkins Competency Assessment Test[51]; HCAI: Hopemont Capacity Assessment Interview[143]; Mac-CAT-CR: the MacArthur Competence Assessment Tool-Research[52]; CCRI: Capacity to Consent to Research Instrument[46]; ICQ: Informed Consent Questionnaire[21]

Table A2

Capacity Assessments used in Studies Aligning to the MCA[1]

Tool	Descriptor	Details
		Treatment Decisions
Capacity to consent to research protocol based on MCA[1,33]	Description	Warner et al.[33] designed a consenting procedure based on the principles of the MCA[1] to assess whether the individual meets criteria for capacity to provide informed consent for research. involved providing sufficiently detailed, salient written and verbal information to potential participants in a form they should be able to understand, including (a) the objectives of the trial, (b) potential risks and inconveniences of participation, (c) the product (Ginkgo) being tested and the possibility of receiving an inert placebo, (d) the concept of randomisation, (e) length of the trial and outcomes measured (f) the opportunity to withdraw at any time. Depending on the ability of the participant to assimilate information, the researcher repeated this information as necessary and at times requested the participant's carer to help impart information. Researchers allowed sufficient time for the individual to understand and retain this information.
	Inter-rater reliability	NR

	Definition of 'Understanding'	Assessed by asking the participant questions about the information to demonstrate their understanding of the information.
	Definition of 'Retention'	Assessed by asking the individual to repeat relevant information.
	Definition of ability to 'use and weigh information'	Assessed by exploring whether the potential participant was able to weigh this information in the balance.
	Definition of 'Communication of choice'	Assessing the ability of the potential participant to communicate their choice, without coercion.
	Scoring/Determination of capacity outcome	Researchers made a judgement about the potential participants' ability to acquit themselves at each stage of the consent process. If a participant fulfilled all four stages he/she was deemed to have met the criteria for informed consent. If a potential participant failed at any stage, they judged them to lack ability to give informed consent.
LIMD[49]	Description	A Swedish quantitative research instrument based on the vignette method. It consists of three vignettes about hypothetical clinical trials, an interview, and a scoring protocol. Three so-called

	criteria of decision-making capacity are evaluated in LIMD: (1) comprehension (understanding the content); (2) evaluation (ability of evaluating risk and benefit); and (3) intelligibility (ability to express a decision). The rationale for the design of LIMD was to perform a detailed and standardized examination of utterances and discourse and avoid interpretations. Responses are sometimes ambiguous or unexpected due to personal attitude and ethical considerations. The LIMD score is based on the evaluator's observations of predefined linguistic features for each of the three established aspects.
Inter-rater reliability	$r = 0.89$[49]
Definition of 'Understanding'	Assesses 'comprehension' (understanding the content of the vignettes). Linguistic features like independence of utterances (i.e., not word-for-word replays), accuracy, inferences, relevance (if utterances were adding new information or not), coherence, specificity and creativity of vocabulary are scored.
Definition of 'Retention'	Elements of retention, although not specified by the measure, are scored within 'comprehension'.

Definition of ability to 'use and weigh information'	Assesses 'evaluation'. Individuals must consider risks and benefits for each of the research vignettes and how the risks and benefits are weighted. Linguistic points are given around coherence between utterances, how content words occurred, use of emotive and evaluating utterances, specificity, prosodic elements and independence in relation to text and the test leader.
Definition of 'Communication of choice'	Assesses 'intelligibility' – an individual's ability to express and formulate a decision and a motivation in an intelligible and relevant way. Linguistic points are awarded for relevance of the reasoning and the decisive utterances, prosodic elements, vocabulary, specificity, formulation of the rationale, occurrence of contradictive utterances or words for ambiguity.
Scoring/Determination of capacity outcome	The LIMD is scored using a linguistically-based analysis. Scoring is based on predefined linguistic features such as independence of the utterances, accuracy, inferences, and coherence for each established criterion. In all scoring verbal (for example syntax, vocabulary), prosodic elements (for example stress and strength of voice) and pragmatic (for example use of meta-linguistic devices like irony, cohesion of utterances, inference) aspects are considered. Principles for scoring of comprehension, evaluation and intelligibility are defined and each criteria receives

		a score (the scores are 0, 1, 2, 3) on a continuous scale on four levels. The summed score (3 x 3 criteria = 9, across 3 vignettes, gives a maximum score of 27). This is used as measure of LIMD.
		Discharge destination decision
Capacity to consent to discharge destination protocol based on the MCA[1,26]	Description	A formal assessment of capacity to make a decision about discharge destination was conducted by a Clinical Neuropsychologist using a semi-structured interview, based on the MCA[1]. The interview incorporated information about the patient that the multidisciplinary team had provided in their questionnaires. Participants were then asked questions about the information provided.
	Inter-rater reliability	$r = 0.44$, $p = 0.06$[26]
	Definition of 'Understanding'	Assessed through questions exploring whether the individual could understand relevant information.
	Definition of 'Retention'	Assessed through questions exploring whether the individual could retain the relevant information whilst making the decision.

	Definition of ability to 'use and weigh information'	Assessed through questions exploring whether the individual could assess the relevant information to arrive at a choice.
	Definition of 'Communication of choice'	Assessed through questions exploring whether an individual could communicate their decision.
	Scoring/Determination of capacity outcome	Each part of the capacity assessment was scored as either 'yes' or 'no', depending on whether the individual was able to meet that standard. If an individual was able to meet all standards, then they would score 'yes' for overall capacity. if they were unable to meet one or more standards, they would score 'no' for overall capacity.
Capacity to appoint an enduring power of attorney (PoA)		
Capacity to appoint an enduring PoA	Description	A semi-structured interview to assess capacity to create an enduring PoA was devised for the study, as there are currently no validated questions available for assessing this type of capacity. This interview was based on the legal judgement given in *RE K, Re F*[146]. The important points

protocol based on the MCA[1,20]		regarding an enduring PoA were explained to patients in a structured format at the start of each interview. Individuals are then asked questions about the content of the information presented.
	Inter-rater reliability	$r = 0.83$[20]
	Definition of 'Understanding'	The patient (donor) must understand the following: - The attorney will be able to assume complete authority over the donors affairs - The attorney will be able to do anything with the donor's property which the donor could have done - The authority will continue if the donor should be or should become mentally incapable - If the donor should be or should become mentally incapable, the power will be irrevocable without confirmation by the court of protection.
	Definition of 'Retention'	Assessed through questions exploring whether the individual could retain the relevant information for long enough to make the decision.
	Definition of ability to 'use and weigh information'	Assessed through questions exploring whether the individual could assess the relevant information to arrive at a choice.

	Definition of 'Communication of choice'	Assessed through questions exploring whether an individual could communicate their decision.
	Scoring/Determination of capacity outcome	The assessment was tape-recorded, and another independent assessment of capacity made by a separate assessor. Both assessors were medical students who had been trained to assess capacity using the interview schedule. Where there was disagreement, this was discussed and resolved. The final decision for each patient was reviewed by a third assessor, a consultant in old age psychiatry experienced in assessing capacity to create an enduring PoA, by listening to the tape-recorded interview.
Variety of decisions		
RCAT Functional Inquiries[29]	Description	RCAT is a multidisciplinary team in Canada with the sole function of assessing and addressing capacity issues. It is important to note that RCAT does not assess all adults in its jurisdiction who require a capacity evaluation. RCAT's mandate is to become involved only with the most complex cases, or where there are gaps in assessment services or where a second opinion is required due to case complexity. The Functional Inquiries are detailed semi-structured

	interviews, based upon the legislation in Canada, that enable the interviewer to quickly and accurately describe potential areas of incapacity, or areas of preserved capacity, across multiple domains.
Inter-rater reliability	NR; As part of the assessment, the team must reach consensus around their assessment.
Definition of 'Understanding'	Assesses ability to understand and identify options: 'adequate factual knowledge base and understanding of options'.
Definition of 'Retention'	N/A – arguably assessed within 'understanding'
Definition of ability to 'use and weigh information'	Assesses 'adequate appraisal of outcome and justification of choices' (explores values/beliefs).
Definition of 'Communication of choice'	Assesses the 'ability to follow-through' (or direct a surrogate to follow-through) with chosen option'.
Scoring/Determination of capacity outcome	Responses are scored as 'sufficient', 'borderline' or 'deficient'. RCAT uses a unique consensus model to arrive at a determination regarding the patient's capacity, thereby reducing the

	possibility of personal bias and making sound use of the resources of the multidisciplinary team. Team members meet to discuss the results of the discipline-specific assessments, any collateral interviews, and the capacity interview. Each team member (Clinical Neuropsychologist, Occupational Therapist, Social Worker) then independently rates the patient's capacity using a 7-point Likert scale. These are then compared and a consensus on capacity is reached.

Note. NR = Not Reported; N/A = Not Applicable; Capacity to consent to research protocol based on MCA[33]; LIMD: the Linguistic Instrument for Medical Decision-Making[49]; Capacity to consent to discharge destination protocol based on the MCA[26]; Capacity to appoint an enduring PoA protocol based on the MCA[20]; RCAT Functional Inquiries: Regional Capacity Assessment Team[147].

Appendix 1.H

Summary of Included Studies

Table A3

Summary of Study Aims, Analyses, and Main Findings

Study	Aim(s)	Analyses	Significant association found? (Y/N)	Reported regression coefficient	SQ
Basset[17]	To explore whether neuropsychological testing can identify a common underlying cognitive operation impaired in patients with AD.	Bivariate correlations were carried out to examine the relationship of variables to performance on HCAT. Authors correlated participant's ability to answer questions correctly and their performance on outcome measures. Stepwise regression was then used to examine the predictive contributions of variables that showed high bivariate correlations with correctly answering the HCAT questions.	Y	Trails A β = NR predicts 89% variance in 'understanding'	6/13
Bertrand et al.[37]	To explore the relationship between behavioural and psychological	Exploratory correlations investigated the relationship between cognitive variables (higher score on MMSE and digit-span indicates better performance; lower scores on ADAS-cog indicate better performance) and clinical competence (total scores in the Mac-CAT-T subscales	Y	N/A	8/13

	symptoms of dementia (BPSD) and clinical competence, especially the capacity to consent to treatment, in Alzheimer's disease (AD).	ranged as follows: 'Understanding' 0–6; 'Appreciation' 0–4; 'Reasoning' 0–8; and 'Expression' 0–2, higher scores indicate increased likelihood of competence).			
Dreer et al.[15i]	To identify cognitive predictors of medical decision-making capacity (MDC) in participants with moderate to severe traumatic brain injury (TBI).	For each CCTI standard, the authors developed three cognitive models: a model of baseline capacity performance (baseline), a model of capacity performance at 6-months follow up using 6-month cognitive predictors (6-months), and a model of capacity performance at 6-month follow-up using baseline cognitive predictors (baseline – 6-month model). Specifically, baseline cognitive predictors were correlated with CCTI performance at baseline (Time 1, n = 34) (baseline model) and 6-month follow-up (Time 2, n = 24) (baseline – 6-month model). Six-month follow-up cognitive predictors were correlated with CCTI performance at 6-month follow-up (6-month model). At all time-points the range of scores on the CCTI were as follows: 'Understanding' (0-70), 'Appreciation' (0-8), 'Reasoning' (0-12) and 'Expression' (0-4). For each group analysis at each time point, the authors first used univariate correlations (Pearson r) as a data reduction step to identify a pool of candidate cognitive predictors for each CCTI standard. This predictor pool was then	Y	WMS-R LM-1 β = 0.938 (*SE* = 0.246) and category fluency β = 1.344 (*SE* = 0.5) predict 56% variance in 'understanding'; WAIS-III arithmetic β = .0382 (*SE* = 0.120) predicted 24% variance in 'understanding'; WMS-R LM-2 β = 0.189 (*SE* = 0.069), and WAIS-R digit-span β = 0.348 (*SE* = 0.156) predicted 32% variance in 'reasoning'.	10/14
Dreer et al [15ii]			Y	Token test β = 2.487 (*SE* = 0.698), SDMT (oral); β = 0.289 (*SE* = 0.129), and WAIS-III arithmetic β = 1.185 (*SE* = 0.531) predicted 72% variance in 'understanding'; Token test β = 0.333 (*SE* = 0.131) predicted 24% variance in 'appreciation'; WAIS-III arithmetic β = 0.566 (*SE* = 0.165) and RAVLT	

		entered into a stepwise multiple linear regression analysis using backward elimination to identify the key multivariate predictors for standards. In this approach, all variables that passed the univariate screening process were entered into the predictive model followed by sequential elimination of the weakest predictor variables until only those making a statistically significant contribution to the model ($p < .05$) remained.		β = 0.117 (*SE* = 0.046) predicted 48% variance in 'reasoning'.	
Dreer et al [15iii]			Y	Token test β = 1.28 (*SE* = 0.503), WAIS-R digit-span β = 1.68 (*SE* = 0.705) and trails B β = -0.042 (*SE* = 0.02) predicted 71% variance in 'understanding'; WAIS-III arithmetic β = 0.323 (*SE* = 0.108) predicted 29% variance in 'appreciation'; Token test β = 0.306 (*SE* = 0.148) and WAIS-III arithmetic β = 0.671 (*SE* = 0.196) predicted 61% variance in 'reasoning'.	
Dymek et al.[38]	To explore the relationship of neurocognitive change to competency loss in in cognitively impaired patients with PD.	Univariate and multivariate analyses were conducted to identify key neurocognitive predictors of performance on each capacity standard for the PD group ('Understanding' range 0-70, 'Appreciation' range 0-8, 'Reasoning' range 0-12, and 'Expression of choice' range 0-4). Univariate correlations were used as a data reduction step to identify potential predictors of performance on each capacity standard for the multivariate analyses. For each capacity standard, the four neurocognitive variables with the strongest univariate correlations were selected for follow-up multivariate analyses. This number of predictor variables reflected the minimum 5:1 subject to predictor ratio. Each set of univariate predictors was entered into stepwise regression analyses for its respective capacity standard, using capacity standard score as the dependent	Y	EXIT-25 β = NR predicts 56% variance in 'understanding', increasing to 68% variance when the DRS memory subscale is added to the model β = NR; DRS memory Subscale β = NR predicts 55% variance in 'communication of a decision'	7/13

		variable and the selected neuropsychological measures as the predictor variables. For each capacity standard, nonparametric discriminant function analyses (DFA) were conducted to identify predictors of competency status (capable, marginally capable, and incapable) for the PD sample. Cut-off scores statistically derived from control performance were used to determine these categories. Cut off scores were established that defined capable as a score equal to or greater than 1½ SD below the control mean on the capacity standard, marginally capable as a score falling between 1½ and 2½ SD below the control group mean on the capacity standard, and incapable as a score falling below 2½ SD below the control mean on the capacity standard. Two DFA were conducted for each capacity standard. The first DFA was run using the one neuropsychological variable with the highest univariate correlation as the predictor of competency status. The second DFA was run using the two neuropsychological variables with the highest univariate correlations as the predictors of competency status.			
Gersteneck er et al.[19]	To explore the effects of cognition on 'understanding' in a medical context, in patients with brain metastasis	'Understanding' scores ranged from 0-82. For all of the cognitive tests, raw scores were used, with higher scores indicating better cognitive performance, except for the Halstead-Reitan test, where lower scores indicate better performance. Pearson product moment correlations were calculated to examine the relationship between understanding scores and neurocognitive performances, age, education, time since diagnosis, and BDI scores. An independent *t* test was used to determine whether understanding scores varied by sex. One-way analysis of	Y	HVLT β = 1.14 (*SE* = 0.17) predicts 58% variance in 'understanding', increasing to 68% when letter fluency is added to the model (HVLT β = 0.79 (*SE* = 0.19); letter fluency β = 0.25 (*SE* = 0.08)); Predicted equation for CCTI understanding score: 510 +	8/12

	variance was conducted to determine whether understanding scores varied by race (i.e. black or African American, white or European-American, and other). Patients were classified as exhibiting intact or impaired understanding based upon psychometric cut-off scores derived from control performance. Intact understanding was defined as a score >1.5 SD below the control group mean. Impaired understanding in this study was defined as a score $\leq$ 1.5 SD below the control group mean. A series of independent t tests was conducted to determine whether neurocognitive performance, age, education, time since diagnosis, and BDI scores varied according to understanding status. Two Pearson's chi-square tests were conducted to determine whether sex or race varied according to understanding status. Variables found to be significantly associated with understanding scores or that varied according to understanding status were then used to construct 2 predictive models. For the first model, a stepwise linear regression was conducted to predict CCTI understanding scores. For the second model, a backwards elimination binary logistic regression was conducted to identify patients with intact and impaired understanding. Predictive accuracy of the resulting model was calculated using receiving operating characteristic (ROC) plots. Using ROC plots, sensitivity and specificity for each potential cut score was obtained by taking sensitivity against 1-specificity. For both models, relative predictive power was obtained through either R^2 or the coefficient of determination of Nagelkerke or pseudo-R^2. Due to the number of comparisons, a more conservative 2-tailed significance level of 0.01 was used for all analyses.	(phonemic fluency x 0.250) + (HVLT-R Total x 0.786). Predicted equation for impaired/intact understanding: 10.249 - (phonemic fluency x 0.349) - (HVLT-R Delayed x 0.312). Scores falling below -0.19 indicate intact 'understanding' with a sensitivity of 94%, specificity of 71%, a negative predictive value of 94%, a positive predictive value of 70% and an overall correct classification rate of 79%, ROC = 0.92.

Gersteneck er et al.[40]	To explore neuropsychologic al predictors of medical decision-making capacity in people with MS	The association between CCTI standards of 'Understanding' (0-70), 'Appreciation' (0-8), and 'Reasoning' (0-12) was evaluated using Pearson's product moment correlations. Neuropsychological variables found to be significantly associated with each capacity standard were then used to construct stepwise linear regression models.	Y	Combined letter and category fluency β = 0.78 (*SE* = 0.15) predicted 59% variance in 'understanding'; SRT β = 0.10 (*SE* = 0.04) predicted 26% variance in 'appreciation'; combined letter and category fluency β = 0.16 (*SE* = 0.04) predicted 40% variance in 'reasoning'.	9/13
Gersteneck er et al.[18]	To determine if cognition can be used to identify persons with cancer at high risk for the impaired ability to understand treatment decisions	Raw scores were used for cognitive tests. Impaired 'Understanding' was defined as a standard deviation score of $\geq$1.5 below the control group mean. A series of independent *t* tests were conducted to determine if neurocognitive performance, age, and education varied according to intact or impaired understanding. Pearson chi-square tests were conducted to determine whether sex or race varied according to 'Understanding' status. For both regression models, the four cognitive variables most associated with 'Understanding' (via correlation or *t* value) were force-entered in a single block. Relative predictive power was obtained through either R^2 or the coefficient of determination of Nagelkerke or pseudo-R^2. Predictive accuracy of the linear regression model was calculated using receiving operating characteristic (ROC) plots. Using ROC plots, sensitivity and specificity for each potential cut-off score was obtained by taking sensitivity against 1-specificity.	Y	LRM: letter fluency β = 0.064 (*SE* = 0.065), HVLT β = 0.526 (*SE* = 0.148), digit-symbol coding β = 0.082 (*SE* = 0.54) and trails B β = -0.21 (*SE* = 0.013) predict 54% variance in 'understanding'. BRM: Predicted equation for impaired/intact 'understanding' = 9.025 − (phonemic fluency x 0.33) − (HVLT-R immediate x 0.235) − (digit-symbol coding x 0.057) − (trails B x 0.005). Scores falling below -9.146 indicate intact 'understanding' with a sensitivity of 80%, specificity of 67%, a negative predictive value of 81%, a positive predictive value of 68% and an	8/13

				overall correct classification rate of 76%, ROC = 0.81.	
Gersteneck er et al.[39]	To investigate the cognitive correlates of medical decision-making in the PSP sample.	Z-scores for CCTI and neuropsychological variables were calculated based on the available normative means and standard deviations. When there was more than one cognitive measure administrated for a domain, an average z-score for the combined measures was used to represent and aggregated domain score. Pearson product moment correlations were calculated to examine the relationship among CCTI variables and neurocognitive performances, age, education, disease duration, and PSPRS scores. Finally, variables that were found to be significantly associated with a particular CCTI capacity standard were then used to construct three forced-entry, linear regression models.	N/A	DRS-2 attention subtest β = 5.5 (*SE* = 3.2), category fluency β = 1.6 (*SE* = 3.3), and combined letter fluency, trails B and frontal assessment battery β = 1.2 (*SE* = 2) predicted 54% variance in 'understanding'; DRS-2 attention subtest β = 1.4 (*SE* = 0.8) predicted 47% variance in 'reasoning'.	8/13
Gersteneck er et al.[34]	To investigate the clinical correlates of the ability to provide informed consent in persons with brain metastasis.	Pearson's product-moment correlations were calculated to determine if age, education, and cognitive variables were significantly associated with CCRI variables. Point biserial correlations were used to determine if gender, sex, and clinical variables were significantly associated with CCRI variables. Variables found to be significantly associated with CCRI variables were then carried forward for use in the regression models. For these models, all variables that correlated significantly with CCRI variables were placed into a forced-entry linear regression. Relative predictive power was obtained using R^2. To help balance the probability of committing a type I or type II error, a two-tailed alpha of .01 was used for all analyses.	Y	Education β = 2.43 (*SE* = 0.62), TMT B β = 2.43 (*SE* = 0.62), and Chemotherapy treatment β = 8.96 (*SE* = 3.16), predicted 52% variance in 'understanding'; HVLT-R immediate β = 0.06 (*SE* = 0.05), HVLT-R delayed β = -0.01 (*SE* = 0.10), and TMT B β = -0.004 (*SE* = 0.01), predicted 22% variance in 'appreciation'; Education β = 0.32(*SE* = 0.10), HVLT-R delayed β = 0.03 (*SE* = 0.11), TMT B β = -0.007 (*SE* = 0.01), whole brain radiation therapy β = 0.66 (*SE* = 0.37), and chemotherapy treatment β	7/13

				= 1.06 (*SE* = 0.50), predicted 43% variance in 'reasoning'.	
Gregory et al.[20]	To investigate if capacity to create an EPA is significantly related to the degree of cognitive impairment in Alzheimer patients, and whether Mini Mental State Examination score (MMSE) is a good predictor of a patient's capacity	Participants were deemed to have capacity or to not have capacity. The association between capacity and degree of cognitive impairment, gender, and presence/absence of qualifications was tested using Pearson chi-Square or Fisher's Exact Test, where appropriate. Difference in MMSE scores, age, and age of leaving school in patients with and without capacity was tested using Mann–Whitney U tests. Stepwise forward conditional binary logistic regression was carried out using capacity status as the dependent variable and MMSE score, age, gender, age of leaving school and presence/absence of qualifications as the independent variables in the model. A receiver operating characteristic (ROC) analysis was performed, plotting the sensitivity of MMSE as a screening tool for capacity against 1-specificity.	N/A	The probability of having capacity to consent increased by 60% for every 1-point increase on the MMSE (*OR* = 1.6, 95% CI 1.3-2.0); ROC analysis: score of 18 on the MMSE showed high specificity (82.6%) and sensitivity (86.2%) in predicting 'understanding'; ROC = 0.921 (95% CI 0.863-0.979).	7/13
Guarino et al.[21]	To examine associations between clinically determined capacity to consent and other characteristics	The associations between capacity (capacity vs no capacity for 'Understanding' deemed by the site investigator), demographics, ICQ perceived understanding scores (range 0-14), and the AD cognitive and functioning measures were analysed by univariate and multivariate logistic regression. All tests of significance were conducted at the 5% level, without correction for multiplicity.	N/A	The odds of a participant lacking necessary 'understanding' increased by 13% for every 1 point decrease on the MMSE (*OR* = 0.87, 95% CI 0.8 - 0.94); The odds of a participant lacking necessary 'understanding' increased by 7% for every 1 point increase on the ADAS-Cog, *OR* = 1.07, 95% CI 1.03-1.10); The odds of a participant lacking necessary 'understanding' increased by 3% for every hour increase in caregivers	6/12

				time, (*OR* = 1.03, 95% CI 1.01 - 1.05); the odds of a participant lacking necessary 'understanding' increased if they were black (*OR* = 2.22, 95% CI 1.23 -4.01).	
Gurrera et al.[22]	To examine the contribution of neuropsychological test performance to treatment decision-making capacity in community volunteers with mild to moderate dementia.	Principal Components Analysis (PCA) was applied to dementia group ability scores across capacity measures. PCA was also applied to dementia group neuropsychological performance data for data reduction because their number was large relative to sample size, and they are highly inter-correlated. Neuropsychological principal components were rotated using Equamax criteria. A series of stepwise linear regressions was conducted with scores for each decisional ability principal component as the dependent variable and neuropsychological component scores as independent variables. Only components with eigenvalues > 1 were considered. Probability of *F* to enter was 0.05, probability of *F* to remove was 0.1. Significance levels were two-tailed.	N/A	Combined WMS-III LM-1 and -2 and the Boston Naming Test β = 0.757 (*SE* = NR) predicted 57.3% variance in 'understanding'; combined digit-span composite score, letter fluency, and vocabulary β = 0.351 (*SE* = NR) predicted an additional 12.3% variance in 'understanding'; combined trails A, trails B, mazes, and VSAT total score β = 0.287 (*SE* = NR) predicted an additional 8.2% variance in 'understanding'; combined WMS-III LM-1 and -2 and the Boston Naming Test β = 0.337 (*SE* = NR) predicted 11.3% variance in 'appreciation'; combined trails A, trails B, mazes, and VSAT total score β = 0.283 (*SE* = NR) predicted an additional 8% variance in 'appreciation'; combined digit-span composite score, letter fluency and vocabulary β = 0.23 (*SE* = NR) predicted an additional 5.3% variance in 'appreciation';	6/13

				combined WMS-III LM-1 and -2, and the Boston Naming Test β = 0.447 (*SE* = NR) predicted 19.9% variance in 'reasoning'; combined trails A, trails B, mazes, and VSAT total score β = 0.341 (*SE* = NR) predicted an additional 11.6% variance in 'reasoning'; combined digit-span composite score, letter fluency, and vocabulary β = 0.282 (*SE* = NR) predicted an additional 7.9% variance in 'reasoning'; combined WMS-III LM-1 and-2, and the Boston Naming Test β = 0.228 (*SE* = NR) predicted 5.2% variance in 'communication of a decision'; combined digit-span composite score, letter fluency and vocabulary β = 0.224 (*SE* = NR) predicted an additional 5% variance in 'communication of a decision'	
Hamann et al.[41]	To analyse the preferences of patients with amnestic mild cognitive impairment (aMCI) and mild dementia in AD regarding different aspects of	Patients with aMCI and mild dementia in AD were pooled for the analyses to increase the variance in the data set. Pearson correlations were used to correlate performance on the capacity standards in the Mac-CAT-T ('Understanding' 0–6; 'Appreciation' 0–4; 'Reasoning' 0–8; and 'Expression' 0–2,) with MMSE score.	Y	N/A	7/13

	decisions and to correlate these findings with different measures of patient decision-making capacity				
Jefferson et al.[23]	To assess decisional capacity performance and the neuropsychological correlates of such performance to better understand higher-level instrumental activities of daily living in individuals with mild cognitive impairment (MCI)	Pearson correlations were conducted between three of the Mac-CAT-CR dimensions 'Understanding' (score range 0-26), 'Appreciation' (score range 0-6), and 'Reasoning' (score range 0-8) and neuropsychological performance in the MCI group.	Y	N/A	8/13
Kim and Caine[24]	To examine the limits and utility of the MMSE as a screening tool for assessing capacity	The authors categorised participants as either 'capable' or 'incapable' on standards of 'Understanding', 'Appreciation', and 'Reasoning' on the basis of validated cut-off scores of 18/26 for 'Understanding', 5/6 for 'Appreciation', and 6/8 for 'Reasoning'. The authors performed a receiver operating characteristic (ROC)	N/A	Scores <19 on the MMSE showed high specificity (85%) but low sensitivity (41%) in predicting 'understanding'. Scores >26 on the MMSE showed high sensitivity (94%) but low specificity (30%) in	5/13

	in AD to consent to research studies	analysis. This analysis plotted the sensitivity of a test against the value of 1-specificity, where sensitivity is the ability of a test to identify a true case among all cases, and specificity is the ability to identify a true 'non-case' among all 'non-cases'. A 'true positive' was defined as a participant who lacked capacity, as determined by one of the four standards of capacity set out in the Mac-CAT-CR, who scored at or below a given MMSE score.		predicting 'understanding'. ROC = 0.756 (95% CI 0.601 to 0.911); Scores <19 on the MMSE showed high specificity (94%) but low sensitivity (45%) in predicting 'appreciation'. Scores >26 on the MMSE showed high sensitivity (95%) but low specificity (35%) in predicting 'appreciation'. ROC = 0.762 (95% CI 0.607 to 0.917); Scores <19 on the MMSE showed high specificity (93%) but low sensitivity (39%) in predicting 'reasoning'. Scores >26 on the MMSE showed high sensitivity (91%) but low specificity (36%) in predicting 'reasoning'. ROC = 0.811 (95% CI 0.672 to 0.950).	
Lui et al.[44]	To assess mental competence in Chinese patients with mild and very mild dementia with a semi structured assessment method and the impact of repeated presentations of information on	Spearman correlation coefficients between Mac-CAT-T ratings 'Understanding' (score range 0-6), 'Appreciation' (score range 0-4), 'Reasoning' (score range 0-8), 'Expressing a choice' (score range 0-2) and demographic and clinical variables were calculated.	Y	N/A	8/13

	patients' mental competence.				
Lui et al.[25]	To explore the relationship of the four decision-making abilities ('understanding', 'appreciation', 'reasoning', and 'expressing a choice'/'communication') and neuropsychological performance in patients with very mild and mild dementia.	Spearman correlation coefficients between the Mac-CAT-T summary scores ('Understanding' 0–6; 'Appreciation' 0–4; 'Reasoning' 0–8 and; 'Expression of choice' 0–2) and demographic and clinical, and cognitive variables were derived. Stepwise linear regression analyses were conducted to evaluate whether the correlated variables predicted each of the four Mac-CAT-T capacity standards. Capacity standards were entered as dependent variables, with the correlated variables as independent variables.	Y	ADAS-Cog β = -0.45 (*SE* = NR) and category fluency β = 0.34 (*SE* = NR) predicted 45% variance in 'understanding'; MMSE β = 0.64 (*SE* = NR) predicted 39% variance in 'appreciation'; category fluency β = 0.42 (*SE* = NR) predicted 20% variance in 'reasoning'; ADAS-Cog β = -0.63 (*SE* = NR) predicted 30% variance in 'communication of a decision'.	8/13
Lui et al.[42a]	To assess if decisional capacity and the four decision-making abilities related to decisions concerning medication management were impaired among community-	Spearman correlation coefficients between ACED ability scores 'Understanding' (score range 0-10), 'Appreciation' (score range 0-8), 'Reasoning' (score range 0-10), 'Expression of choice' (score range 0-2) and demographic and cognitive measures were calculated. For each ACED ability score, linear regression analyses were conducted to evaluate whether correlated variables were predictive in each participant group and for all participants. The ability score was entered as the dependent variable, with cognitive and demographic scores entered as independent variables.	N/A	MMSE β = 0.297 (*SE* = 0.085) together with the Mac-CAT-T β = 0.351 (*SE* = 0.249) predicted 27% variance in 'understanding'. The MMSE was not a significant predictor of 'appreciation' β = 0.057 (*SE* = 0.067) or 'reasoning' β = -0.079 (*SE* = 0.072).	8/13
Lui et al.[42b]			N/A	MMSE was not a significant predictor of 'understanding' β = 0.122 (*SE* = 0.079), however age	

	dwelling Chinese older persons in Hong Kong with amnestic mild cognitive impairment (MCI) and mild Alzheimer's disease (AD), as compared with cognitively normal older adults			was β = -0.276 (*SE* = 0.028) and together with Mac-CAT-T score β = 0.371 (*SE* = 0.164) they predicted 34% variance in 'understanding'; MMSE was not a significant predictor of 'appreciation' β = 0.144 (*SE* = 0.077), however years of education was β = 0.220 (*SE* = 0.055) and predicted 16% variance in 'appreciation'; MMSE was not a significant predictor of 'reasoning'	
Lui et al.[43a]	To examine the relationship between these decisional abilities and cognitive and functional measures among patients with MCI or early dementia	Spearman correlation coefficients were calculated between the ACED ability scores ('Understanding' (0-10), 'Appreciation' (0-8), 'Reasoning' (0-10), and 'Expressing a choice' (0-2)) and the demographic and clinical variables and cognitive and functional measures. Linear regression analysis was carried out to explore the interrelationships among correlated variables.	N/A	MMSE β = 0.285 (*SE* = 0.067) together with participant sex β = 0.225 (*SE* = 0.349) and Mac-CAT-T score β = 0.308 (*SE* = 0.214) predicted 38% variance in 'understanding'; MMSE did not significantly predict 'appreciation' or 'reasoning'	8/13
Lui et al.[43b]			N/A	MMSE β = 0.293 (*SE* = 0.088) together with Mac-CAT-T score β = 0.280 (*SE* = 0.190) predicted 40% variance in 'understanding'; category fluency β = 0.320 (*SE* = 0.035) together with Mac-CAT-T score β = 0.289 (*SE* = 0.195) predicted 30% variance in	

				'appreciation'; category fluency β = 0.255 (*SE* = 0.05) together with Mac-CAT-T score β = 0.254 (*SE* = 0.157) predicted 22% variance in 'reasoning'	
Mackenzie et al.[26]	To investigate the effect of cognitive problems and other factors on the capacity to make a decision about discharge destination and to compare the impressions of multidisciplinary team (MDT) members regarding capacity with a formal assessment	Participants were scored as either having capacity or not having capacity. T-tests were employed for parametric data, and Mann–Whitney tests and chi-squared for non-parametric data, to compare performance on cognitive tests for those with and without capacity.	N	N/A	9/12
Marson et al.[36]	To identify cognitive predictors of competency performance and status in Alzheimer's disease (AD) using three	As a data reduction step, neuropsychological test scores were correlated with capacity standards of 'Understanding', 'Appreciation' and 'Reasoning' scores using Pearson *r* (no score ranges given for capacity standards). Because of the large number of univariate correlations, a significance value of $p < 0.025$ was used. The resulting set of univariate predictors was then entered into stepwise multivariate regression analyses to identify key neuropsychological predictors of capacity	Y	DRS conceptualisation subtest β = NR predicted 70% variance in 'understanding'; Controlled Oral Word Fluency predicted 58% variance in 'appreciation', which increased to 61% variance when category fluency β = NR was added to the model; auditory comprehension screen β = NR	6/13

	differentially stringent legal standards for capacity to consent	standard performance within groups and into parametric and nonparametric discriminant. function analyses to identify predictors of competency status for the entire sample. The nonparametric analyses were added because of' the non-normal multivariate distribution of the competency classes for the legal standards.		predicted 44% variance in 'communication of a decision'.	
Marson et al.[45]	To investigate qualitative behavioural changes associated with declining medical decision-making capacity (competency) in patients with AD	CCTI scores for each domain were calculated quantitatively 'Understanding' (0-70), 'Appreciation' (0-8), 'Reasoning' (0-12) and 'Expression' (0-4). The authors also developed a system of qualitative scores for identifying behavioural changes and errors reflected in AD patient responses on the CCTI test items. The majority of the error codes were developed based on the investigators' experience with control and AD patient verbal responses produced during piloting and standardization of the CCTI. The qualitative scoring system consists of 16 mutually exclusive error codes conceptually organized into four domains: 1) language dysfunction, 2) executive dysfunction, 3) affective dysfunction, and 4) compensatory responses. The individual codes were developed based on behaviours that had first been noted during pilot testing of the CCTI. They were then grouped into domains based on behavioural similarities and the authors' initial views regarding underlying mechanisms. Regression analyses were carried out to identify error types that predicted performance on each capacity standard. Depending on LS score range, either logistic or linear regression was used. The authors identified neurocognitive correlates of selected error codes in the	N/A	Trails A predicted 32% variance in the error 'loss of Task' β = NR, which explained 23% variance in 'understanding' and 18% variance in 'appreciation'; the Token test predicted 43% variance in the error 'loss of detachment' β = NR, which explained 29% variance in 'understanding' and 14% variance in 'reasoning'; the WAIS-R similarities subtest predicted 16% variance in the error 'incoherent responses' β = NR, which explained 36% variance in 'understanding'; DRS construction subtest predicted 15% variance in the error 'nonresponsive answers' β = NR, which explained 15% variance in 'understanding' and 24% variance in 'appreciation'; performance on the Boston Naming Test and DRS memory subtest predicted 15% and 21% variance in the error 'delegation' respectively β = NR,	6/13

		AD sample using univariate Spearman correlation and linear regression. The neuropsychological measures with the highest univariate correlations with each specified error code were entered into regression equations to identify neurocognitive predictors of that code.		which predicted 33% variance in 'communication of decision'.	
Marson et al.[46]	To investigate research consent capacity (RCC) in patients with MG and identify cognitive abilities and clinical factors associated with RCC in MG	Data reduction was carried out through bivariate correlations, which were computed to examine cognitive and demographic predictors of 'Appreciation' (score range 0-4), 'Reasoning' (score range 0-6), and 'Understanding' (score range 0-91) using Pearson's test. For each standard, the authors selected the three top cognitive correlates reaching at least the .05 significance level and entered them into a multivariate stepwise regression analysis. Cognitive variables were retained in each model if the change in R^2 achieved by their entry was significant at the .05 level.	Y	Letter fluency β = NR predicted 63% variance in 'understanding', which increased to 71% variance when category fluency β = NR was added to the model; category fluency β = NR predicted 61% variance in 'appreciation'; letter fluency β = NR predicted 30% variance in 'reasoning'.	8/13
Moelter et al.[27*]	To examine the association between cognitive domains and research consent capacity in a PD sample	Associations between cognitive domains for the DRS-2 and the MoCA and the Mac-CAT-CR 'Understanding' (0-26), 'Appreciation' (0-6), 'Reasoning' (0-8), and 'Expressing a choice' (0-2) were tested with Spearman rank order correlations, owing to the non-normal distributions of the Mac-CAT-CR ability domains and MoCA subscale measures.	Y	N/A	8/13
Moro et al.[35]	To explore which cognitive functions impact comprehension.	Pearson's coefficient (r) was employed to evaluate potential associations between comprehension of the consent form and clinical variables. In order to minimize the possibility of false-positives from multiple comparisons, we calculated a composite index for each cognitive domain in the neuropsychological tests (memory, attention, apraxia, executive functions, and	Y	N/A	6/13

		language). Tests for five a priori hypotheses were conducted using Bonferroni adjusted alpha levels of .01 per test (.05/5).			
Okonkwo et al.[28a]	To investigate cognitive predictors of medical decision capacity in people with MCI.	Participants' scores on each CCTI standard were summed across vignettes A and B to create a composite variable ('Understanding' score range 0-78, 'Appreciation' score range 0-8, 'Reasoning' score range 0-12, 'Expressing choice' range 0-4). Comparisons of group performance on these composite variables were performed using one-way analysis of variance. As a data reduction strategy, zero-order correlations were computed within each study group to assess the simple bivariate relationship between CCTI standards and neuropsychological variables. Next, maintaining a minimum ratio of 10 subjects per variable, those neuropsychological measures correlated with the CCTI standards at the .01 significance level were selected for entry into multiple variable stepwise linear regression analyses to create models of consent capacity. Neuropsychological variables were included in each model if the change in R^2 achieved by their entry was significant to the .05 significance level.	Y	WMS-III LM 2 β = NR predicted 44% variance in 'understanding', CVLT-2 β = NR predicted a further 5% variance in 'understanding', trails 3 β = NR predicted a further 4% variance in 'understanding'; WMS-III LM2 β = NR predicted 16% variance in 'appreciation'; CVLT-2 β = NR predicted 24% variance in 'reasoning', trails 3 β = NR predicted a further 6% variance in 'reasoning'.	8/13
Okonkwo et al.[28b]			Y	Trails A β = NR predicted 50% variance in 'understanding', WMS-III LM-1 β = NR predicted a further 17% variance in 'understanding' and DRS-2 memory subtest β = NR predicted a further 6% variance in 'understanding'; trails A β = NR predicted 33% variance in 'appreciation'; category fluency β = NR predicted 25% variance in 'reasoning'; 10/36 Spatial Recall test β = NR predicted a further 10% variance in 'reasoning'; perseveration errors on Conners' continuous performance test β = NR predicted 35% variance in	

				‘communication of a decision’ and CLOX 2 predicted a further 16% variance in ‘communication of a decision’	
Pachet et al.[29c]	To determine if cognitive deficits alone, as identified by the MMSE, predict capacity versus the value of clinical judgment in capacity assessments	A chi-square analysis was used to examine the relationship between 2 dichotomous variables: cognitive impairment (as determined by MMSE score) and determination of capacity (capacity vs no capacity; as determined by RCAT). The sensitivity, specificity, positive predictive values (PPV), and negative predictive values (NPV) were calculated for scores between 17 and 26 on the MMSE. A chi-square analysis was also completed for each diagnostic group and the sensitivity, specificity, PPV, and NPV were again calculated for each group. Lastly, to further investigate whether there is an ‘‘optimal cut point,’’ they completed receiver operating characteristic (ROC) curves, which allowed them to assess whether there was an optimal MMSE cut point that maximizes sensitivity and specificity. *P* values below .01 were considered statistically significant throughout the analysis.	N/A	Score of ≤22 on the MMSE showed high specificity (93-100%) but variable sensitivity (22-45%) in predicting ‘understanding’; ROC = NR	6/13
Pachet et al.[29d]			N/A	Score of ≤24 on the MMSE showed high specificity (100%) but variable sensitivity (18-41%) in predicting ‘understanding’; ROC = NR	
Palmer et al.[30]	To compare decisional capacity among 3 diagnostic groups, and to examine the degree to which impaired ‘understanding’ can be detected with a brief set of	The authors used Spearman rho to examine the degree to which age, education, and MMSE totals were each significantly correlated with the total scores from the 3-item questionnaire and with each of the 4 Mac-CAT-CR subscale scores ‘Understanding’ (score range 0-26), ‘Appreciation’ (score range 0-6), and ‘Reasoning’ (score range 0-8), and ‘Expression of choice’ (range 0-2). within each diagnostic group. The authors also examined the sensitivity and specificity of each potential cut score from the 3-item questionnaire to impaired vs intact	Y	Score of <24 on the MMSE showed high specificity (87.5%) and sensitivity (61.5%) in predicting ‘understanding’; ROC = 0.88 (95% CI 0.79-0.96). There is no instance where the balance of sensitivity and specificity on the MMSE exceeded that associated with an alternative 3-item questionnaire, involving direct enquire of ‘understanding’.	9/13

	screening questions	understanding as measured with the understanding subscale of the Mac-CAT-CR (≥ 15 = capacity).			
Reen et al.[14]	To evaluate whether the BRIMMS protocol can improve MS patients' treatment understanding and reduce conflict in treatment decisions compared to consultation as usual.	Bivariate Pearson's product-moment correlations assessed the relationship between treatment understanding scores following consultation style with raw scores on measures of numerical reasoning, health literacy, premorbid IQ and symptoms of MS. Treatment understanding scores were collapsed across aural and visual forms of each consultation style. That is, understanding scores for BRIMMS and usual consultation were computed in the correlation. Due to multiple correlations, a stringent alpha of $p<.01$ was applied and accepted as significant.	Y	N/A	9/13
Spataro et al.[16]	To explore the effects of demographic and clinical variables on capacity to consent to treatment in an ALS cohort.	Multinomial regression analysis was performed to assess which variables are likely to be independently associated with Mac-CAT-T sub-scores. Wilcoxon matched-pairs test was used to compare differences between CCT outcomes at baseline and 1-year follow-up. For all analyses, p values < 0.05 were considered significant.	Y	The odds of a participant lacking necessary 'understanding': increased by 10% for every 1-year increase in age *OR* = 0.9 (95% CI 0.86 – 0.95); decreased by 15% for every 1-year increase in education *OR* = 1.15 (95% CI 1.04-1.27); and decreased by 10% for every one point increase on a phonemic fluency measure *OR* = 1.10 (95% CI 0.89-0.10). The odds of a participant lacking necessary 'reasoning' increased by 5% for every 1-year	11/13

				increase in age *OR* = 0.95 (95% CI 0.91-0.98).	
Stormoen et al.[31]	To identify the most powerful cognitive component(s) that best predicted medical decision-making capacity in patients with AD and MCI	A principle component analysis was used to reduce the number of cognitive test measures into independent components covering as much as possible of the total test variance. Eigen values >1 were used to decide the number of components, and all components should have high loadings (>0.70) in at least two test measures. Varimax rotation was used to identify a parsimonious structure. A stepwise linear regression analysis was used to examine the relationship (common variance, r^2) between the LIMD score (overall capacity score range 0-27) and possible cognitive predictors (component scores). The degree of importance for each component regarding LIMD was presented as β weights.	N/A	Combined similarities, information, and inference tests β = 0.66 (*SE* = NR) combined Rey-Osterrieth Retention, RAVLT (learning), and RAVLT (retention) β = 0.43 (*SE* = NR), combined forward word sequence, trails A, and trails B β = 0.32 (*SE* = NR) and combined Wechsler digit-span forwards, Wechsler digit-span backwards, and Stroop colour word-interference test β = 0.23 (*SE* = NR) together predicted 73% variance in 'overall capacity'	8/13
Triebel et al.[47]	To investigate the relationship between cognitive functioning and consent capacity in patients with malignant glioma	Bivariate and multivariate analyses were conducted to identify key neurocognitive predictors of CCTI performance ('Understanding' range 0-70, 'Appreciation' range 0-8, 'Reasoning' range 0-12, and 'Expression of choice' range 0-4). As a data reduction step, bivariate correlations were computed to identify candidate neuropsychological predictors of CCTI performance using the Pearson test for 'Understanding', 'Appreciation' and 'Reasoning' and a Spearman rho test for 'Expressing a choice'. The top three neuropsychological correlates reaching the 0.05 significance level were entered into separate multivariate stepwise linear regression models for each standard. Neuropsychological variables were retained in each	Y	HVLT β = NR predicted 67% variance in 'understanding', which increased to 72% when category fluency β = NR was added to the model; HVLT β = NR predicted 45% variance in 'appreciation' which increased to 54% variance when category fluency β = NR was added to the model; HVLT β = NR explained 33% variance in 'reasoning'	7/13

		model if the change in R^2 achieved by their entry was significant at the 0.05 significance level. Exploratory correlational analyses were also conducted examining the relationship between performance on the CCTI standards and demographic factors (age, education, and gender) and clinical variables (Karnofsky score, time since diagnosis, and BDI scores).			
Triebel et al.[32e]	To examine neurocognitive predictors of MDC in cognitively healthy adults and in patients with mild and moderate/severe TBI evaluated within 6 weeks of injury using an established capacity assessment instrument (Capacity to Consent to Treatment Instrument (CCTI))	The authors examined bivariate correlations between demographic, clinical variables (age, years of education, race/ethnicity, gender, GOAT score), and cognitive domain scores with the CCTI standard scores 'Understanding' (0-70), 'Appreciation' (0-8), 'Reasoning' (0-12). The purpose of this analysis was to examine associations between demographic and clinical variables with CCTI consent standard performance. The authors next analysed the relationships between the cognitive domain scores and the CCTI scores. They used ordinary least squares regression models to examine the association between cognitive domain scores and 'Understanding', and ordinal regression to examine the association between domain scores and the odds of higher scores on 'Appreciation' and 'Reasoning'. Separate regression analyses were performed for each group (e.g., controls, mTBI, and msevTBI). All four neuropsychological domain scores were simultaneously entered into the regression equation for each standard. The proportional odds assumption was evaluated using the test of parallel lines and by comparing the observed and expected cell counts at each threshold; no substantial deviations from proportional odds were observed. Model fit was measured using R^2 and adjusted R^2 for the	N/A	Combined CVLT trials 1 -5, short delay free recall, long-delay free recall, WMS-III LM-1 and -2 β = 2.464 (*SE* = 0.580) predicted 62% variance in 'understanding'; Combined WAIS-III arithmetic and WTAR reading *OR* = 1.408 (95% CI 1.082 -1.399) predicted 23% variance in 'appreciation'; combined trails A, trails B, digit-symbol coding, symbol search *OR* = 1.264 (95% CI 0.996 - 1.605) and combined CVLT trials 1 -5, short delay free recall, long-delay free recall, WMS-III LM-1 and -2 *OR* = 1.309 (95% CI 1.048 -1.634) predicted 34% variance in 'reasoning'.	8/13
Triebel et al.[32f]			N/A	Combined WAIS-III arithmetic and WTAR reading β = 2.835 (*SE* = 0.631) and combined CVLT trials 1 -5, short delay free recall, long-delay free recall, WMS-III LM-1 and -2 β = 3.499 (*SE* = 1.018)	

		'Understanding' model and goodness of fit test and Nagelkerke pseudi-R^2 for 'Appreciation' and 'Reasoning'. For 'Appreciation' and 'Reasoning', the authors analysed odds rations and the 9% confidence intervals around them for cognitive variables that were significant in the models. Secondary analyses were run examining bivariate correlations between CCTI consent standards for 'Understanding', 'Appreciation' and 'Reasoning', and the cognitive domain scores separately for complicated and uncomplicated mild TBI cases.		predicted 76% variance in 'understanding'; category fluency *OR*= 1.754 (95% CI 1.013-3.034) together with combined WAIS-III arithmetic and WTAR reading *OR*= 1.289 (95% CI 1.031-1.611) predicted 48% variance in 'appreciation'; combined WAIS-III arithmetic and WTAR reading *OR* = 1.289 (95% CI 1.041-1.598) predicted 53% variance in 'reasoning.'.	
Warner et al.[33]	To assess characteristics associated with capacity to consent to research in a dementia sample	Participants were assessed to either have capacity or not have capacity. Associations between capacity (consent versus assent) and the following predictors were investigated: clinical diagnosis (Alzheimer disease versus vascular dementia); gender; ethnic origin (other versus white); MMSE score (>24 vs <24), age (years); length of education (years); duration of dementia (months) and ADAS-Cog score. Single variable analysis was undertaken by fitting separate logistic regression models for each of the predictors. Odds ratios with 95% confidence intervals were obtained, along with p-values using the likelihood ratio chi-square test. Multivariable logistic regression was then undertaken. All the above variables were entered into a single model. The significance of each variable in the model was then assessed using the likelihood ratio chi-square test. The least significant term was removed and the model re-fitted. This process was iterated to identify which of the	Y	The probability of having capacity to consent increases by 17% for every 1 point decrease on the ADAS-Cog, (*OR* = 0.830, 95% CI 0.771-0.893); The probability of having capacity to consent significantly increases if you score over 24 on the MMSE, *OR* = 6.271, 95% CI 2.920-13.471); The probability of having capacity to consent increases by 8.4% for every 1 year decrease in age, *OR* = 0.916, 95% CI 0.872-0.962); The probability of having capacity to consent increases by 65.8% if you have AD compared to vascular dementia, *OR* = 0.342, 95% CI 0.146-0.799).	6/13

	above variables were significantly independently associated with the presence of capacity.	

Note. 'Significant association found? Y/N?' and 'Reported regression coefficient' refer to the reported relationship between capacity outcomes and cognitive performance, in line with the question set out in this review; N/A = Not Applicable; SQ = study quality assessment; study quality rating using the NIH tool[11]. SQ is calculated as Total "Yes" – Total NA per paper. This is in line with tool guidance which suggests that NA scores should not count negatively towards the quality rating.

Appendix 1.I

Cognitive Tests Used in Included Studies

Table A4

Description of Outcome Measures used in Studies

Test name	Reference(s) for versions used	Brief Description
10/36 Spatial Recall Test	Boringa et al.[112]	A subtest in the Brief Repeatable Battery of Neuropsychological tests (BRB-N). Assesses visuospatial learning and delayed recall. Consists of a 6x6 checkerboard with 10 checkers. The board is put in front of the subject for 10s. After presentation, the subjects attempt to reproduce the original design on an empty board. This process is repeated twice and after 15 minutes, the subject is asked to recall the design again. The score is the total number of correct responses for the three trials and the delayed recall trial.
7/24 Spatial Recall Test	Rao[67]	Assesses visuospatial learning, susceptibility of such learning to proactive and retroactive interference and ability to recall visuo-spatial information following a period of delay. Consists of a 4x6 checkerboard with seven checkers randomly placed. The board is put in front of the subject for 10s. After presentation, the subjects attempt to reproduce the original design on an empty board.
Animal Naming	Spreen and Strauss[101]	Involves the generation of words to semantic categories (e.g., animals) within a limited time-period (1 minute).

Arithmetic	Wechsler Adult Intelligence Scale 3rd Edn (WAIS-III; Wechsler[54])	Assesses attention and working memory. Requires subjects to manipulate mental mathematical problems.
Attentional Matrix	Spinnler and Tognoni[107]	A selective visuo-perceptual attention test in which the patient has to cross out target-digits (visual search). Involves three different matrices (1-digit, 2-digit, and 3-digit targets) made up of 11 strings of 10 digits each with 45 seconds to complete each matrix. The overall score range is 0-60. The test has no memory load and only a presumptively small spatial exploratory load.
Behavioural Assessment of the Dysexecutive Syndrome (BADS) Rule Shift	Wilson et al.[99]	Evaluates a subject's ability to change an established pattern of responding, using familiar materials. In part 1, a response pattern is established according to a simple rule. In part 2, the rule is changed, and clients have to adapt their responses, inhibiting their original response set.
Benton Visual Discrimination Test (BVDT)	Benton et al.[62]	Assessment of perceptual matching, using multiple-choice stimuli. Subjects are required to identify and select the image that matches the target image. The score is determined by the number of correct responses.
Block Design	Wechsler Adult Intelligence Scale Revised (WAIS-R; Wechsler[54])	Measure of spatial visualization ability and motor skill. The subject uses hand movements to rearrange blocks that have various colour patterns on different sides to match a pattern. The items in a block design test can be scored both by accuracy in matching the pattern and by speed in completing each item.
Boston Naming Test (BNT)	Goodglass et al.[73]	Assesses confrontation naming and lexical retrieval abilities. Consists of 60 black and white line drawings of objects, which subjects are required to name.
California Verbal Learning Test (CVLT)	Delis et al.[93] Delis[91]	Assesses verbal learning abilities and verbal retrieval abilities.

Cantonese Mini Mental State Examination (CMMSE)	Chiu et al.[94]	It has been approved for translation and validated in Hong Kong. See MMSE for further details.
Category Fluency	Lam et al.[97]; Lezak et al.[71]	A measure of semantic fluency. Subjects are asked to generate exemplars in three categories (animals, fruits, and vegetables) for 1 minute per category. Information about the number of exemplars generated in 1 minute was recorded. The score represents the total number of exemplars generated after the three 1-minute trials
Category Fluency	Gladsjo et al.[105]; Spinnler and Tognoni[107]	Involves the generation of words to semantic categories (e.g., animals) within a limited time period (1 minute).
Cognitive Competency Test - Verbal Reasoning subtest	Wang and Ennis[114]	A measure of abstraction. The subject is asked questions about such topics as time management and safety hazards. Higher scores indicate better insight and judgement.
Cognitive Estimation	Carlesimo et al.[109]	A measure of reasoning and self-monitoring. The task requires the "selection and regulation of cognitive planning", in order to answer questions that are not immediately answerable but the response to which can be effectively guessed using general knowledge e.g. "What is the height of a traffic light?" The test comprised 21 questions that required participants to give oral, numerical responses. For each item, the bizarreness score was either 0 (not bizarre) or 1 (bizarre). Hence, the total bizarreness score ranged from 0 and 21. The higher the bizarreness score, the greater the impairment in cognitive estimation.
Cognitive subscale of the "Alzheimer's Disease Assessment Scale" (ADAS-Cog)	Mohs[87]; Schultz, et al.[58]; Rosen et al.[127]	A brief neuropsychological assessment used to assess the severity of cognitive symptoms of dementia. The ADAS-Cog consist of 11 questions: Word Recall Task; Naming Objects and Fingers; Following Commands; Constructional Praxis; Ideational Praxis; Orientation; Word Recognition Task; Remembering Test Directions; Spoken Language; Comprehension; Word-Finding Difficulty. Score represents the number of errors in each task.

Comprehension	Wechsler Memory Scale-Revised (WMS-R; Wechsler[64]); Wechsler Adult Intelligence Scale 3rd Edn (WAIS-III; Wechsler[59]); Wechsler Adult Intelligence Scale Revised (WAIS-R; Wechsler[54])	Assesses a subject's ability to deal with abstract social conversations, rules, and expressions.
Concept Formation	No reference given	Subjects were presented with three pairs of objects and asked to explain what each of pair of items had in common. Each comparison was rated at 2 points if it entailed an abstract generalisation, 1 point if a response entailed a specific 'concrete' likeness, and 0 points if no similarity was observed. The total score ranges from 0 to 6.
Controlled Oral Word Fluency from the Multilingual Aphasia Examination (COWF)	Benton and Hamsher[61]	This task involves supplying a subject with a letter, after which he or she has a limited time-period (most commonly 60s) during which to orally produce as many words as possible that begin with the designated letter.
Constructive Apraxia	Spinnler and Tognoni[107]	A test of copying and visuospatial abilities. Patient is asked to copy two-dimensional drawings.
Delayed Word Test (Recall)	Lam et al.,[96]	A word list of 10 items is read to each subject and they are asked to recall the list 10 minutes later. The total score is the number of items correctly recalled.

Dementia Rating Scale (DRS) and (DRS-2)	Jurica et al.[82]; Mattis[75]	The Dementia Rating Scale (DRS) is considered a very useful instrument to assess patients with dementia. The tasks are grouped into five subscales, each one evaluating different cognitive areas, namely: Attention, Initiation/Perseveration (I/P), Construction, Conceptualization and Memory.
Digit- and Visual-Spans	No reference given	A measure of attention and working memory. Consists of sequences of ascending and descending digit- and visual-spans. Subjects are required to repeat them back. Scores indicate the number of correct responses.
Digit-Span	Wechsler Memory Scale-Revised (WMS-R; Wechsler[64]); Wechsler Memory Scale 3rd Edn (WMS-III; Wechsler[79]); Wechsler Adult Intelligence Scale (WAIS; Wechsler[100]); Wechsler Adult Intelligence Scale 3rd Edn (WAIS-III; Wechsler[59]); Wechsler Adult	Assesses attention and concentration by asking subjects to repeat back sequences of digits forwards and in reverse.

	Intelligence Scale Revised (WAIS-R; Wechsler[54]); Spinnler and Tognoni[107]	
Digit Symbol Coding	Wechsler Adult Intelligence Scale 3rd Edn (WAIS-III; Wechsler[59]); Wechsler Adult Intelligence Scale Revised (WAIS-R; Wechsler[54])	It consists of (e.g., nine) digit-symbol pairs (e.g., 1/-,2/⊥ ... 7/Λ,8/X,9/=) followed by a list of digits. Under each digit, the subject should write down the corresponding symbol as fast as possible. The number of correct symbols within the allowed time (e.g., 90 or 120 sec) is measured.
Executive Clock Drawing Test (CLOX)	Royall et al.[113]	Subjects are asked to draw a clock face and set the time. The CLOX is divided into an unprompted task that is sensitive to executive control (CLOX1) and a copied version that is not (CLOX2).
Forward and Backwards word sequences	Ostberg et al.[124]	Involves a speech motor task to control for sequential motion rate and two-word production tasks that contrast forward and backward production of the same helical word sequence, namely, the names of the months.
The Frontal Systems Behaviour scale (FRsBS)	Grace[119]	A 46-item rating scale exploring the frontal-lobe related behavioural impairment, consisting of three subscales: apathy, disinhibition, executive dysfunction. A total score of $\geq$65 is associated with a clinically significant frontal-lobe mediated dysfunction.

Hopkins Verbal Learning Test (HVLT) and (HVLT-R)	Brandt[103]; Brandt and Benedict[77]	A list learning test, which consists of 12 nouns within three semantic groups. The acquisition variable consists of three acquisition trials in which the administrator reads the words aloud and then asks the subject to repeat as many as he/she can remember in any order. A delayed recall trial is introduced after 20–25 min, in which the subject is asked to simply retrieve as many of the words listed in the acquisition trial as he/she can remember. The subject is not informed about the delayed recall trial beforehand. Lower raw scores indicate difficulties with the task.
Information	Wechsler Adult Intelligence Scale Revised (WAIS-R; Wechsler[54])	Assesses degree of general information acquired from culture.
Information/Orientation	Wechsler Memory Scale-Revised (WMS-R; Wechsler[64])	Assessing general personal information and orientation to person, place, and time.
Judgement Test	Stern and White[92]	Measures judgment pertaining to home safety, health, and medication issues
Letter and Category Fluency	Tallberg et al.[126]	Swedish norms for tests of word fluency. Subjects are given 1 minute apiece to name as many words as possible beginning with the letters, “F”, “A”, and “S”. Also for category naming - involves the generation of words to semantic categories (e.g. animals) within a limited time period (1 minute).
Letter Fluency Test	Spreen and Strauss[81]; Spinnler and Tognoni[107]	Subjects are given 1 minute apiece to name as many words as possible beginning with the letters, “F”, “A”, and “S”

Logical Memory 1 and 2	Wechsler Memory Scale-Revised (WMS-R; Wechsler[64]); Wechsler Memory Scale 3rd Edn (WMS-III; Wechsler[79]); Wechsler Adult Intelligence Scale Revised (WAIS-R; Wechsler[54])	Logical Memory I—assessing recall of orally presented stories; Logical Memory II—assessing ability to recall information from the stories in Logical Memory I after a 30-minute delay, as well as assessing recognition of information from stories.
Matrix Reasoning	Wechsler Adult Intelligence Scale 3rd Edn (WAIS-III; Wechsler[59]);	Measure of nonverbal abstract problem solving, inductive reasoning and spatial reasoning. Subjects are required to Identify the missing part of a visual matrix.
Mazes Test (large print version)	Christensen et al.[90]	Visuospatial, time limited test assessing planning behaviour.

Mental control	Wechsler Memory Scale-Revised (WMS-R; Wechsler[64])	Assessing speeded performance for reciting the alphabet, counting forward and backward, days of the week forward and backward, months of the year forward and backward, counting by 6s while stating days of the week in order.
Omission and Commission errors on the Conners' Continuous Performance Test	Conners[111]	Primarily used to differentiate between subjects with ADHD and controls. Requires subjects to maintain vigilance and react to the presence (or absence) of a specific stimulus within a set of distracters presented continuously. Assesses 'errors of commission', whereby responses occur when no response is required and is assumed to reflect impulsivity. It also measures 'errors of omission', whereby there is an absence of a response to a target and is assumed to reflect symptoms of inattention.
Oral Trail Making Test	Ricker and Axelrod[83]	An oral version of the standard TMT that has no visual or graphomotor components.
Paired Verbal Associations 1 and 2	Wechsler Memory Scale-Revised (WMS-R; Wechsler[64])	Verbal Paired Associates I—assessing recall memory for orally presented word pairs that have been previously learned; Verbal Paired Associates II—assessing ability to recall associations from Verbal Paired Associates I after a 30-minute delay, as well as assessing recognition of word pairs.
Raven's Progressive Matrices	Raven et al.[108]	A series of multiple-choice items of abstract reasoning. Each item depicts an abstract pattern in a 2 x 2 or 3 x 3 matrix; all cells contain a figure except for the cell in the right lower corner. Participants are asked to identify the missing segment that would best complement the pattern constituted by the other cells among a set of alternatives that are positioned beneath the matrix.
Reading and Writing	Johansson[123]	Swedish norms for assessing logico-grammatical sentences, reading speed and reading aloud.

Similarities	Wechsler Adult Intelligence Scale (WAIS; Wechsler[100]); Wechsler Adult Intelligence Scale 3rd Edn (WAIS-III; Wechsler[59]);	Assesses abstract verbal reasoning through a series of questions asking how two words are alike.
Simple Auditory Comprehension Test (SAC)	Eisenson[74]	Assesses auditory verbal comprehension of oral sentences.
Spatial Span	Wechsler Memory Scale-Revised (WMS-R; Wechsler[64]); Wechsler Memory Scale 3rd Edn (WMS-III; Wechsler[79])	Assessing ability to reproduce increasingly longer spatially separated sequences of forward and backward taps on blocks.
Stroop Colour and Word Test	Golden and Freshwater[121]	Assessment of cognitive flexibility. The subject is required to look at each page and move down columns reading words or naming the ink colours as quickly as possible, without interference, within a given time limit. Scores represent the number of items completed on each of the three stimulus sheets.

Story Recall	Spinnler and Tognoni[107]	Patients are presented with a story orally and asked for immediate verbatim recall, and then for delayed recall an hour later. The stories are in Italian.
Symbol Search	Wechsler Adult Intelligence Scale 3rd Edn (WAIS-III; Wechsler[59])	A measure of visual perception and speed. Requires the subject to identify identical symbols under time pressure.
The Brief Visuospatial Memory Tests Revised, recall trials (BVMTR)	Langdon et al.[118]	The BVMTR is a visuospatial memory task consisting of an array of six figures presented three times to participants for 10 seconds each. Once the array is removed, participants are required to draw the figures from memory, ensuring that the correct shapes are drawn in the correct location. Figures are scored for their correct shape and their correct location, and patients could score a maximum of 12 points in each trial. The total score is calculated by summing the scores of all three trials.
The Chinese version of the Cognitive subscale of the "Alzheimer's Disease Assessment Scale" (ADAS-Cog)	Chu[95]	A standard cognitive instrument that was specifically designed for the evaluation of cognitive deficit in patients with AD, translated. See ADAS-Cog for further details.
The consortium to establish a registry for Alzheimer's disease (CERAD) Battery: Word list learning (both immediate and delayed recall)	Morris et al.[53]	Immediate: This is a free-recall memory test that is also included in the Alzheimer's Disease Assessment Scale (ADAS; 20) and assesses learning ability for new verbal information. Three trials of a 10-item word list are presented. On each trial, the 10 words are presented in a different order. The subject is instructed to read aloud each word as it is presented. Immediately following each trial, the subject is asked to recall as many items as possible. The maximum score on each trial is 10. Delayed: This test assesses the ability to recall, after a 5-minute delay, the 10 words given in the word-list memory test. The maximum number of correct responses is 10. For each subject, a savings score (Savings) is computed and is presented as a percentage, reflecting the relative amount of verbal information retained over the delay interval ([Delay/Trial 31 X 100 = Savings).

The Controlled Oral Word Association Test from the Multilingual Aphasia Examination (COWA)	Benton et al.[62]	Subjects are given 1 minute apiece to name as many words as possible beginning with the letters, "C", "F", and "L". The score represents the total number of words generated for each letter within the time limit.
The Executive Interview (EXIT-25)	Royall et al.[68]	A bedside test of simple executive abilities, including verbal and spatial fluency, inhibition, primitive reflexes, and set flexibility.
The Frontal Assessment Battery (FAB)	Dubois et al.[84]	A battery of 6 subtests, exploring the functions of the frontal lobes. Subtests include: Conceptualisation (abstract reasoning/similarities); Mental Flexibility (Letter fluency); Motor Programming (Luria Sequencing); Sensitivity to Interference (subjects must provide an opposite response to the examiner's alternating signal); Inhibitory Control (go-no go paradigm); Environmental Autonomy. Total score is determined by adding the scores of the subtests.
The Galveston Orientation and Amnesia Test (GOAT)	Levin et al.[60]	Involves questions assessing orientation to time, place, and person and recall of recent events. The score represents the number of error points.
The Grooved Pegboard	Heaton et al.[72]	Assessment of motor speed and fine coordination The subject is required to place 25 pegs in board with their dominant hand. The score represents the time that this takes to complete.
The Middlesex Elderly Assessment of Mental State: MEAMS	Golding[98]	A screening tool for gross impairment of cognitive skills in the elderly. It allows assessors to differentiate between functional illnesses and organically based cognitive impairments. Includes 12 subtests covering the following areas of functioning: Orientation; Memory; New Learning; Naming; Comprehension; Arithmetic; Visuo-spatial skills; Perception; Fluency; Motor Perseveration. The score represents the total number of correct responses.

The Mini Mental State Examination (MMSE)	Bertolucci, et al.[56]; Folstein et al.[57]	A commonly used set of questions for screening cognitive function. It provides measures of orientation, registration (immediate memory), short-term memory (but not long-term memory) as well as language functioning. The score represents the number of correct responses.
The Montreal Cognitive Assessment (MoCA)	Nasreddine et al.[106]	A brief, 30-question assessment to detect cognitive impairment. It assesses the following cognitive domains: short-term memory; visuospatial abilities; executive functioning; attention; language; abstract reasoning and orientation. The score represents the total number of correct responses.
The Paced Auditory Serial Addition Test (PASAT)	Benton et al.[148]	A measure of sustained attention and information processing speed. Requires the subject to add pairs of digits presented at two rates of speed. The subject is instructed to add 60 pairs of digits such that each number is added to the one that immediately precedes it and report the outcome verbally. The digits are presented by tape, first with a speed of every 3 s one digit, the second trial with every 2 s one digit. The subject is required to respond verbally quickly, inhibit encoding of his own response while attending to the next stimulus in a series, and perform at an externally determined pace. The score is the number of correct responses per trial.
The Rey Auditory Verbal Learning Test (RAVLT)	Rey[65]; Taylor[66]	A measure of word list learning. Participants hear 15 words over 5 trials. Their score is the total number of words recalled.
The Selective Reminding Test (SRT)	Buschke[78]	Designed to measure verbal learning and memory through the use of a list-learning procedure over multiple trials. involves reading the subject a list of 12 unrelated words and then having the subject immediately recall as many of these 12 words as possible. Every trial after the first involves selectively presenting only those words in which the subject did not recall on the immediately preceding trial. The selective reminding trials proceed in this manner until the subject is able to correctly recall all 12 words on three consecutive trials, or until 12 trials have been completed.

The Symbol-Digit Modalities Test (SDMT) and (SDMT-R)	Smith[63,85]	Examines sustained attention and concentration by primarily assessing complex visual scanning and tracking. The subject examines a series of nine meaningless geometric symbols which are labelled 1 to 9. During 90 s the subject substitutes symbols in a row by the corresponding number and responds verbally. The score is the number of correct substitutions within the time limit.
The Wide Range Achievement Test 3rd Edn (WRAT-3)	Wilkinson[115]	A measure of basic academic skill of reading, spelling, and mathematical computation necessary for effective learning, communication, and thinking. Subtests include reading, recognition, spelling, and arithmetic. The score represents the number of correct responses.
The Wisconsin Card Sorting Test (WCST)	Berg[69]; Grant and Berg[70]	Assessment of reasoning/concept formation and set-shifting. The subject is required to deduce principles by which to sort cards via feedback on performance. The score is determined by the number of perseverative responses.
Token Test	Benton and Hamsher[61]; Spellacy and Spreen[102]	Measures verbal comprehension of single and multiple step commands. The subject follows commands using coloured tokens.
Trail Making Test (TMT)	Army Individual Test Battery[88]; Delis et al.[93] ; Reitan[55]; Reitan and Wolfson[104]; Spinnler and Tognoni[107]	Trails A: measures visual-scanning and attention abilities; higher scores indicate worse performance. Trails B: Assesses sequencing and mental flexibility; higher scores indicate worse performance
Unusual Views	Lezak et al.[76]	This is a test of perceptual ability. An item is presented from a conventional view and an unconventional view and the subject must decide whether the items are the same or different.

Verbal Judgements	Spinnler and Tognoni[107]	In this test, the patient is asked to discern the differences between pairs of items, to explain the meanings of some proverbs, to discern the absurdity of some phrases and to classify some items. The score can vary within the range from 0 to 60.
Verbal and Spatial reasoning task (VESPAR): series completion subtask	Langdon and Warrington[116]	A test of spatial reasoning. The series completion task consisted of 25 items, progressively increasing in difficulty from simple arithmetic series to more complex numerical patterns. Participants were required to select the subsequent number to complete a series (i.e. 2, 4, 8, ?) from a choice of four numbers (16, 10, 12, 20).
Visual Reproduction	(Wechsler Memory Scale 3rd Edn (WMS-III); Wechsler[79])	Assessing ability to reproduce difficult-to-verbalize designs after a brief exposure.
Visual Search and Attention Test (VSAT)	Trenerry et al.[89]	This is a measure of attentional abilities. Subjects are presented with a visual array consisting of letters or symbols. In trials one and two of the VSAT, subjects are asked to rapidly search the array and cross out all of the items that are the same form as a target (single-feature search coalitions). In trials three and four, subjects are asked to rapidly cross out all of the items in the array that match the target in terms of both form and colour (dual-feature search conditions). In the standard administration of the VSAT, subjects 'are given 60 seconds to cross out as many target items as they can, and their performances are scored on only the last two trials of the test. The score represents the number of targets crossed out in 60 seconds on the left and right side of the array as well as the two sides combined.
Verbal Span	Spinnler and Tognoni[107]	In this short-term verbal memory test, the patient is asked to repeat, in their order of presentation, lists of spoken disyllabic words. The score is the length of the longest sequence that is repeated correctly in two out of three trials.

Vocabulary	Wechsler Adult Intelligence Scale 3rd Edn (WAIS-III; Wechsler[59])	Assess the degree to which a subject is able to learn, appropriately comprehend and verbally express vocabulary.
Wechsler Test of Adult Reading (WTAR)	Wechsler[117]	The WTAR provides an estimate a subjects level of intellectual functioning before the onset of injury or illness. It is is composed of a list of 50 words that have atypical grapheme to phoneme translations. The score represents the number of correct responses.

Appendix 2.A

SIP Instructions for Authors

To assist the reader, all tables and figures have been integrated into the main text, as opposed to submitted separately.

1. SUBMISSION

Authors should kindly note that submission implies that the content has not been published or submitted for publication elsewhere except as a brief abstract in the proceedings of a scientific meeting or symposium.

Once the submission materials have been prepared in accordance with the Author Guidelines, manuscripts should be submitted online at http://www.editorialmanager.com/bjhp

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The British Journal of Health Psychology publishes original research on all aspects of psychology related to health, health-related behaviour and illness across the lifespan including:

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- management of acute and chronic illness
- responses to ill-health
- screening and medical procedures
- psychosocial mediators of health-related behaviours
- influence of emotion on health and health-related behaviours
- psychosocial processes relevant to disease outcomes
- psychological interventions in health and disease
- emotional and behavioural responses to ill health, screening and medical procedures

- psychological aspects of prevention

3. MANUSCRIPT CATEGORIES AND REQUIREMENTS

The types of paper invited are:

- papers reporting original empirical investigations, using either quantitative or qualitative methods, including reports of interventions in clinical and non-clinical populations;
- theoretical papers which report analyses on established theories in health psychology;
- we particularly welcome review papers, which should aim to provide systematic overviews, evaluations and interpretations of research in a given field of health psychology (narrative reviews will only be considered for editorials or important theoretical discourses); and
- methodological papers dealing with methodological issues of particular relevance to health psychology.

Authors who are interested in submitting papers that do not fit into these categories are advised to contact the editors who would be very happy to discuss the potential submission.

Papers describing quantitative research (including reviews with quantitative analyses) should be no more than 5000 words (excluding the abstract, reference list, tables and figures). Papers describing qualitative research (including reviews with qualitative analyses) should be no more than 6000 words (including quotes, whether in the text or in tables, but excluding the abstract, tables, figures and references). In exceptional cases the Editor retains discretion to publish papers beyond this length where the clear and concise expression of the scientific content requires greater length (e.g., explanation of a new theory or a substantially new method). Authors must contact the Editor prior to submission in such a case.

All systematic reviews must be pre-registered. The pre-registered details should be given in the methods section but blinded for peer review (i.e., 'the review was preregistered at [BLINDED]'); the details can be added at proof stage. Registration documents should be uploaded as title page files when possible, so that they are available to the Editor but not to reviewers.

Please refer to the separate guidelines for **Registered Reports**.

COVID-19 Research

The BJHP has received an overwhelming number of COVID-19 related submissions. We can only consider papers that are providing new and novel data on COVID-19. We particularly welcome submissions of intervention studies.

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you to read, they will also be difficult for the editors and reviewers. If your manuscript is difficult to read, the editorial office may send it back to you for revision.

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The manuscript should be submitted in separate files: title page; statement of contribution; main text file; figures/tables; supporting information.

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Abstract

For articles containing original scientific research, a structured abstract of up to 250 words should be included with the headings: Objectives, Design, Methods, Results, Conclusions. Review articles should use these headings: Purpose, Methods, Results, Conclusions. As the abstract is often the most widely visible part of your paper, it is important that it conveys succinctly all the most important features of your study. You can save words by writing short, direct sentences. Helpful hints about writing the conclusions to abstracts can be found **here**.

Keywords

Please provide appropriate keywords.

Acknowledgments

Contributions from anyone who does not meet the criteria for authorship should be listed, with permission from the contributor, in an Acknowledgments section. Financial and material support should also be mentioned. Thanks to anonymous reviewers are not appropriate.

Statement of Contribution

All authors are required to provide a clear summary of ‘what is already known on this subject?’ and ‘what does this study add?’. Authors should identify existing research knowledge relating to the specific research question and give a summary of the new knowledge added by your study. Under each of these headings, please provide 2-3 (maximum) clear outcome statements (not process statements of what the paper does); the statements for 'what does this study add?' should be presented as bullet points of no more than 100 characters each. The Statement of Contribution should be a separate file.

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- Tables and figures (each complete with title and footnotes)
- Appendices (if relevant)

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References

References in published papers are formatted according to the Publication Manual of the American Psychological Association (6th edition). However, references may be submitted in any style or format, as long as it is consistent throughout the manuscript.

Tables

Tables should be self-contained and complement, not duplicate, information contained in the text. They should be supplied as editable files, not pasted as images. Legends should be concise but comprehensive – the table, legend, and footnotes must be understandable without reference to the text. All abbreviations must be defined in footnotes. Footnote symbols: †, ‡, §, ¶, should be used (in that order) and *, **, *** should be reserved for P-values. Statistical measures such as SD or SEM should be identified in the headings.

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Although authors are encouraged to send the highest-quality figures possible, for peer-review purposes, a wide variety of formats, sizes, and resolutions are accepted.

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Legends should be concise but comprehensive – the figure and its legend must be understandable without reference to the text. Include definitions of any symbols used and define/explain all abbreviations and units of measurement.

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- **Language:** Authors must avoid the use of sexist or any other discriminatory language.

- **Abbreviations:** In general, terms should not be abbreviated unless they are used repeatedly and the abbreviation is helpful to the reader. Initially, use the word in full, followed by the abbreviation in parentheses. Thereafter use the abbreviation only.
- **Units of measurement:** Measurements should be given in SI or SI-derived units. Visit the Bureau International des Poids et Mesures (BIPM) website for more information about SI units.
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Except where otherwise stated, the journal operates a policy of anonymous (double blind) peer review. Please ensure that any information which may reveal author identity is blinded in your submission, such as institutional affiliations, geographical location or references to unpublished research. We also operate a triage process in which submissions that are out of scope or otherwise inappropriate will be rejected by the editors without external peer review. Before submitting, please read **the terms and conditions of submission** and the **declaration of competing interests**.

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- Manuscripts that report the collection and integration of qualitative and quantitative data
- Manuscripts that report new data collections regardless of research design

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Funding

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All listed authors should have contributed to the manuscript substantially and have agreed to the final submitted version. Authorship is defined by the criteria set out in the APA Publication Manual:

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The journal expects that where possible all data supporting the results in papers published are archived in an appropriate public archive offering open access and guaranteed preservation. The archived data must allow each result in the published paper to be recreated and the analyses reported in the paper to be replicated in full to support the conclusions made. Authors are welcome to archive more than this, but not less.

All papers need to be supported by a data archiving statement and the data set must be cited in the Methods section. The paper must include a link to the repository in order that the statement can be published.

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In some cases, despite the authors' best efforts, some or all data or materials cannot be shared for legal or ethical reasons, including issues of author consent, third party rights, institutional or national regulations or laws, or the nature of data gathered. In such cases, authors must inform the editors at the time of submission. It is understood that in some cases access will be provided under restrictions to protect confidential or proprietary information. Editors may grant exceptions to data access requirements provided authors explain the restrictions on the data set and how they preclude public access, and, if possible, describe the steps others should follow to gain access to the data.

If the authors cannot or do not intend to make the data publicly available, a statement to this effect, along with the reasons that the data is not shared, must be included in the manuscript.

Finally, if submitting authors have any questions about the data sharing policy, please access the **FAQs** for additional detail.

Publication Ethics

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For help with submissions, please contact: Hannah Wakley, Associate Managing Editor, **bjhp@wiley.com** or phone +44 (0) 116 252 9504.

Author Guidelines updated April 2019

Appendix 2.B

SIP Study Materials





Oxford University Hospitals
NHS Foundation Trust

Private & Confidential

Oxford Children's Hospital
Headley Way
Headington
Oxford
OX3 9DU
Switchboard: 01865 741166
Tel: 01865 234187

Re: Research project - Understanding perceived barriers to the disclosure of psychological difficulties by young people with Coeliac Disease

We would like to invite your child to take part in a research study to understand what it is like for young people to have coeliac disease. We are specifically interested in why young people might feel able or unable to tell their doctor about how coeliac disease is affecting their wellbeing, in order to try and improve support for young people. We are also interested in young peoples' experiences of talking with others about the condition, how well it is understood and how it is viewed.

If your child would like to take part in this research, I will arrange a time to meet with you and your child in person to discuss the research study and what it involves and to answer any queries you/they may have. This could be on the same day that your child takes part in the research study, or a few days before, whichever you and your child would prefer. Due to concerns around limiting contact to minimise risk of COVID-19, there is also the option for you and your child to meet me via Skype video call.

You will be asked to give written consent for your child to take part in this study. Your child will also need to provide written assent to take part. Following this, your child will be interviewed for a maximum of 90 minutes about their experience of having coeliac disease and their experiences of disclosing psychological difficulties associated with coeliac disease to clinicians in the service. This will happen in a private, quiet room at the hospital or at your home, whichever they prefer. We could also carry out the interview via Skype video call. As the purpose of this study is to **understand young peoples' experiences from their perspective, you will not be** required to be present during the interview. If you have any queries about this, please discuss these with me, and I will do my best to address your concerns. There will also be an opportunity for you to share your thoughts at the end of the interview.

This research is being conducted in line with the educational requirements of my doctoral thesis. The results of this research will be used to help us think about how best to support young people with coeliac disease in the service. The medical team will not be directly involved in the research but are inviting your child to take part. Your **child's** care will not be affected by whether or not they choose to participate.

You will find enclosed an information sheet for yourself and one for your child, which will provide all the details of the study. If you or your child would like to find out more information, please contact me using the details in the information sheet. Your child will be under no obligation to participate, but I will be able to answer any questions that you or your child have. If your child would still like to take part in this research, I will arrange a time to meet with you both.

Yours Sincerely

Miranda Wheeler
Chief Investigator

Oxford University Hospitals
NHS Foundation Trust

Oxford Children's Hospital
Headley Way
Headington
Oxford
OX3 9DU
Switchboard: 01865 741166
Tel: 01865 234187

Private & Confidential

Re: Research project - Understanding perceived barriers to the disclosure of psychological difficulties by young people with Coeliac Disease

We would like to invite you to take part in a research study to understand what it is like for young people to have coeliac disease. We are specifically interested in why young people might feel able or unable to tell their doctor about how coeliac disease is affecting how they are feeling emotionally. This is in order to try and improve support for young people. We are also interested in young peoples' experiences of talking with others about the condition; how well it is understood and how it is viewed.

If you choose to take part in this research, I will arrange a time to meet with you in person to discuss the study and what will happen. This could be on the same day that you take part in the research, or a few days before you take part**, it's up to you.** You can also bring a parent/ guardian with you to meet me if you would like to. I will try my best to answer any questions that you or your parent/ guardian may have.

You will be asked to give written consent to take part in this study. You will be asked to talk about your experience of having coeliac disease and your experiences of **talking to professionals in the Gastroenterology service at the Children's hospital** about difficulties relating to how you are feeling emotionally. This will happen in a private, quiet room at the hospital or at your home, whichever you prefer.

As the purpose of this study is to understand your experiences from your point of view, we would prefer to meet with you individually during the discussion. However, your parent/ guardian can be present during the rest of the study visit if you would like them to be. If you have any questions or concerns about this, please discuss these with me, and I will do my best to help. If your parent/ guardian attends the study visit with you, there will be an opportunity for them to share their thoughts at the end of the study visit.

I am carrying out this research as a University student, so results will be written up in a report and given to my teachers. It will be written in a way that makes sure nobody can work out who the young people are that took part. The results of the study will also be used to help us think about how best to support young people with coeliac **disease at the Children's Hospital.** The medical team will not be directly involved in the research but are inviting you to take part. Your care will not be affected by whether or not you choose to participate.

You will find enclosed an information sheet, which will provide all the details of the study. If you would like to take part, you can contact me using the contact details in the information sheet. You will not need to agree to participate at this point, but I will be able to answer any questions that you have. Following this, if you still would like to take part in this research, I will arrange a time to meet with you.

Yours Sincerely

Miranda Wheeler
Chief Investigator



Oxford University Hospitals
NHS Foundation Trust

Oxford Children's Hospital
Headley Way
Headington
Oxford
OX3 9DU
Switchboard: 01865 741166
Tel: 01865 234187

Private & Confidential

Re: Research project - Understanding perceived barriers to the disclosure of psychological difficulties by young people with Coeliac Disease

You recently received some information regarding the above study, which hopes to explore what it is like for young people to have coeliac disease. We are specifically interested in why young people might feel able or unable to tell their doctor about how coeliac disease is affecting their wellbeing, in order to try to improve support for young people. We are also interested in **young peoples'** experiences of talking with others about the condition; how well it is understood and how it is viewed.

We are still recruiting for this study and hope you may consider your child taking part. If you or your child would like to find out more information, you can contact me using the below contact details. Your child will be under no obligation to participate at this point, but I will be able to answer any questions that you or they have.

Following this, if your child would like to take part in this research, a single study visit will take place. During this visit, you will be asked to give written consent for your child to take part in this study. Your child will also need to provide written assent to take part. Following this, your child will be interviewed for a maximum of 90 minutes about their experience of having coeliac disease and their experiences of disclosing psychological difficulties associated with coeliac disease to clinicians in the service. This will happen in a private, quiet room at the hospital or at your home, whichever they prefer. As the purpose **of this study is to understand young peoples' experiences** from their perspective, you will not be required to be present during the interview. If you have any queries about this, please discuss these with the researcher, who will do their best to address your concerns. There will also be an opportunity for you to share your thoughts at the end of the interview.

Please contact me using the contact details below if you have any questions or would like your child to take part. If I do not hear from you, I will assume that you are not interested in taking part in this study and you will not be contacted again.

Contact details:
Email: miranda.wheeler@hmc.ox.ac.uk
Phone: 07469432868

Yours Sincerely

Miranda Wheeler
Chief Investigator



NHS
Oxford University Hospitals
NHS Foundation Trust

Private & Confidential

Oxford Children's Hospital
Headley Way
Headington
Oxford
OX3 9DU
Switchboard: 01865 741166
Tel: 01865 234187

Re: Research project - Understanding perceived barriers to the disclosure of psychological difficulties by young people with Coeliac Disease

You recently received some information regarding the above study, which hopes to explore what it is like for young people to have coeliac disease. We are specifically interested in why young people might feel able or unable to tell their doctor about how coeliac disease affects how they are feeling emotionally. This is in order to try to improve support for young people. We are also interested in young peoples' experiences of talking with others about the condition; how well it is understood and how it is viewed.

We are still recruiting for this study and hope you may consider taking part. If you would like to find out more information, you can contact me using the below contact details. You will not need to agree to participate at this point, but I will be able to answer any questions that you have.

Following this, if you would still like to take part in this research, I will arrange a time for a single study visit where I would meet with you. During this visit, you will be asked to give written consent to take part in this study. You will be asked to talk about your experience of having coeliac disease and your experiences of talking to professionals in the Gastroenterology service at the Children's Hospital about difficulties relating to how you are feeling emotionally. This will happen in a private, quiet room at the hospital or at your home, whichever you prefer.

As the purpose of this study is to understand your experiences from your point of view, we would prefer to meet with you individually during the discussion. However, your parent/ guardian can be present during the rest of the study visit if you would like them to be. If you have any questions or concerns about this, please discuss these with the researcher, who will do their best to help. If your parent/ guardian attends the study visit with you, there will be an opportunity for them to share their thoughts at the end of the visit.

Please contact the me using the contact details below if you have any questions or would like to take part. If I do not hear from you, I will assume that you are not interested in taking part in this study and you will not be contacted again.

Contact details:
Email: miranda.wheeler@hmc.ox.ac.uk
Phone: 07469432868

Yours Sincerely

Miranda Wheeler
Chief Investigator

Oxford University Hospitals
NHS Foundation Trust

Lead Researcher: Miranda Wheeler, Trainee Clinical Psychologist
Email: Miranda.wheeler@hmc.ox.ac.uk
Phone: 07469432868

Parent Information Sheet

Study Title: Understanding perceived barriers to the disclosure of psychological difficulties by young people with Coeliac Disease

We would like to invite your child to take part in a research study. Before you decide, you need to understand why we are doing this research, and what it would involve. Please ask the researcher if there is anything you don't understand.

Why are we doing this research?
The aim of this study is to understand what it is like for young people to have coeliac disease. We are specifically interested in their experiences of talking to medical professionals in the service about the impact that having coeliac disease has on their wellbeing and their thoughts and feelings about this. We want to understand this so that we can think about the best way to help young people who might have difficulties in this area. This research is being undertaken by a University student and will contribute to the fulfilment of an educational requirement (e.g. a doctoral thesis).

Why has my child been invited?
Your child has been invited to help us with this research because they are aged 11-16 and were diagnosed with coeliac disease a minimum of three months ago. We hope that at least 8 young people will be interviewed.

Does my child have to take part?
No, this is optional. Whatever you decide will not affect your child's care at the hospital.

What will happen to my child if they take part?
If your child would like to take part in this research, I will arrange a time to meet with you and your child in person to discuss the research study and what it involves and to answer any queries you/they may have. This could be on the same day that your child takes part in

the research study, or a few days before, whichever you and your child would prefer. You will be asked to give written consent for your child to take part in this study. Your child will also need to provide written assent to take part. Following this, your child will be interviewed for a maximum of 90 minutes about their experience of having coeliac disease and their experiences of disclosing psychological difficulties associated with coeliac disease to clinicians in the service. This will happen in a private, quiet room at the hospital or at your home, whichever they prefer. The interview will be digitally audio recorded. As the purpose of this study is to understand young people's experiences from their perspective, you will not be required to be present during the interview part of the visit. If you have any queries about this, there will be an opportunity to discuss these with the researcher, who will do their best to address your concerns. If your child feels strongly about having you present, then an agreement will be reached whereby you are present for part or all of the interview, but are asked not to contribute during the interview. There will be an opportunity for you to share your thoughts at the end of the interview.

Your child will not have to answer any questions that they do not want to and will not have to tell the researcher anything they do not want to. They can take a break during the interview if they would like to, and they can end the interview at any time. They will be able to withdraw their data from the study up to two weeks after their interview session, at which point data will be de-identified. None of this will affect the care that they receive from the hospital.

What are the possible disadvantages or risks of taking part?

Due to the topic of the study it is possible that your child may become upset during the interview. However, they will be able to stop the interview or take a break at any time. The researcher is a Trainee Clinical Psychologist who has a degree in psychology and experience working with young people with health conditions. This means they have had training in talking to people about emotional topics and supporting them if they become upset. If the researcher thinks that your child is experiencing a high level of distress, you will be informed and there will be a discussion about what the most appropriate support would be. This will either happen in person at the time of the interview, or you will be contacted afterwards. In this instance, a clinical psychologist will write to your GP informing them of this and inviting you to seek support from the psychological team within your local Gastroenterology service. This letter will also be copied to you. If the researcher is concerned that your child or someone else is at risk of harm, they might need to tell you about this straight away. They might also need to tell other professionals to ensure the safety and best interests of your child.

What are the possible benefits of taking part?

We hope that by taking part in this study, your child will be helping us learn more about the emotional consequences of living with coeliac disease and hopefully improve support for young people who have similar experiences in the future.

Will my child be reimbursed for taking part?
Your child will receive £5 for participation. In addition, if you and/or your child travel to the hospital purely to participate in the research, reasonable travel costs will be reimbursed.

What if there is a problem?
If you have a concern about any aspect of this study, you should contact the researcher (details below) who will do their best to answer your questions. Alternatively, you can contact the University of Oxford Clinical Trials and Research Governance (CTRG) office on 01865 616480, or the head of CTRG, email ctrg@admin.ox.ac.uk. If you remain unhappy and wish to complain formally, you can do this through the NHS Complaints Procedure. The Patient Advisory Liaison Service (PALS) is a confidential NHS service that can provide you with support for any complaints or queries you may have regarding the care you receive as an NHS patient. PALS is unable to provide information about this research study. If you wish to contact the PALS team, please contact 01865 221473 or PALS@ouh.nhs.uk.

What will happen with the results of the study?

The results of the study will be used to help the researchers think about how best to support young people with coeliac disease in the service. The research being undertaken will also contribute to the fulfilment of an educational requirement (e.g. a doctoral thesis). On successful submission of the thesis, it will be deposited both in print and online in the University archives to facilitate its use in future research. If so, the thesis will be openly accessible. Additionally, a report will be written for publication in an academic journal. Quotes from interviews may be used in the report however these will be de-identified.

If your child participates in this study, you will have the opportunity to be sent a summary of results from this study. You will be asked to indicate if you would like to receive a summary on the consent form, in addition to how you would like to receive it e.g. by post/email.

How will we use information about your child?
We will need to use information given by you and your child for this research project. This will include **demographic information and information relating to your child's coeliac** disease, including:

- **Your child's age when they were diagnosed with coeliac disease**
- Your family history of coeliac disease
- The services that your child attends for the coeliac disease

People will use this information to do the research or to check your child's records to make sure that the research is being done properly. People who do not need to know who your child is will not be able to see their name or identifiable details. Their data will have a code number instead.

We will keep all information about your child safe and secure.

Will my child's participation in the study be kept confidential?
Yes, all information that is collected about your child during the course of the research will be coded with a participant ID and kept confidential. As explained above we may need to inform your **child's** GP or other professionals that they are experiencing a high level of distress. We would only ever share the information that was necessary to keep your child safe, and we would always do our best to talk to you about it beforehand.

No information that identifies your child will be included in the write-up. Audio recordings will be transcribed and then deleted. Interview transcripts will be labelled with your **child's** participant ID only.

Once we have finished the study, we will keep some of the data so we can check the results. We will keep identifiable information about your child for 6-12 months after the study has finished. This excludes any research documents with personal information, such as consent forms, which will be held securely **and separately at the Children's Hospital** for 5 years after the end of the study and will then be disposed of securely

What are your choices about how your child's information is used?

- Your child can stop being part of the study at any time, without giving a reason. You **will be able to withdraw your child's data up to two weeks after they take** part in the research.
- **We need to manage your child's records in specific ways for the research to be reliable. This means that we won't be able to let you see or change the data we hold** about your child.

Data protection regulation provides you with cont**rol over your child's personal data and how it is used. When you agree to your child's information being used in research,** however, some of those rights may be limited in order for the research to be reliable and accurate.

Where can you find out more about how your child's information is used?
You can find out more about how we use your child's information

- at www.hra.nhs.uk/information-about-patients/

- at https://compliance.web.ox.ac.uk/individual-rights
- by asking one of the research team.

Who is conducting the study?
The Chief Investigator (CI) is Miranda Wheeler, a Trainee Clinical Psychologist at the Oxford Institute of Clinical Psychology Training and Research. The CI is undertaking this as part of an educational project and is supervised by a Senior Clinical Psychologist in the Paediatric Gastroenterology Service.

Who is organising and funding the study?
The University of Oxford is sponsoring the study and it is funded by the Oxford Institute of Clinical Psychology Training and Research.

Who has reviewed the study?
All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee. This is in order to protect your safety, rights, wellbeing and dignity. This study has been reviewed and approved by the HRA and Health and Care Research Wales (HCRW) Approval Research Ethics Committee (REC reference: 19/WA/0323).

What do I do next?
If having read this information sheet you are happy for your child to participate and they would like to be interviewed, please contact the Chief Investigator (CI) using the telephone number or email address below. The CI will be able to answer any questions you have and can then organise an interview if your child still wishes to take part.

Researcher contact details:
Miranda Wheeler, Chief Investigator
Email: Miranda.Wheeler@hmc.ox.ac.uk
Phone: 07469432868

Dr Annabel David, Senior Clinical Psychologist
Email: Annabel.David@ouh.nhs.uk
Phone: 01865 234187

Lead Researcher: Miranda Wheeler, Trainee Clinical Psychologist
Email: Miranda.Wheeler@hmc.ox.ac.uk
Phone: 07469432868

Participant Information Sheet – 11-15 years

Study Title: Understanding perceived barriers to the disclosure of psychological difficulties by young people with Coeliac Disease

What is research?
Research is done to find out the answer to an important question. We would like to invite you to take part in a research study. Please read this information and talk about it with your parent / guardian. Please ask us if there is **anything you don't understand**.

Why is this project being done?
My name is Miranda; I am a student at University. I am doing this research with a research team to find ways to improve support for young people with coeliac disease. This is part of my university work. We want to understand what it is like for young people to have coeliac disease. We are very interested to find out why young people do or do not tell their doctor at the Paediatric Gastroenterology service about how having coeliac disease makes them feel. We hope that this will help us improve the support that young people can get if they are having difficult feelings. **We are also interested in young people's experience of talking** with their friends, teachers and family about having coeliac disease. For example, how other people might think or feel about it. The results of this research will be written up in a report and given to my teachers. I will write the report without naming anyone who took part.



Why me?
You have been chosen to help us with this research because you are aged 11-15 and have had coeliac disease for at least 3 months. I hope that at least 8 young people like you will be come and talk to me.

Do I have to take part?

No you do not! It is up to you. Whatever you decide will not affect your care at the hospital. **You can also change your mind later. If you don't want to take part, just say no!**




What will happen?

If you choose to take part in this research, I will arrange a time to meet with you and your parent/guardian to explain the study and what will happen. I can also answer any questions that you or your parent/guardian might have. This could be on the same day that you take part in the research, or a few days before you take part, **it's up to you**. You will be asked to sign a form to say that you agree to take part (an assent form). We will also ask your parent/guardian to sign a form to say that they agree to you taking part (a consent form).




After this, we will spend up to 90 minutes talking about what it is like to have coeliac disease. We will also talk about your experiences of talking to doctors in the Gastroenterology service at the **Children's hospital** about how having coeliac disease makes you feel. This will happen in a private, quiet room at the hospital or at your home, whichever you prefer. Our conversation will be audio-recorded so that I can listen to it.



As we would like to understand your thoughts and feelings, your parent/guardian will not need to be there during this conversation. If you have any questions or worries about this, please talk to me and I will do my best to help. If you would really like your parent/guardian to be there when you are talking to me, then they can be. However, I would ask them to sit quietly so that you can answer my questions. Your parent/guardian will be able to share their thoughts at the end of the study visit.

You will not have to answer any questions that you do not want to and will not have to tell me anything you do not want to. You can take a break during the conversation if you would like to. You can end the conversation at any time. You can ask me not to use things you said during the conversation in the research up to two weeks after your study visit. None of this will affect the care that you receive from the hospital.

What else might happen?
Some young people can find that talking about having coeliac disease is upsetting. If you think that you might find this upsetting too, you can let me know and I will do my best to help. We can stop talking or take a break at any time. I have learnt a lot about supporting young people with their feelings and have spent a long time working with young people with health conditions, which means that I have had training in supporting people if they become upset. If I think that you are very upset or am worried about yours or someone **else's safety,** I will need to tell your parent / guardian and your GP. This is so that you can be offered some support to feel better. I might also need to tell other professionals. This is to make sure that you are safe.

We hope that by taking part in this study, you will be helping us to learn more about how young people with coeliac disease feel. We hope that this will help us to improve support for young people who have similar thoughts and feelings in the future.

What will I get for taking part?
In exchange for your time and effort we will be offering you £5. Also, if you and your parent/guardian travel to the hospital just to take part in the research, then you will receive money to cover any reasonable travel costs.

What if something goes wrong?
Your parent/guardian will be able to talk to someone who will be able to tell them what they need to do about it.

What happens to what the researchers find out?
The results of the study will be used to help us think about how best to support young people with coeliac disease in the service. The report I write will also be available for other students to look at, to help with their research. When the study finishes, we plan to put the research findings in medical magazines that doctors read. We also plan to present the findings to doctors at meetings. Things that you say during our conversation may be used in the report, however no one would know it was you who said it.



If you participate in this study, your parents will have the opportunity to be sent a summary of results from this study.

Will anyone else know I'm doing this?

All of the information that you give during the study visit, including what you say during the conversation, is **called "research data" and this will be kept** private. At the start of the study, you will be given a participant ID number. Anything you say during the research study will be linked to only this number, your name will be removed from the data. This information will be password protected and only the research team would be able to look at this. Audio recordings will be written out and then the recordings will be deleted. The only time we would share the information you give us with someone else e.g. your parent/guardian or GP, would be if you told us something that made us worried that you or someone else was at risk of serious harm. We would only ever share the information that was needed to keep you safe, and we would always do our best to talk to you about it before we do this. This is part of our responsibility to you to make sure you are safe.

How will we use information about you?

We will need to use information given by you and your parent/ guardian for this research project. This will include information about you (e.g. your age) and information about your coeliac disease, including:

- How old you were when a doctor told you that you have coeliac disease
- If anyone else in your family has coeliac disease
- Other doctors or nurses that you see for your coeliac disease

People will use this information to do the research or to check your records to make sure that the research is being done properly. People who do not need to know who you are will not be able to see your name or details that identify you. Your data will have a code number instead.

We will keep all information about you safe and secure.

Once we have finished the study, we will keep some of the data so we can check the results. We will keep identifiable information about you for 6-12 months after the study has

finished. Consent forms will be held safely and privately at the **Children's Hospital** for 5 years after the end of the study.

Who is in the research team?

I am the lead researcher, I am a Trainee Clinical Psychologist at the Oxford Institute of Clinical Psychology Training and Research. I will be the researcher that meets with you, if you choose to take part.



I am supervised by Dr Annabel David, Senior Clinical Psychologist in the Paediatric Gastroenterology Service.

Did anyone else check the study is ok to do?

Before any research goes ahead it has to be checked by a Research Ethics Committee. This is a group of people who make sure that the research is OK to do. This study has been looked at by the HRA and Health and Care Research Wales (HCRW) Approval Research Ethics Committee (REC reference: 19/WA/0323).



What do I do next?

If you would like to take part in this research, please contact me using the telephone number or email address below or ask your parent/ guardian to. I will be able to answer any questions you or your parent/ guardian have. If you still would like to take part in this research, I will arrange a time to meet with you and your parent/guardian.

Researcher contact details:
Miranda Wheeler, Chief Investigator
Email: Miranda.Wheeler@hmc.ox.ac.uk
Phone: 07469432868

Dr Annabel David, Senior Clinical Psychologist
Email: Annabel.David@ouh.nhs.uk
Phone: 01865 234187



Lead Researcher: Miranda Wheeler, Trainee Clinical Psychologist
Email: Miranda.Wheeler@hmc.ox.ac.uk
Phone: 07469432868

Participant Information Sheet – 16 years

Study Title: Understanding perceived barriers to the disclosure of psychological difficulties by young people with Coeliac Disease

We would like to invite you to take part in a research study. Before you decide, you need to understand why we are doing this research, and what it would involve. Please read this information and ask the researcher if there is **anything you don't understand**.

Why are we doing this research?
The aim of this study is to understand what it is like for young people to have coeliac disease. We are particularly interested in the reasons why young people either feel able or unable to tell their doctor at the Paediatric Gastroenterology service about how coeliac disease is affecting how they are feeling emotionally during their appointments. We hope that this will help us improve emotional support for young people. We are also interested in **young people's experience of talking with other people about the condition, how well it is** understood and how it is viewed. This research is being carried out by a University student, so results will be written up in a report and given to their teachers.

Why have I been invited?
You have been invited to help us with this research because you are aged 16 and were diagnosed with coeliac disease a minimum of three months ago. We hope that at least 8 young people will be come and talk to us.

Do I have to take part?
No, this is optional; it is up to you to decide. Whatever you decide will not affect your care at the hospital.

What will happen to me if I take part?
If you choose to take part in this research, I will arrange a time to meet with you in person to discuss the study and what will happen. This could be on the same day that you take part **in the research, or a few days before you take part, it's up to you.** You can also bring a

parent/ guardian with you to meet me if you would like to. I will try my best to answer any questions that you or your parent/ guardian may have. You will be asked to give written consent to take part. Following this, you will have a discussion with the researcher for a maximum of 90 minutes. You will be asked to talk about your experience of having coeliac disease and your experiences of talking to professionals in the Gastroenterology service at **the Children's hospital** about difficulties relating to how you are feeling emotionally. This will happen in a private, quiet room at the hospital or at your home, whichever you prefer. The discussion will be digitally audio-recorded.

As the purpose of this study is to understand your experiences from your point of view, we would prefer to meet with you individually during the discussion. However, your parent/ guardian can be present during the rest of the study visit if you would like them to be. If you have any questions or concerns about this, please discuss these with the researcher, who will do their best to help. If you would really like someone to be there during the discussion, then they can join you. However, we would ask them to sit quietly so that you can answer questions. They will be able to share their thoughts at the end of the study visit.

You will not have to answer any questions that you do not want to and will not have to tell the researcher anything you do not want to. You can take a break during the discussion if you would like to, and you can end the discussion at any time. You will be able to withdraw your data up to two weeks after your study visit, at which point data will be de-identified. None of this will affect the care that you receive from the hospital.

What are the possible disadvantages or risks of taking part?

Due to the topic of the study it is possible that you may become upset during the discussion. You will be able to stop the discussion or take a break at any time. The researcher has a psychology degree and experience working with young people with health conditions, which means they have had training in supporting people if they become upset. If the researcher thinks that you are experiencing a high level of distress, or is concerned about yours or **someone else's safety, they will need to tell your parent/** guardian, and your GP. This is so that you can be offered some support to feel better. They might also need to tell other professionals. This is to make sure that you are safe.

What are the possible benefits of taking part?

We hope that by taking part in this study, you will be helping us learn more about the emotional consequences of living with coeliac disease and hopefully improve support for young people who have similar experiences in the future.

Will I be reimbursed for taking part?
You will receive £5 for your time. In addition, if you travel to the hospital purely to participate in the research, then you will receive money to cover any reasonable travel costs.

What if there is a problem?
If you have a concern about any aspect of this study, you should contact the researcher (details below) who will do their best to answer your questions. Alternatively, you can contact the University of Oxford Clinical Trials and Research Governance (CTRG) office on 01865 616480, or the head of CTRG, email ctrg@admin.ox.ac.uk. If you remain unhappy and wish to complain formally, you can do this through the NHS Complaints Procedure. For help with making a formal complaint you can contact the Patient Advice and Liaison Service (PALS) on 01865 221473 or PALS@ouh.nhs.uk.

What will happen with the results of the study?
The results of the study will be used to help the researchers think about how best to support young people with coeliac disease in the service. This research is being carried out by a student, so results will be written up in a report and given to their teachers. Their report will also be available for other students to look at, to help with their research. When the study finishes, we plan to put the research findings in medical magazines that doctors read, and we plan to present the findings at conferences and meetings. Quotes from interviews may be used in the report however these will be de-identified.

If you participate in this study, you will have the opportunity to be sent a summary of results from this study. You will be asked to indicate if you would like to receive a summary on the consent form, in addition to how you would like to receive it e.g. by post/email.

Will my taking part in the study be kept private?
Yes. All of the information that you give during the study visit, including what you say during the discussion, is **called "research data" and this will be kept** private. At the start of the study, you will be given a number. All information that you provide throughout this study will be linked to only this number. Information that may directly identify you (e.g., your name) will be removed from the data so that you can no longer be recognised from it. This information will be kept in password protected audio files and only the research team would have access to these. This data will be securely stored for 5 years at the end of the study. Audio recordings will be transcribed and then deleted. Interview transcripts will be labelled with your participant ID only.

The only situation in which we would share the information you give us with someone else e.g. your parent/guardian and GP would be if you told us something that made us concerned that you or someone else was at risk of serious harm. We would only ever share

the information that was necessary to keep you safe, and we would always do our best to talk to you about it beforehand. This is part of our responsibility to you to make sure you are safe. Responsible members of the University of Oxford and the relevant NHS Trust(s) may be given access to data for monitoring and/or audit of the study to ensure that the research is being run properly.

How will we use information about you?
We will need to use information given by you for this research project. This will include demographic information (e.g. your age) and information relating to your coeliac disease, including:

- How old you were when you were diagnosed with coeliac disease
- Your family history of coeliac disease
- The services that you attend for your Coeliac Disease

People will use this information to do the research or to check your records to make sure that the research is being done properly. People who do not need to know who you are will not be able to see your name or details that identify you. Your data will have a code number instead.

We will keep all information about you safe and secure.

Once we have finished the study, we will keep some of the data so we can check the results. We will keep identifiable information about you for 6-12 months after the study has finished. This does not include any research documents with personal information, such as consent forms, which will be held securely at the **Children's Hospital** for 5 years after the end of the study. We will write our report in a way that no-one can work out that you took part in the study.

What are your choices about how your information is used?
- You can stop being part of the study at any time, without giving a reason. You will be able to ask the researcher not to use your data in this project up to two weeks after you take part in the research.
- We need to manage your records in specific ways for the research to be reliable. This **means that we won't be able to let you see or change the data we hold** about you.

Data protection regulation provides you with control over your personal data and how it is used. When you agree to your information being used in research, however, some of those rights may be limited in order for the research to be reliable and accurate.

Where can you find out more about how your information is used?
You can find out more about how we use your information
- at www.hra.nhs.uk/information-about-patients/
- at https://compliance.web.ox.ac.uk/individual-rights

- by asking one of the research team.

Who is conducting the study?
The Chief Investigator (CI) is Miranda Wheeler, a Trainee Clinical Psychologist at the Oxford Institute of Clinical Psychology Training and Research. The CI is undertaking this as part of an educational project and is supervised by a Senior Clinical Psychologist in the Paediatric Gastroenterology Service.

Who has reviewed the study?
All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee. This is in order to protect your safety, rights, wellbeing and dignity. This study has been reviewed and approved by the HRA and Health and Care Research Wales (HCRW) Approval Research Ethics Committee (REC reference: 19/WA/0323).

Who is organising and funding the study?
The University of Oxford is sponsoring the study and it is funded by the Oxford Institute of Clinical Psychology Training and Research.

What do I do next?
If having read this information sheet you would like to take part in this research, please contact the CI using the telephone number or email address below. The CI will be able to answer any questions you have. If you still would like to take part in this research, we will arrange a time to meet with you.

Researcher contact details:
Miranda Wheeler, Chief Investigator
Email: Miranda.Wheeler@hmc.ox.ac.uk
Phone: 07469432868

Dr Annabel David, Senior Clinical Psychologist
Email: Annabel.David@ouh.nhs.uk
Phone: 01865 234187



NHS
Oxford University Hospitals
NHS Foundation Trust

Lead Researcher: Miranda Wheeler, Trainee Clinical Psychologist
Email: Miranda.wheeler@hmc.ox.ac.uk
Phone: 07469432868

Study Code: *Site ID Code:* *Participant identification number:*

Consent Form for Parents

Study Title: Understanding perceived barriers to the disclosure of psychological difficulties by young people with Coeliac Disease

If you agree, please initial each box

1. I confirm that I have read and understand the information sheet dated 18.11.19 (version 2.0) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐
2. I understand that my **child's** participation is voluntary and that they are free to withdraw at any time without giving any reason, without my **child's** medical care being affected. In this, I understand that I can also request that their data is withdrawn from the study up to two weeks after their interview. ☐
3. I understand that relevant sections of my **child's** medical notes and data collected during the study may be looked at by the University of Oxford, authorised individuals from the research team, the ethics committees, or the NHS Trust where it is relevant to my child taking part in this research. I give **permission for these individuals to have access to my child's records.** ☐
4. I agree to audio recording and the use of de-identified direct quotes in research reports and publications. ☐
5. I understand that if the researcher thinks that my child is experiencing a high level of distress, or is concerned about their safety, they will need to tell relevant professionals e.g. **my child's** GP. ☐

6. I agree to my child taking part in the above study. ☐

Additional:

7. I would like to receive a summary of findings at the end of the study by ☐
email: _____

or by post: _____

_____

_____

_____

_____	_____	_____
Name of Parent	Signature	Date

_____
Name of Child

Miranda Wheeler	M.Wheeler	18.09.20
Name of person taking consent	Signature	Date



NHS
Oxford University Hospitals
NHS Foundation Trust

Lead Researcher: Miranda Wheeler, Trainee Clinical Psychologist
Email: Miranda.wheeler@hmc.ox.ac.uk
Phone: 07469432868

Study Code: *Site ID Code:* *Participant identification number:*

Assent Form

Study Title: Understanding perceived barriers to the disclosure of psychological difficulties by young people with Coeliac Disease

If you agree, please initial each box

1. I confirm that I have read and understand the information sheet dated 18.11.19 (version 2.0) for the above study. I have had the opportunity to think about it and ask any questions if I wanted to. ☐
2. I understand that I do not have to take part in this study if I do not want to and can stop at any time, without giving a reason. If I choose not to take part, it will not affect my medical care. ☐
3. I understand that some of my medical information and what I talked about during the study may be looked at by the University of Oxford, the research team, the ethics committees, or the NHS Trust where it is relevant. I give permission for these people to access my records. ☐
4. I am happy for things I say to be included in the write-up of the research in a way that people would not know who said it. ☐
5. I understand that if the researcher thinks I am feeling very upset, or is worried about my safety, they will need to tell other people e.g. my parent / guardian and my GP to keep me safe. ☐
6. I agree to take part in the study. ☐

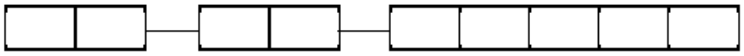
Name	Signature	Date
Miranda Wheeler	M.Wheeler	18.09.20
Name of person taking consent	Signature	Date



NHS
Oxford University Hospitals
NHS Foundation Trust

Lead Researcher: Miranda Wheeler, Trainee Clinical Psychologist
Email: Miranda.wheeler@hmc.ox.ac.uk
Phone: 07469432868

Study Code: *Site ID Code:* *Participant identification number:*



Consent Form for 16-year olds

Study Title: Understanding perceived barriers to the disclosure of psychological difficulties by young people with Coeliac Disease

If you agree, please initial each box

1. I confirm that I have read and understand the information sheet dated 18.11.19 (version 2.0) for the above study. I have had the opportunity to think about the information, as questions and have had these answered satisfactorily. ☐
2. I understand that I do not have to take part in this study if I do not want to. If I chose not to take part, it will not affect my medical care. ☐
3. I know that I can stop the discussion at any time and do not need to say why. I know that I can also ask for my recording not to be used in the study up to two weeks after my interview session. ☐
4. I agree to audio recording and the use of de-identified direct quotes in research reports and publications. ☐
5. I understand that relevant sections of my medical notes and data collected during the study may be looked at by authorised individuals from the University of Oxford, the research team, the ethics committees, or the NHS Trust where it is relevant to me taking part in this research. I give permission for these individuals to have access to my records. ☐
6. I understand that if the researcher thinks I am experiencing a high level of distress, or is concerned about my safety, they will need to tell other people e.g. my parent / guardian or my GP to keep me safe. ☐

7. I agree to take part in the study. ☐

Additional:

8. I would like to receive a summary of findings at the end of the study by email: _____ ☐

or by post: _____

_____

_____

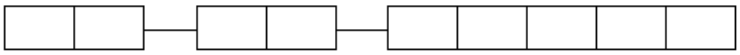
_____	_____	_____
Name	Signature	Date
_____	_____	_____
Name of person taking consent	Signature	Date



NHS
Oxford University Hospitals
NHS Foundation Trust

Lead Researcher: Miranda Wheeler, Trainee Clinical Psychologist
Email: Miranda.wheeler@hmc.ox.ac.uk
Phone: 07469432868

Study Code: *Site ID Code:* *Participant identification number:*



Demographic Questionnaire

Study Title: Understanding perceived barriers to the disclosure of psychological difficulties by young people with Coeliac Disease

Please answer the questions below as accurately as you can. If you don't know how to answer a question, please ask the researcher and they will help. If you don't want to answer a question then you don't have to, just leave it blank.

1. What is your gender?

   Female ☐

   Male ☐

   Other ☐

   Prefer not to say ☐

2. What is your biological sex?
   Girl ☐
   Boy ☐

   Prefer not to say ☐

3. What is your ethnicity? e.g. White/ Black/ Asian _____

4. What is your nationality? e.g. British/ Irish/ Indian _____

Please continue on the next page.

5. How old were you when you were diagnosed with coeliac disease?

   _____Years, _____Months.

6. How old are you now? _____Years, _____Months.

7. What year are you in at school? _____

8. Does anybody else in your family have a diagnosis of coeliac disease?

   Y / N / Don't know / would prefer not to say

   a. If yes, what relation are they to you? _____

9. Do you attend any other services for your coeliac disease? E.g. dietician/GP

   Y / N / Don't know / would prefer not to say

   If yes, please write down which services are involved:

   _____

Thank you for completing this questionnaire.
Please let the researcher know that you have finished.

Oxford University Hospitals
NHS Foundation Trust

Participant Debrief Sheet

Title of Project: Understanding perceived barriers to the disclosure of psychological difficulties by young people with Coeliac Disease

Thank you for participating in this research study. Your participation will help us think about how best to support young people with coeliac disease in the service.

We hope that you have found it interesting and have not been upset by any of the topics discussed. However, if you have found any part of this experience to be upsetting and you would like NHS support, please contact us using the details below.

Researcher contact details:
Miranda Wheeler, Chief Investigator
Email: Miranda.Wheeler@hmc.ox.ac.uk
Phone: 07469432868

Dr Annabel David, Senior Clinical Psychologist
Email: Annabel.David@ouh.nhs.uk
Phone: 01865 234187

Appendix 2.C

Semi-Structured Interview Schedule

Title of Project: Understanding perceived barriers to the disclosure of psychological difficulties by young people with Coeliac Disease

Lead Researcher: Miranda Wheeler, Trainee Clinical Psychologist

[Parent/ guardian present if participant is < 16 years old. If participant is 16 years old, parent/ guardian will not need to be present].

Introduction

Hello, I am Miranda Wheeler, I am a Trainee Clinical Psychologist. Thank you very much for agreeing to talk to me and for making the time to meet with me today. When you were first told about the research, you will have been given/sent an information sheet. Have you read through the information sheet? Do you or your parent/ guardian have any questions about this?

We also need to sign the consent form. Do you/ does your parent/ guardian have any questions about anything on the consent form?

[sign consent form]

You will have also been sent a short questionnaire to collect some more information about your age, background and coeliac history. This information will help me to think carefully about how the service can improve emotional support for young people who might have things in common to you, or might be different to you. Did you receive this and complete it?

[complete demographic questionnaire if they haven't brought a completed version with them, parent/lead researcher provides assistance when necessary]

Do you remember from the information sheet you received that I would like to record what we talk about so that I can write up our conversation word for word? The recording will be downloaded onto an encrypted laptop. This means that only the I will listen to it and, as I said before, all information that could identify you will be removed when it's written out.

Do you have any questions?

[Parent/ guardian leaves for next part of interview if participant is < 16 years].

I'm going to switch on the recorder now and then I'll remind you what we're going to be talking about today.

I am interested in the reasons why young people either feel able or unable to talk to their doctor at the gastroenterology service about how coeliac disease is affecting how they are feeling emotionally and the impact it is having on their life. I am not part of the medical team; I am just doing some research in the service. All information from our discussion that could identify you will be removed when I write up the research, so that your answers remain private. I am interested in hearing both things that made it easier to talk to your doctor and things that made it more difficult.

I would like to ask you about your experiences of having Coeliac Disease and about your experiences of going to see your doctor at the hospital. This should take about 45 minutes. If I use a word or phrase that is difficult to understand just let me know and I can explain what it means.

Before we start, I wanted to ask you what you call your coeliac disease, as I know different people can call it different things, such as 'coeliac's'. What would you like me to call it?

I will now switch on the recorder and ask you my questions.

SIP Interview Schedule:

Introduction questions/ experiences of being a young person with coeliac disease

1. To start with, I would like to find out a bit about you. Tell me about your hobbies or interests?

Prompts: Are you into sport/ music etc.?

2. Can you tell me a bit about what it's like to have [coeliac disease]?

Prompts: Do you think [coeliac disease] has affected your relationships with your family or friends? How? How has it affected your day to day activities or hobbies? Has it caused any problems? What happened? How did you feel? How did you manage?

3. Can you tell me when you first became aware that you had [coeliac disease]?

Prompts*: What happened? What kind of physical problems did you notice? How did that feel in your body? What did you think was happening?

***Here the interview can progress down one of two pathways: 1. If they talk about medical appointments/diagnosis then can move onto 'Experiences with Children's services' after prompts; 2. If they don't continue with current section.*

4. What impact has having [coeliac disease] had on how you are feeling emotionally?

Prompts: How has it made you feel about yourself? Has it ever had a negative effect on how you are feeling emotionally? Has it ever had a positive effect on how you are feeling emotionally?

***** *Here the interview can progress down one of two pathways: 1. If they say it has negatively affected their wellbeing then can skip to 'What is your experience of talking to your doctor about how you feel about coeliac disease?'; 2. If they say it has not negatively affected their wellbeing then continue in this section.*

5. How have your thoughts and feelings about [coeliac disease] changed over time?

Prompts: How has your experience of having [coeliac disease] changed over time, as you are getting older? How important is [coeliac disease] to you? How important does it feel for you?

6. Who have you told about your [coeliac disease]?

Prompts*: Who did you first tell about your diagnosis? How do you tell them? Who did you speak to? How did they respond? What has been helpful about telling others? What has been unhelpful about telling others? Do you think telling others has changed how they see

you? Has telling these people changed things for you? Are there people you can't tell or don't want to tell? Why?

***Here the interview can progress down one of two pathways: 1. If they talk about medical professionals, can move onto 'Experiences with Children's services'; 2. If they talk about friends/family/school staff can continue in this section.*

7. What do you say when you talk to others about your [coeliac disease]?

Prompts: Do you talk to anyone about how it makes you feel? Are there things that you feel worried about talking to others about relating to your [coeliac disease]? How do you feel about telling people about the problems you go through? Does that change? Has that changed over time? Are there things you don't want to talk about with other people?

8. How do you think people with [coeliac disease] are seen by others?

Prompts: How is [coeliac disease] seen by your parents/ family/ teachers/ friends/ healthcare professionals? Do people treat you differently to others? How seriously is [coeliac disease] taken by others? How do you think [coeliac disease] is viewed compared to other health conditions, like a asthma or other allergies (by family, friends, school (peers and teachers) and healthcare professionals)?

Experiences with Children's services

I'd now like to ask you about your experiences of the gastroenterology service, at the Oxford Children's Hospital, can you remember which service I'm talking about?

9. What is/was it like to go to appointments at the paediatric gastroenterology service at the children's hospital?

Prompts: When appointments come up how does that make you feel emotionally? How does/did it feel being there? What does it feel like afterwards? What do you think the appointments are for? Is there anything you like about the service/ think that they do well? Is there anything you don't like about the service/ think that they could do better?

10. What do you think your doctors in the gastroenterology service at the Children's Hospital are interested in hearing about?

Prompts: What is it that makes you think that? What is that like for you?

11. What is your experience of talking to your doctor about how you feel about [coeliac disease] or talking to them about [insert shared difficulties]?

Prompts: How did you let them know how you were feeling? What made this easier? What made this more difficult? What might make you feel unable to talk to your doctor about this? Do you feel more able to talk to certain professionals than others?

12. Thinking about other young people with [coeliac disease], what do you think might make them feel unable to talk about their difficulties (for example, with following the gluten free diet, or with concerns about social situations that involve food, or feeling different to others or feeling worried) to doctors in the service?

Prompts: Do you think your experience of talking to doctors in the service is the same as others?

Service improvements

13. What could the service do to make it easier to talk about how you feel?

Prompt: How do you think that might help?

Ending

14. Is there anything that we haven't talked about that you would like to mention about your experience of having [coeliac disease]?
15. Anything that I haven't asked about that you think is important for me to know related to your experience of the service?

Thank you for taking the time to talk to me today and for talking about your experiences so openly.

[turn recorder off]

[Give participant and parent (where applicable) debrief and allow time for any questions they may have about the study or their participation].

[Reimburse the parent for any travel costs and the participant £5 for their time, ensure signed receipt for payment is taken].

Appendix 2.D

SIP Ethical Approvals

Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

Ariennir gan Lywodraeth Cymru
Funded by Welsh Government

Wales Research Ethics Committee 4
Wrexham

Mailing address:
Health and Care Research Wales Support Centre
Castlebridge 4
15-19 Cowbridge Road East
Cardiff
CF11 9AB

Telephone: 07976 982591

Email: Wales.REC4@wales.nhs.uk
Website : www.hra.nhs.uk

Please note: This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

23 December 2019

Ms Miranda Wheeler
Oxford Institute of Clinical Psychology Training & Research
Oxford Health NHS Foundation Trust, Isis Education Centre, Warneford Hospital
Headington, Oxfordshire
OX3 7JX

Dear Ms Wheeler

Study title:	**Understanding Perceived Barriers to the Disclosure of Psychological Difficulties by Young People with Coeliac Disease**
REC reference:	**19/WA/0323**
Protocol number:	**000000**
IRAS project ID:	**271265**

Thank you for your letter of 18 December 2018, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Confirmation of Capacity and Capability (in England, Northern Ireland and Wales) or NHS management permission (in Scotland) should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for HRA and HCRW Approval (England and Wales)/ NHS permission for research is available in the Integrated Research Application System.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations

Registration of Clinical Trials

It is a condition of the REC favourable opinion that **all clinical trials are registered on a publicly accessible database. For this purpose, 'clinical trials' are defined as the first four project** categories in IRAS project filter question 2. Registration is a legal requirement for clinical trials of investigational medicinal products (CTIMPs), except for phase I trials in healthy volunteers (these must still register as a condition of the REC favourable opinion).

Registration should take place as early as possible and within six weeks of recruiting the first research participant at the latest. Failure to register is a breach of these approval conditions, unless a deferral has been agreed by or on behalf of the Research Ethics Committee (see here for more information on requesting a deferral: https://www.hra.nhs.uk/planning-and-improving-research/research-planning/research-registration-research-project-identifiers/

As set out in the UK Policy Framework, research sponsors are responsible for making information about research publicly available before it starts e.g. by registering the research project on a publicly accessible register. Further guidance on registration is available at:

https://www.hra.nhs.uk/planning-and-improving-research/research-planning/transparency-responsibilities/

You should notify the REC of the registration details. We will audit these as part of the annual progress reporting process.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

After ethical review: Reporting requirements

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study, including early termination of the study
- Final report

The latest guidance on these topics can be found at https://www.hra.nhs.uk/approvals-amendments/managing-your-approval/.

Ethical review of research sites

NHS/HSC sites

The favourable opinion applies to all NHS/HSC sites listed in the application subject to confirmation of Capacity and Capability (in England, Northern Ireland and Wales) or management permission (in Scotland) being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non-NHS/HSC sites

I am pleased to confirm that the favourable opinion applies to any non-NHS/HSC sites listed in the application, subject to site management permission being obtained prior to the start of the study at the site.

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	*Version*	*Date*
GP/consultant information sheets or letters [GP Letter version 1 18.11.19]	1	18 November 2019
Interview schedules or topic guides for participants [Interview Schedule]	1	15 October 2019
IRAS Application Form [IRAS_Form_21102019]		21 October 2019

IRAS Checklist XML [Checklist_16122019]		16 December 2019
Letter from sponsor [Letter from Sponsor]		15 October 2019
Letters of invitation to participant [Parent Invitation]	2	18 November 2019
Letters of invitation to participant [16 year old Invitation]	2	18 November 2019
Non-validated questionnaire [Demographic Questionnaire]	1	15 October 2019
Other [Parents No Contact Letter]	1	15 October 2019
Other [16 year old No Contact Letter]	1	15 October 2019
Other [Debrief Form]	1	15 October 2019
Other [Receipt]	1	15 October 2019
Other [Response to provisional approval table 18.11.19]	N/A	11 November 2019
Participant consent form [Parent Consent Form]	2	18 November 2019
Participant consent form [Assent Form]	2	18 November 2019
Participant consent form [16 year old Consent Form]	2	18 November 2019
Participant information sheet (PIS) [Parent PIS]	2	18 November 2019
Participant information sheet (PIS) [16 year old PIS]	2	18 November 2019
Participant information sheet (PIS) [11 -15 years (tracked)]	2	18 December 2019
Participant information sheet (PIS) [11-15 years (clean)]	2	18 December 2019
Research protocol or project proposal [Research Protocol]	1	15 October 2019
Summary CV for Chief Investigator (CI) [Chief Investigator CV]		15 October 2019
Summary CV for student [Student/CI CV]		15 October 2019
Summary CV for supervisor (student research) [Academic Supervisor CV]		15 October 2019
Summary CV for supervisor (student research) [Field Supervisor CV]		15 October 2019

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website:
http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/

HRA Learning

We are pleased to welcome researchers and research staff to our HRA Learning Events and online learning opportunities – see details at:
https://www.hra.nhs.uk/planning-and-improving-research/learning/

19/WA/0323	Please quote this number on all correspondence

With the Committee's best wishes for the success of this project.

Yours sincerely



Dr Kathryn Ann Clarke
Chair

Email:Wales.REC4@wales.nhs.uk

Enclosures: **"After ethical review –** guidance for **researchers"**

Copy to: CTRG

Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

NHS
Health Research Authority

Ms Miranda Wheeler
Oxford Institute of Clinical Psychology Training & Research
Oxford Health NHS Foundation Trust, Isis Education Centre, Warneford Hospital
Headington, Oxfordshire
OX3 7JX

Email: HCRW.approvals@wales.nhs.uk

Issued 23 December 2019
Revised 14 January 2020

Dear Ms Wheeler

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title:	**Understanding Perceived Barriers to the Disclosure of Psychological Difficulties by Young People with Coeliac Disease**
IRAS project ID:	**271265**
Protocol number:	**000000**
REC reference:	**19/WA/0323**
Sponsor	**University of Oxford, Clinical Trials and Reserach Governance**

I am pleased to confirm that **HRA and Health and Care Research Wales (HCRW) Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, **in line with the instructions provided in the "Information to support study set up" section towards** the end of this letter.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?
HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.

Please see IRAS Help for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?
HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?

The standard conditions **document "***After Ethical Review – guidance for sponsors and investigators***", issued with your REC favourable opinion, gives detailed guidance on reporting** expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

Who should I contact for further information?
Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **271265**. Please quote this on all correspondence.

Yours sincerely,
Tracy Biggs
Approvals Specialist

Email: HCRW.approvals@wales.nhs.uk

Copy to: *CTRG*

List of Documents

The final document set assessed and approved by HRA and HCRW Approval is listed below.

Document	*Version*	*Date*
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Evidence of Sponsor Insurance]		15 October 2019
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Evidence of Sponsor Insurance]	Email confirmation	18 November 2019
GP/consultant information sheets or letters [GP Letter version 1 18.11.19]	1	18 November 2019
HRA Schedule of Events [Validated]	1	01 November 2019
Interview schedules or topic guides for participants [Interview Schedule]	1	15 October 2019
IRAS Application Form [IRAS_Form_21102019]		21 October 2019
IRAS Checklist XML [Checklist_16122019]		16 December 2019
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Organisation Information Document [Organisation Information Document]	1	15 October 2019
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Summary CV for supervisor (student research) [Academic Supervisor CV]		15 October 2019
Summary CV for supervisor (student research) [Field Supervisor CV]		15 October 2019

IRAS project ID	271265

Information to support study set up

The below provides all parties with information to support the arranging and confirming of capacity and capability with participating NHS organisations in England and Wales. This is intended to be an accurate reflection of the study at the time of issue of this letter.

Types of participating NHS organisation	Expectations related to confirmation of capacity and capability	Agreement to be used	Funding arrangements	Oversight expectations	HR Good Practice Resource Pack expectations
One site type undertaking all research activities.	Research activities should not commence at participating NHS organisations in England or Wales prior to their formal confirmation of capacity and capability to deliver the study.	An Organisation Information Document has been submitted and the sponsor is not requesting and does not expect any other site agreement to be used.	No funding to be made available to sites.	It is expected that there should be a Principal Investigator where locally employed staff take responsibility for research procedures.	No Honorary Research Contracts, Letters of Access or pre-engagement checks are expected for local staff employed by the participating NHS organisations. Where arrangements are not already in place, research staff not employed by the NHS host organisation undertaking any of the research activities listed in the research application would be expected to obtaina Letter of Access based on standard DBS checks and occupational health clearance.

Other information to aid study set-up and delivery

This details any other information that may be helpful to sponsors and participating NHS organisations in England and Wales in study set-up.

NHS
Oxford University Hospitals
NHS Foundation Trust
From the Head of Research Governance
OUH Research & Development
Joint Research Office
OUH Cowley

Tel: 01865 223714
Email: Rebecca.Davies1@ouh.nhs.uk
Date: 16/01/2020

Confirmation of Trust Management Approval

Re: Understanding Perceived Barriers to the Disclosure of Psychological Difficulties by Young People with Coeliac Disease
IRAS Reference: 271265
Research and Development Reference: 14574
Research Ethics Committee Reference: 19/WA/0328
Sponsor: University of Oxford

Dear Miss Miranda Wheeler,

On behalf of the Oxford University Hospitals NHS Foundation Trust, I am pleased to confirm Trust Management Approval and Indemnity for the above research as outlined in the application received.

Approved Documents

Protocol Version 1.0 dated 15/10/2019 is recognised as the most current to date.

The documents approved for use at this Trust are as listed in the following approval documents:

Health Research Authority Letter dated: 23/12/2019
Research Ethics Full Approval Letter dated: 23/12/2019

Any subsequent, relevant amendments are additionally included to date.

Metrics and Recruitment

First Participant target	Agreed Total recruitment target
Target date:11/02/2020	Target minimum: 7
	Recruitment Target date 01/06/2020

To comply with national research performance requirements the OUH Trust will monitor and publish recruitment for your study:
1. **Performance of the recruitment of the first participant to your study calculated from the time of receipt of a valid research application in R&D;**
2. **Recruiting planned participants to time and target.**

Signed electronically in Studylive by Mrs Shahida Hussain on 16/01/2020 09:20

NHS
Oxford University Hospitals
NHS Foundation Trust

Recruitment of participants needs to be reported via the Siteline system. Contact your department Siteline administrator to request auserid. If you miss this target you will be required to give reasons that can be reported to the DOH/NIHR.

Study Staff
The CV and relevant training of the PI has been reviewed.

Conditions of Approval
The following conditions apply to this approval:

a) The study is conducted in compliance with all the relevant legislation and the relevant OUH policies.
b) All staff working on the study have the appropriate training and experience and have responsibilities formally delegated to them.
c) Serious Breaches of GCP or the protocol will be notified to the Head of Research Governance within one working day of awareness.
d) Should you plan to leave your position in OUH, you will notify R&D immediately of your plans, to enable discussions around the future of study conduct.
e) All relevant documents will be maintained and will be made available to R&D personnel, to facilitate compliance checks, formal audits and regulatory inspections..
f) You will notify R&D of any subsequent protocol amendments.
g) You will promptly inform R&D of the end of the study.

I wish you every success with the study.

Yours sincerely,

Mrs Shahista Hussain
Head of R&D Governance

Copy to relevant support departments (attach copy contract):

Department / Location	Email
Miranda Wheeler	miranda.wheeler@hrnc.ox.ac.uk
OUH Study Finance	study.finance@nhs.net
Paediatrics	Rebecca.beckley@paediatrics.ox.ac.uk Tamar.Cameron@paediatrics.ox.ac.uk

Signed electronically in Studylive by Mrs Shahista Hussain on 16/01/2020 09:20

Appendix 2.E

Stages of Data Analyses (in Accordance with Smith et al. (2009))

Example of Initial Noting (step 2; Interview 2: Maya)

Initial Noting	Transcript	Emerging Theme
Going over to others houses is hard. Repetition of 'biggest' when discussing problems. Emphasis? Importance of familiar people. Awareness of impact of diagnosis on others. Assumption that others find diagnosis burdensome? – then corrects this to make it hypothetical. Awareness of assumption? 'try and find' – emphasising that catering for GF is challenging. Perception that others find catering for the diet burdensome. Impact on others is the most difficult thing. Others having to make food plans ?or compromises? around her needs is difficult. Importance of friends being familiar with condition.	**P02:** umm. Definitely going over to other people's houses. That was the biggest, especially people that I don't *normally* hang out with or it's just a *one time* I'm going over because *obviously*, they don't wanna really. If they don't wanna go out in their time and get a different, try and find the gluten free things, or. So *that's* probably the *biggest* way that it's affected me is. That. If people are going out to *eat* it's like oh can [P02] come, because. Where, where do we have to eat because [P02] is here and that. But. Once, once my friends get used to it they, they know that, where I can eat and where I can't eat so. **M:** mm. ok, so it's sounds like it's something your friends have had to get used to.	
Friends keep food for them at their house. Importance of friends keeping her (and her CD) in mind. Feeling included?	**P02:** yeah. My friends' got a stack of gluten free chicken nuggets in her freezer for me so (smiling whilst saying) **M:** oh (laughs), that's nice. So like they keep gluten free food for you. **P02:** yeah. **M:** that must. How does that feel?	
Importance of friends (and friends' mums) going out to buy gf food. Social visits involving food are planned. Is it nice not to have to ask? That others keep her in mind? Does this make her feel accepted and/or cared for?	**P02:** That feels good, that to know that. Um, she knows when I'm *coming* and her mum knows she goes out if I'm coming over she'll be like oh she got you a gluten free pizza so that's. it feels nice.	

Note. Pink = Descriptive comments; Blue = Linguistic comments; Yellow = Conceptual comments

Example of Developing Emerging Themes (step 3; Interview 7: Billy)

Initial Noting	Transcript	Emerging Theme
	so how did it make you feel not be able to do that?	
Feeling left-out	**P07:** umm. Well. I felt a bit left out in a way. But I-I did-in errr. December last year I think I went to a party of my quite good friend and they asked	Diagnosis transforming self as different and excluded
Importance of peer support and others' sharing responsibility for diet	their parents to get me a gluten free pizza because they were having pizza so I had-I had um gluten free pizza and I	Importance of peers advocating for dietary requirements
The importance of feeling included in social situations involving food	said-I said like I might be able to have the nachos but I'll have to check with like the counter. Cus the place we went they made sure that they	
Life-stage – more responsibility around diet than expected?	had stuff I could eat and yeah. So I asked the counter and they said they can do the nachos gluten free in a separate like fryer. Yeah, so I was quite happy with that.	YP adopting an adult role in managing diet
	M: fantastic, so it sounds like your friends more recently make sure that where they go for their birthday that you can have something there-	
Importance of others' noticing when they get it wrong	**P07:** -yeah like I-say, on Tuesday my friend offered me a bit of a cookie and then like a few seconds after he said oh no sorry I didn't mean to cus obviously I've told them and told them like how it can affect	Importance of others' holding my condition in mind
A want to communicate the seriousness of the condition to friends	me and yeah. So that made me. Yeah.	
	M: mm ok, and um. When you first got diagnosed, who did you tell about being gluten free then?	
Life-stage – disclosing to adults first then friends.	**P07:** umm. I can't really (laugh) remember. I think it was like my teachers and then my friends but yeah I can't- 100% remember.	

Example of Clustering Emerging Themes (step 4; Interview 5: Sam)

Cluster Label	Emerging Theme	Indicative Quote
1.The need for adults to provide me with a developmentally appropriate understanding of my condition to share with others.	The way a young person understands the consequences of their condition is important in adherence to diet	I think some people mistaken it for coeliac disease and gluten *intolerance* cus when you're gluten *intolerant* um you mainly get bloated and I don't think-I'm not too sure about this, um er it mainly makes you bloated and it doesn't like *harm* your body as much as when you're like fully allergic to it um. err. Yeah when you're like proper coeliac disease…they may carry on eating it thinking that it's not going to be as bad for them as it can be. -23
	Importance of developmentally appropriate explanation of impact of non-adherence to diet	over time the more you eat of those little bits they're just gunna build up to be like a *full* meal of *wheat* and that's *not* healthy. -19
	Barrier to disclosure: lack of developmentally appropriate understanding and explanation of condition	I knew I couldn't have. I couldn't have a certain *piece* of food but I just like, I didn't know what it was so I could say to people that I was gluten free but I couldn't explain to people what it- what I *actually* had. -9**;** my teacher would like explain to some other students maybe if like they tried to hand me something when I was younger. Because obviously I didn't understand so I would go to take it and my teacher would have to explain that I couldn't have it. -10
	Role of parents as educators	like I just *learnt* cus mum obviously taught me. she said you can't have these things. -9; My mum she's had it from when she was

	around eighteen, nineteen so she's had it a while and I guess she's kind of just learnt and she's just taught me about it. And so now I just kind of, kind of know about it a bit more. -28
Developmental stage: role of adult in taking responsibility for disclosure of condition	My *teacher* was the first to *know*. Cus I was obviously like three four ..I guess she was the first to know and I didn't really know what it was so I didn't really say that I had it because I guess I didn't know that I had it at the start. -9; My mum. She would normally like say to my teachers and then if like my teacher would like explain to some other students maybe if like they tried to hand me something when I was younger -10; it's also just like to like remind like other people maybe that you know to just like remember that she can't have this so just like y'know yeah. -42
Young people update their understanding of the condition as they get older	…I didn't really *understand* like what I read on the back of the packet. But as I grew-up I guess I just got used to it...I just think I just *learnt* that you have to just read it before I would eat it. Cus um. y'know my body can't have *certain* things. -5
Challenges of disclosure – mismatch between young person's use of labels and peer understanding	Half the time I say I have coeliac disease and then they just like oh right why have you got a *disease* and I'm like well and then I shorten it so I say I'm gluten free and I guess they've heard that- they've heard that word before and ummm. I guess it kind of like *stops* them. And then they're like oh what so you can't

	have milk? And that's what they *always* say. For some reason but um. no. it's-wheat. -17
Condition 'label' used in disclosure conveys meaning	When people ask me I normally say that I'm gluten free cus it's easier to understand hence the name. but. Normally like when I'm like talking to my friends I just say coeliac disease -1
It's frustrating when others' misunderstand the condition	So like they're like what's coeliac disease and I'm like yeah I'm gluten free and they're like oh you can't have *milk* and *no.* that's like lactose intolerant. I'm not lactose intolerant I'm *gluten free*. I don't know why. Everyone just always asks if it's something to do with milk. -17
Barrier to disclosure: when peers are developmentally less able to hold my emotions and condition in mind	In primary I just wouldn't really *tell* anyone. I just *didn't*. I don't know. I guess I would get *annoyed* but I just didn't *tell* people because they didn't really. I don't *know*. Hmm. -16
Challenges parents have in providing a developmentally appropriate explanation of condition	when I was younger I didn't really understand when mum said I couldn't have it because I couldn't really. I didn't really *understand* like what I read on the back of the packet. -5
Importance of parents providing language for disclosure	mum obviously taught me. she said you can't have these things. So I guess that when people *ask* that's when I could kind of *tell* them because like, I could sort of understand a bit more. -9
Importance of developmentally appropriate understanding and explanation of condition in facilitating disclosure	Most of the time they *ask* what I can't have and I say well I can't have gluten, barley, rye I *think*. Um. just like other parts of the wheat. Um. because if I digest it it can damage my

		digestive system. Um. that's just kind of what I tell them. Yeah.
2. Developmental stage: adults as gatekeepers to diet vs growing autonomy over diet	Importance of teachers sharing responsibility for managing diet and preventing exclusion/distress	teachers they're like really important I guess and it's kind of their *role* and responsibility to like *remember* that y'know like when another student brings in a cake that that specific student can't have it because of this reason -41
	Adults as gatekeepers to food	their mums will like go out and get like brownies if I can't like have what they're having -11
	Importance of parents restoring young person's sense of inclusion and belonging	my dad would like, he would always take me to the shops afterwards when he picked me up and get me a sweet or something special -4
	Changing role of parents in providing support around medical appointments	I remember I *used* to like get a *treat* because I had my bloods taken I would always get something from hospital after like a kinder egg or something. Now it just, it's just something that we just *do, yearly*. And you know we just go we get it done we go home -28
	Overcoming limitations of diet through growing autonomy for diet.	when I go to school and there's only like a few options...so I generally take in like my own lunch to avoid having the same thing every day -2
	Self-responsibility and accountability in managing diet	I know that I stick to my diet and I don't eat wheat -25
	Engaging in valued activity minimises impact of the diagnosis	my hobbies include baking so sometimes I can't make specific things sometimes like bread or doughnuts because you know the yeast doesn't always react um because it's not

		really *wheat* but um no I just make things I bake that I can make which is normally like cake or biscuits 24-25
	Taking self-responsibility for managing diet reduces need for reliance on others	people it's just getting into the habit of always looking at the back of a packet. Or just like always you know just oh are those gluten free before you eat them. Otherwise it's constantly like *asking* other people. Like is this got wheat, barley, gluten etc in -5
	Self-responsibility in protecting health	they're like oh can't you try just a little bit just to see what it tastes like and it's always. It's *no.* because it's not. It's not that simple. -19; after a few times that they've said it it can get annoying. Just like well no I can't have it. -20
3. Lack of personal value in appointments	No personal value in appointments	it just gets annoying because you're waiting so long for them to just say yep you're fine see you next year. -25; I remember going in with my friend and she like looked at the clock and we were in there for like 45 seconds to a minute...they normally just say are you sticking to your diet and I'll say yep and then they're like have you had any vomiting or diarrhoea? No. um have you made any changes to your diet have you eaten any wheat? No. great keep up with the good work see you next year. -26; Sometimes (when i get an appointment) it's just like oh again. -27; I guess it's just habit. Y'know I just go in there, they ask you the same

		questions you answer the same and then its not really much different -31
	Developmental stage determines understanding of appointments	I think they're there to check to make sure that my like body's doing *ok* and that I'm not consuming wheat and I'm I'm sticking to my diet and there's no like problems -28
	Importance of understanding the purpose of the appointment	it just feels *so boring*. Sometimes it can go really quickly like sometimes it takes like 20 minutes but I remember like for a few *years* it did take a good like hour and a half to two hours to *wait* to go in there for like 3 minutes. And for them just to say I'm *fine*. -26
	Role as passive and powerless recipient of care	because I know that I stick to my diet and I don't eat wheat we have to sit there for like an hour and a half or two hours just to like *go in* and for them to like ask me are you sticking to your diet are you doing this, are you doing that and for me to just say and then for them just to say I'm like *fine* and it just gets annoying because you're waiting so long for them to just say yep you're fine see you next year. -25
	Doctor as responsible for setting agenda for what is discussed in appointments through the questions they ask	(why do you think they are interested in that?) Mainly because that's like the only question they ask. Um. they don't really ask much about anything I guess -31
	Perception that doctors have high expectations regarding my management of the condition	I guess that they're just hoping to hear that y'know I'm sticking to my diet and I'm not. Trailing off course because um. my doctor is just like *used* to me y'know staying on my

		diet and not really trailing off and. Y'know the other things -30
4. Importance of peers understanding, accepting, and actively supporting me with my condition.	My peers do not define me by my condition	most of my friends they don't mind they don't really, they don't really care they just look past it -2
	Importance of peers not defining my by the condition	I just guess I told them and they just were sort of like oh cool and then they kind of just dismissed it. -10
	Condition as acceptable to others	when they first meet me they don't (treat me differently), at all. And then when I tell them I guess they just see me as the girl who can't eat *wheat* and that's just y'know, *normal*. But. They don't really treat me differently. -19
	Importance of peers accepting responsibility for diet	I don't really think they mind *at all*. Um. they're kind of like my family it's just *normal*. It's oh P05 is coming over, we gotta get something gluten free. It's never really. A problem. -18
	Growing autonomy over diet results in reduced experiences of exclusion with peers	that was mainly in *primary* school because *now* um obviously we don't really bring sweets in for our whole class because obviously our classes are really big and you know we're *not* always in the same classes as our tutor group um so we don't normally bring things in now but if we *do* it's normally my friend and they normally buy something else. -43
	It is important that my peers see me as more than my condition	My friend group is very good they y'know they don't take it into account, obviously they still *care* but it's not something that they ever

	get annoyed about or mad about um y'know they always like *share* what they can like sweets and stuff -39
Importance of peers sharing responsibility for managing condition/diet	they always like *share* what they can like sweets and stuff they've always check on the back of the packet but I guess that for some people they may not have had that friend group y'know they may have just been completely left out and their friends may not have even made an effort at all. To y'know find them something. -39
Importance of feeling included in situations that could be excluding (around food)	I guess at *first* I was just like y'know like grateful that they *wanted* me to try it and that they were being kind -20
Importance of others' holding my condition in mind	It just feels like they *care* kind of. Like they're not just gunna be like ohh can you have you have this? Oh you can? Here you go they wouldn't like rummage through their kitchen like they'd go out and like get some things specifically -11
Importance of peers holding my condition and emotions in mind.	they kind of understand- but most of my friends would go out and get something especially so I don't really feel it (annoyed) as often as I *used* to in primary -15
Role of peers in providing emotional support	Some of my friends do *ask* like isn't it *hard* to like *have* coeliac disease and I'm just. *Sometimes* it is. -14
Importance of knowing others share emotional experiences (normalising)	I guess they sort of *understand* that it can be really annoying when you like can't have something and you want it because I guess

		everybody's felt that. I just feel it a bit more regularly. -15
5. It's hard when my peers don't understand or respond appropriately to disclosure.	Younger children misunderstand the impact of non-adherence	mainly when we were a bit *younger* and it was just like family cousins and friends like oh can't you just have a little bit to see what it tastes like it's cus they were *young* and I guess they just wanted me to try it but I couldn't. -20
	Experienced stigma	He still finds it *shocking*. That I can't eat KFC. -12
	Peers as problematic broadcasters (using disclosure to magnify difference)	They don't really *understand* like some of the boys in my class. And. I told them that I was coeliac disease and he was like what so you can't have this and can't have that and I was like *yeah*. And then he *still* he's just like ohh she can't have this.-12
	Self-control around how and when disclosure is made is important	it's just like he brings it up with like new classes. Because we have new classes this year and he just like brings it up. And *then* other people ask and it just like upbrings most questions. -13
	It's important people understand the diet is not a choice.	coeliac disease is where y'know you just *can't* eat it like at *all* in comparison to like lactose intolerant. Some lactose intolerant people they normally like they have chocolate bars and they have normal milk just because y'know they *want* to and sometimes it doesn't really *affect* them and I guess they kind of relate it to *that* as like gluten intolerant oh it can't do you much harm. -23

Stigma associated with 'disease'	When you kind of like say oh I have coeliac disease I guess they kind of like freak out a bit they don't know what it is cus you've said the word *disease*. Y'know they're just like oh what's *that?* Why have you got a *disease?* Is it contagious? And stuff. -16; sometimes they're like ohh you've got a disease what disease is it? And they like get a bit. Weird and panicky. Because I've said the word disease and disease isn't always like a good word. -17
Problematic when others don't respond appropriately to disclosure	it can get annoying sometimes when they're like oh can you have this, can you have this, can you have this. -12
Peer pressure to eat gluten reduces over time	I was younger it was *definitely* difficult but now I've just got used to it and people just take it, they don't really ask again and again. -20
Experience of peer pressure to eat gluten	Some people don't take it *as* seriously. They're just like oh. You um. you've got coeliac disease oh can't you just eat a bit of wheat, can't you just try it -19
The process of disclosure temporarily transforms others' perceptions of the self	I guess they're seen as *normal* at *first*. And then. When you kind of like say oh I have coeliac disease I guess they kind of like freak out a bit they don't know what it is cus you've said the word *disease*...And then. When you explain to them and say no it's just like you just can't eat something like its fine then you kind of just understand and then they just. move on. -16

	Disclosure as transforming others' perceptions of the self as 'weird'	they just find it *weird* that I don't like eat like fast food and stuff, like KFC and stuff like that. So they find it really weird. -2
6. Diagnosis when young reduces illness identity	It's all I've ever known	I mean for *me* it didn't really take many months obviously I *grew up* with it. -5
	Importance of peers adjusting to condition alongside me	I guess it's just like I've kind of *grown up* with the friends I like had...And they've just kind of grown up with knowing that I've had coeliac disease. Cus they were obviously there from a young age -10
	Impact of societal shift in increasing range of gf foods important in supporting adjustment and inclusion	*now* I'm just kind of- I'm fine with it. Y'know. I know that there's like *more* now like I can like *go* to a shop and like pick up some cakes -*6*
	Diagnosis when young mitigates challenges of adjustment to condition	It's not really a problem because I've just grown up with it. I guess I've just got used to it. Because it's something that I've just *had*. So It's not something that's like *new* to me. like where I forget to do it. It's something that i just do automatically. -15
	Importance of others in the family accepting responsibility for diet	when we're going over its like oh I need to get something gluten free for P05 and [mum]. It's not much or like oh they're coming over *great.* It's just normal kind of. -18
	Others create safety and shared understanding through experiencing the condition alongside me	because mum already has it and so we're just kind of used to going somewhere and *not* having something or not having a sandwich or y'know going to a restaurant and I guess my *dad*. He, he just kind of like because we have coeliac disease he's just kind of eaten gluten-free food as well -36

	Diagnosis is less visible in safe spaces (and safe relationships)	I guess it's just *normal* (to my family) - 18
	No strong illness identity	I don't really get symptoms. I can sometimes feel bloated, but the only symptom I get regularly is like ulcers in my mouth -4
7. Transforming relationship with diagnosis: from rejection to acceptance and personal growth.	Older and wiser	*Now*. I'm like *fine* in like my older years. -*5*
	Transforming relationship with the diagnosis through process of self-reconciliation	at *first* I *hated* it. I didn't *want it* at *all*. Because y'know I was just *different* from everybody else. Um. but now it's just something that y'know just kind'a I think makes me *me*. -7
	Young people experience different emotions at different stages of adjustment post-diagnosis	it would be different for some people because some people they may not have *learnt* to deal with it and they may still be like *angry* maybe with like their parents if their parents have it they may still be like annoyed and angry that they have it too that they're *different*. -38
	Condition as a companion – carrying the diagnosis with me	I think it's just something like. It just *comes* with me. it's like my characteristic kind of. Like it's always just like oh I'm P05 I have coeliac disease. -8
	Diagnosis as transforming the self as unique	I don't really mind being different. I used to not like it but now I feel like it's a *positive* thing. Like now I feel like I'm *unique* as my grandad would say -37
	Diagnosis as transforming the self as special	like specific school trips I would be like the one person to have like that special lunch. I remember I felt *really* special one time… being gluten free meant that they didn't have

		any sandwiches so I got pizza. And I felt so special and happy and I shared it with my friends, but I just felt so happy in that moment. -24
	Overcoming fear of needles	when I was a bit *younger* I didn't really like it because of the needle but. As I've just grown up a bit I'm just used to it. Cus I used to have a butterfly needle but now I just have the big *adult* ones. So I guess I'm just used to it and then the blood test doesn't faze me -27
	Submitting to condition	I guess it's just something that I can't change so there's no point in *complaining* about it because I can't *change* it -7; I guess it was just me realising that no matter how much I got annoyed about it and how much I was angry about it it wasn't gunna change like, y'know how much I didn't want it it wasn't just gunna go away because I wanted it to. So I guess I just had to like-I just had to deal with it. So over time I just have got used to it. -37
	Personal growth in overcoming fears	once it (blood test) was done I just felt very *proud* -27
8. Barriers to disclosure of emotional difficulties: power, characteristics of HC professional, perceived stigma, perceived seriousness of problems.	Safety important in disclosure	sometimes you just feel a bit more like *safe* talking to somebody about it who can give you like empathy about it. -33; I don't know it's just something like about them they just. Doctors look very serious and some of them look quite like scary because they are very serious so nurses they just look, some of them

	are just very kind and somebody who you'd wanna talk to. -34
Importance of healthcare professional cultivating safety to reduce power imbalance	the *nurse* I think she's a children's nurse still and um. she's not. You know. *Mean*. She's very quick with the needle, she doesn't you know hesitate. She's like very reassuring. Um. she's just very gentle. I think the blood test is pretty good yeah -29
Systemic barrier to disclosure: the hospital environment does not elicit safety	I think that the hospital can be like a really scary place with all these syringes and IVs and like *drips* and the *smell* and the *look* and its like, it just doesn't really *look* you know like a nice place sometimes where bad *things* happen sometimes. It's just not very. Yeah. It's not very nice. -38
Power and safety: Fear of disclosure leading to punishment	for some people it may like um, may make them like say, like if they told that person it may make them feel like *angry* kind of. They may think that if they tell them like they may be angry about it -34
Stigma in disclosure of emotional difficulties	Maybe like sometimes boys might be *embarrassed* to talk about it because it's not seen as manly to talk about your *feelings*. -34
Role of power and position in creating safety	Doctors look very *serious* and some of them look quite like *scary* because they are very serious so nurses they just look, some of them are just very kind and somebody who you'd *wanna* talk to. -34
Recognition of systemic barriers to disclosure (lack of resources)	nurses can be a bit more *laid back* and just a bit more *lenient* I guess. And like sort of

	y'know let you have the chance to just like talk if you need to. -34
Responsibility of doctors in asking how I am coping emotionally (giving permission to share)	Them *asking* that question I guess (would make it easier to talk about) because they never really *ask* so I feel like if they asked maybe more like kids would like come forward and say about it but they *don't* really. As far as I can remember. -32
Previous bad experiences with disclosure can prevent future attempts to disclose emotional distress	when you say it at first they're like oh well it can't be *that* bad because this is way worse or this is way worse and then you may not wanna talk about it because you worry someone else is gunna do that too -41
My emotions don't meet the threshold of seriousness for disclosure to my doctor	*no* I really *haven't* (spoken to my doctor about how I am feeling). mainly just because. I dunno. I never really struggled. It was only like three *days* like in a year where I would get annoyed and so it was never really like a regular thing -31
Doctor's would not understand the psychosocial or emotional challenges posed by the condition	I guess it's just not really something that you talk about with your *doctor* it's more like with your like parents or somebody else who has coeliac disease because they can kind of relate to it more. And they like understand a bit better -35
Doctors' characteristics elicit or reduce sense of safety	Maybe if like maybe if it's like errm. A girl like me and they're talking to the doctor maybe if it was like a *female* doctor because sometimes if you're talking to like the same sex, I don't know, if that makes sense. Like sometimes you just feel a bit more like *safe*

	talking to somebody about it who can give you like empathy about it. 32-33
Doctors' non-verbal communication is important in facilitating disclosure	They may think that if they tell them like they may be angry about it because they don't have that like gentle face, they have a very serious face. 34-35
Experience of feelings being invalidated by others following disclosure	some people when sometimes maybe when you bring it up people may say oh well it's not as bad as this and it's not as bad as that and that maybe like puts somebody *down* so they don't wanna talk about it. -40
Doctors perceived as unapproachable and serious	I feel like the doctors are good but the doctors can be very *serious* and the nurses can be a bit more *laid back* and just a bit more *lenient* I guess. -34
Physical consequences of condition determine how seriously it is seen by doctors	I think it's not necessarily *extremely* serious if like. Like my doctor- we go obviously for a follow-up every year and he can see that I've sticked to my diet and I don't eat wheat and I'm fine so I guess for *him* it's not that serious in comparison to some y'know coeliacs who may eat wheat because they *want* to be normal -22
Doctors are interested in physical symptoms	(are they interested in hearing about anything else?) Maybe just like if I have any new symptoms but I think that's it -30
Discussing emotions is not part of a doctors' role	I feel like people think doctors are more like for medical reasons and if there's a problem not necessarily about how you feel all of the time -37

	Power as barrier to disclosure of emotional difficulties	I feel like sometimes kids feel *shy* to ask. I mean *I* don't but I feel like some children who are maybe a bit younger they may feel shy to ask - 32
	Role of doctor in redressing power imbalance	they may feel shy to ask so maybe if the question is asked *to* them then they may open up a bit. -31
9. The benefits of disclosure via broadcasting: curating understanding and acceptance in others	Educating people through disclosure can reduce stigma	I mean I don't really *mind* I don't mind explaining it. Just so that people like *know*..most of the time I don't mind explaining it to people just so that they know what it is. -12
	It's important others' know my diet is not my choice	I feel like um they could *understand* that it's not like a *choice* that that person has. Like it's not like being vegan or a vegetarian where it's a *choice*, it's something you can't just like take away it's not an *option* and like people do struggle with it and I guess some people don't understand and if they were a bit more educated they might. -40
	Disclosure via broadcasting in different contexts can reduce barriers to talking about the condition	I guess they could sort of *educate* more people, maybe like put up more videos and like maybe like *learn* it in school -39
	Disclosure through broadcasting	I don't *mind* telling *anyone*. If people want to *know* then I'll tell them. -13
	Not the content of disclosure, but the process of disclosure that can be problematic	most people in my year. *Well not most people in my year* but like from past tutors because they like change, most people *know* so. It's not really a problem.-12
	Disclosure as a gateway to inclusion in situations involving food	It's really helpful because pretty much all of my family members like when I'm coming

	over to their house they'll go out and because my mum has coeliac disease too obviously um they'll just go out and they'll buy like some specific sausages or specific cakes or specific bread um for me and mum...even with my friends, like, I go over to my friends sometimes for like sleepovers and stuff and their mums will like go out and get like brownies if I can't like have what they're having -11
Disclosure via broadcasting to educate others' and thus manage others' perceptions of self	Pretty much everyone I know. All my friends. My family members *obviously*. Umm. And just like some of my teachers know, but not *all* of them it's only like when they're just like handing out stuff and like I just say no and they're like *oh why* (said in annoyed voice) and I'm like oh I'm gluten free blah blah blah. But um. mostly all my friends know and they don't *care*. -8
Others' misunderstand the impact and seriousness of the condition as it is not well known	I think because people aren't as *aware* of what it actually *is* and like what it can *do* to like some people's bodies. I don't think they really understand. Because it's not like a really well known disease like asthma or like lactose intolerance…I think sometimes people think oh well it's not that well known so it can't be that serious. And it wouldn't be important. -21
Importance of early disclosure in cultivating acceptance in peer-group	when I *first* met them they would like offer me something and I would say no and it was because I had coeliac so they would know it

		from like really early on in our friendship and then just like as our friendship like *built* they just.- It's just something I've always had. Something that is like-is part of *me.* like oh P05, she has this she has that she has coeliac disease. -13
	Disclosure as a gateway to cultivating understanding and thus safety in others around condition	They may have heard the term gluten-free but I don't think they've ever like *asked* like what it *actually* was. So they just never really *learn* and sometimes yeah people do they're just like oh *what's that* and then they *understand* and then they take it seriously. -21
	Role of self as educator important in reducing stigma	When you explain to them and say no it's just like you just can't eat something like its *fine* then you kind of just *understand* and then they just. move on. -16
10. Difficult social and emotional experiences caused by my condition	Coeliac as transforming the self as different and excluded	when I was younger I *definitely* felt *different* when I was like seven to like nine tenish um because I just felt like everybody else can have like these *cool* lunches and they can like *go* and when there was like bake sales and have cakes and stuff aaand I just felt like I was just like the odd one out like the *one* friend who couldn't have that specific thing. -36
	Fear regarding medical procedures	Mum said that I wouldn't have the butterfly needle and I would have like the *big* needle and I was *scared* because that was like the first time I would have the proper needle rather than the butterfly one that's very thin -27

It's not fair	um I remember one time I got really upset at Rainbows because there was like three girls and it was like their birthdays that week and they like all bought in cake and I couldn't have *any* of it and I was quite upset -4; when I was younger it just annoyed me. because everybody else had something and I couldn't and I would just get upset and annoyed because y'know I couldn't have what they were having -6
Condition can be distressing in situations involving food	When you're like going out to a restaurant or you're trying to find something for lunch like when they don't sell sandwiches or *anything* gluten free so you just have to buy like crisps and some apple slices. To eat. -14
Feeling envious around food	sometimes it was *annoying* just because everyone had like these really nice cakes and like sweets and stuff but I just had like little biscuits -4
Repeated experiences of exclusion around food	everyone bought stuff in and I couldn't *have it* and where like on pancake day I *couldn't have it* and just other like celebrations on people's birthdays in primary school or like when the teachers bought in something and I have to just like say *no*. because I couldn't eat it. -7
Impact of condition on building relationships around food	when the other kids had like birthday cake and they bought in cake and sweets to like *share* when it was like err a celebration or something I could never like *have* it -3

	Condition as transforming the self as different and excluded around food	everybody else had something and I *couldn't* and I would just get upset and annoyed because y'know I couldn't have what they were having. And on like pancake day I couldn't have the pancakes. So I just had to *sit there*. While y'know my teacher ate my pancake -6
	Diagnosis as visible and defining in situations involving food	that's when I was like aware because I couldn't really like have the cakes that teachers bought in and stuff like that -3

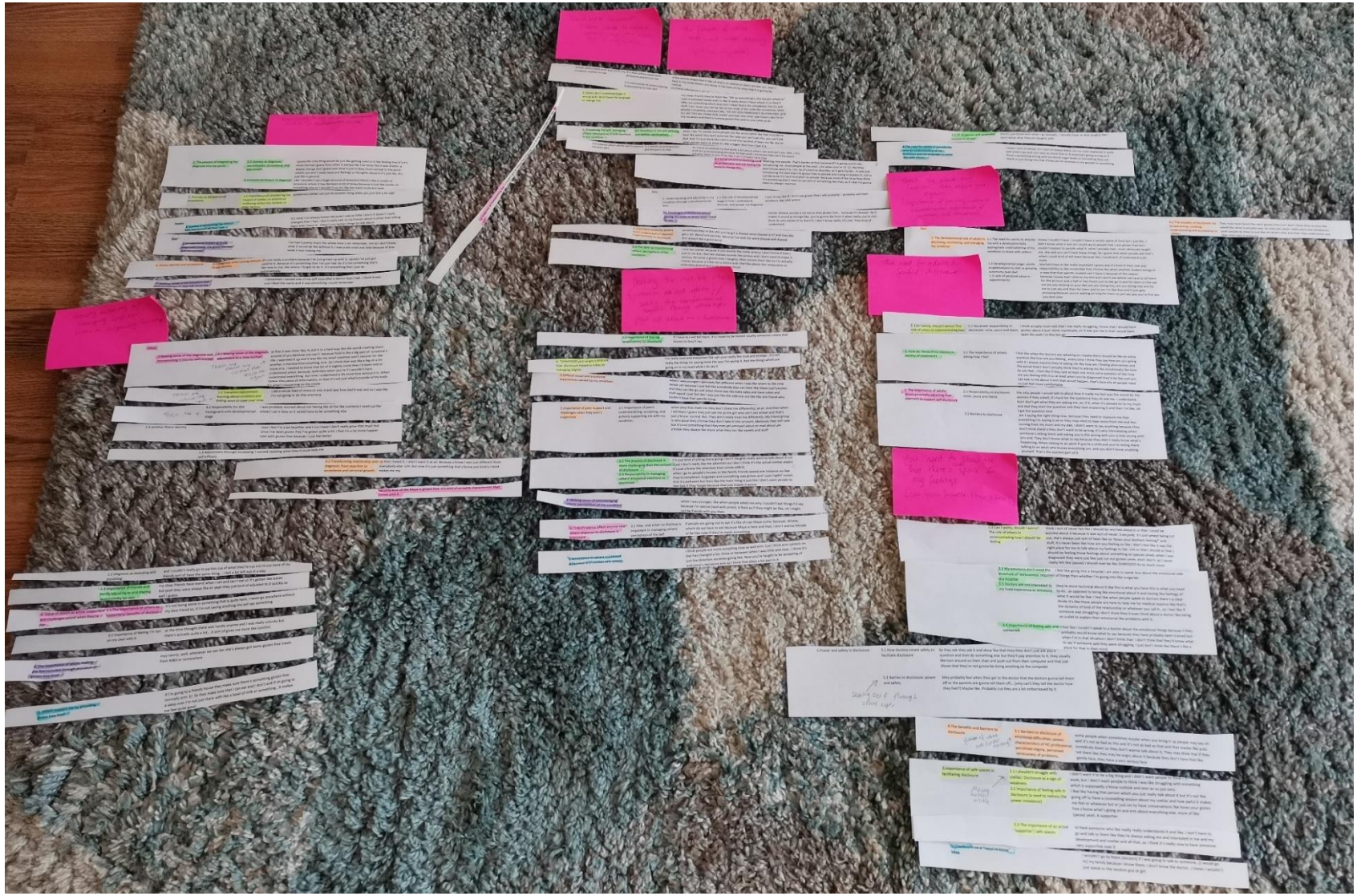
Example of Subsequently Reducing Clusters to Create Master Themes (step 4; Interview 5: Sam)

Master Theme	Cluster Title	Indicative Quote (page number)
1. The developmental role of adults in disclosing, monitoring, and managing my condition	The need for adults to provide me with a developmentally appropriate understanding of my condition to share with others.	I knew I couldn't have. I couldn't have a certain piece of food but I just like, I didn't know what it was so I could say to people that I was gluten free but I couldn't explain to people what it- what I actually had...mum obviously taught me. she said you can't have these things. So I guess that when people ask that's when I could kind of tell them because like, I could sort of understand a bit more. (9)
	Developmental stage: adults as gatekeepers to diet vs growing autonomy over diet	..teachers they're like really important I guess and it's kind of their role and responsibility to like

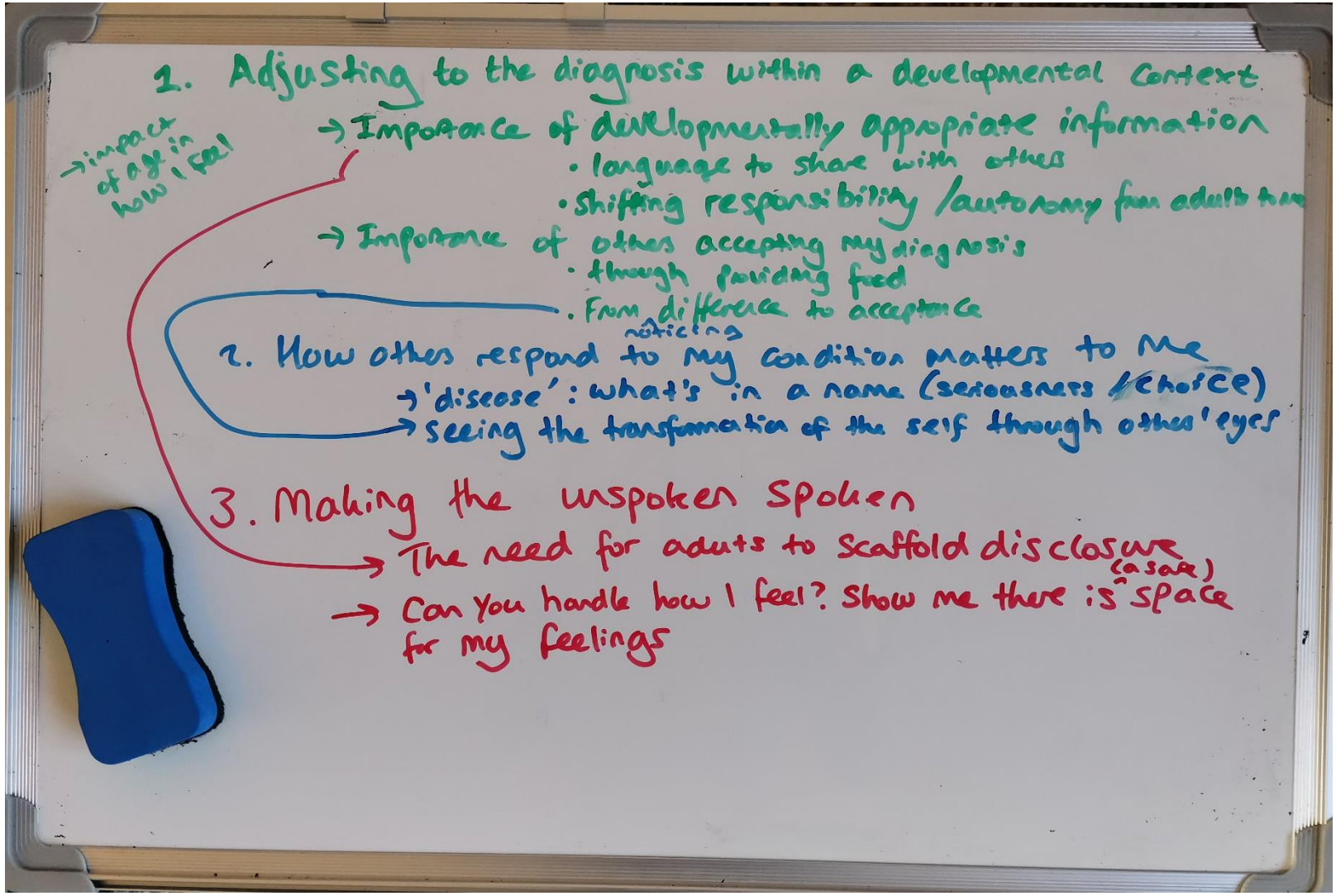
		remember that y'know like when another student brings in a cake that that specific student can't have it because of this reason. (41)
	Lack of personal value in appointments	because I know that I stick to my diet and I don't eat wheat we have to sit there for like an hour and a half or two hours just to like go in and for them to like ask me are you sticking to your diet are you doing this, are you doing that and for me to just say and then for them just to say I'm like fine and it just gets annoying because you're waiting so long for them to just say yep you're fine see you next year. (25)
2.Importance of peer support and challenges when they aren't supportive	Importance of peers understanding, accepting, and actively supporting me with my condition.	when they first meet me they don't (treat me differently), at all. And then when I tell them I guess they just see me as the girl who can't eat wheat and that's just y'know, normal. But. They don't really treat me differently. (19); My friend group is very good they y'know they don't take it into account, obviously they still care but it's not something that they ever get annoyed about or mad about um y'know they always like share what they can like sweets and stuff. (39)
	It's hard when my peers don't understand or respond inappropriately to disclosure.	sometimes they're like ohh you've got a disease what disease is it? And they like get a bit. Weird and panicky. Because I've said the word disease and disease isn't always like a good word. (17)
3. Illness identity and acceptance	Diagnosis when young reduces illness identity	It's not really a problem because I've just grown up with it. I guess I've just got used to it. Because it's something that I've just had. So It's not something that's like new to me. like where I

		forget to do it. It's something that i just do automatically. (15)
	Transforming relationship with diagnosis: from rejection to acceptance and personal growth.	at first I hated it. I didn't want it at all. Because y'know I was just different from everybody else. Um. but now it's just something that y'know just kind'a I think makes me me. (7)
4.The benefits and barriers to disclosure	Barriers to disclosure of emotional difficulties: power, characteristics of HC professional, perceived stigma, perceived seriousness of problems.	some people when sometimes maybe when you bring it up people may say oh well it's not as bad as this and it's not as bad as that and that maybe like puts somebody down so they don't wanna talk about it. (40); They may think that if they tell them like they may be angry about it because they don't have that like gentle face, they have a very serious face. (34-35)
	The benefits of disclosure via broadcasting: curating understanding and acceptance in others	They may have heard the term gluten-free but I don't think they've ever like asked like what it actually was. So they just never really learn and sometimes yeah people do they're just like oh what's that and then they understand and then they take it seriously. (21)
5. Difficult social and emotional experiences caused by my condition	Being the odd one out	when I was younger I definitely felt different when I was like seven to like nine tenish um because I just felt like everybody else can have like these cool lunches and they can like go and when there was like bake sales and have cakes and stuff aaand I just felt like I was just like the odd one out like the one friend who couldn't have that specific thing. (36)

Developing Superordinate and Subordinate Themes (Step 5; grouping Master Themes across transcripts 1 -7)



Developing Superordinate and Subordinate Themes (Step 5; Developing Superordinate and Subordinate Labels based on Groupings)



Final Superordinate and Subordinate Themes with Indicative Quotes (Step 5)

Superordinate Theme	Subordinate Theme	Indicative Quotes
1. "As soon as food started going away it was like oh *ok* this is what it is": Adjusting to the diagnosis within a developmental context	1.2 "Over time it just-it feels more normal": Growing with the condition	*It's what I've always known because I was so little I don't-it doesn't really changed how I feel.* (Finley) *I've had it pretty much the whole time I can remember. Um so I don't know what it would be like without it.* (Eve) *It's not really a problem because I've just grown up with it. I guess I've just got used to it. Because it's something that I've just had.* (Sam) *I guess the only thing would be just like getting used to it like feeling that it's-it's more normal I guess from when it started like I've never felt it was drastic -a drastic change but I guess over time it just-it feels more normal to the point where you don't really have any feelings or thoughts about it it's just like, it's just life in general.* (Anna) *At first it was more like, to put it in a hard way like the world crashing down around of you because you can't- because food is like a big part of someone's life; It was really more when things started changing was when I was aware that, oh right that I can't eat THAT and I can't eat this. So that was the. When I was told it was a bit like, oh ok. But it was as soon as food started going away that it was like oh ok this is what it is.* (Maya) *I would see-if I went shopping I'd see like these coco pop bars that I usually love and I'll think oh they used to be my favourite.* (Billy)
	1.3 "Why can't I just have one?..Will it really have an effect on me?": The need	*I knew I couldn't have. I couldn't have a certain piece of food but I just like, I didn't know what it was so I could say to people that I was gluten free but I couldn't explain to people what it- what I actually had.* (Sam)

	for adults to scaffold my understanding over time	*Err I sort of always knew cus my mum explained it quite well what I can and can't eat; To check that er if anything's going wrong so if there's something wrong with my blood sugar levels or something…I know they take my blood but don't know what they do with it.* (Finley) *I did a whole load of research into it and saw how bad it was and so I was like I'm not going to do that anymore.* (Sarah) *there's just blood tests when I go basically...I actually have no idea (laughs) like I don't know what they are (laughs)… yeah.* (Anna) *[it's] to check how much blood they can get from you without me going pale or something.* (Eve) *..my small intestine won't recover for like another 3 months from anything, erm. So I think that was like a big oh a bit more of a. I needed to know that bit of it slightly more than I'd been told or understood when, because, definitely when you're 9 I wouldn't have understood everything. But now I understand a bit more how serious it is; They told me.. my stomach is like an un-blown up balloon and then when I eat gluten, the balloon blows up. So it gets really inflamed and my body is trying to fight it but it can't. but I just get tired. And that's probably the simplest way, I say the exact same thing every time.* (Maya) *she gave me a bit more information about it and after that I was-I was a bit like- a bit shocked in a way cus the first-one of the first things I saw was that I couldn't have like some biscuits or chocolate but-and I thought there just wouldn't be anything and I thought like. That. Because I can't have that I wouldn't ever be able to have that.* (Billy)

	1.4 "My friend group is very good...they always like share what they can like sweets and stuff": the importance of others creating (a safe) space for me	*If I'm going to a sleep over I'm not just there with like a bowl of milk or something...it makes me feel quite good.* (Finley) *My close friends have learnt what I can and can't eat so it's gotten like easier.* (Anna) *I feel like having that person which you just really talk about it...just um to have conversations like how's your gluten free y'know what's going on and erm about everything else, more of like (pause) yeah. A supporter.* (Sarah) *I think it's normally like my friend who will be like oh Maya can you eat there? I never go anywhere without my best friend so, if I'm not saying anything she will say something.* (Maya) *They're like oh can't you try just a little bit just to see what it tastes like and it's always. It's no. because it's not.. It's not that simple.* (Sam) *I went to, round her house for a sleepover with two other friends and her mum made gluten free eggy bread for me.* (Eve) *Last year I think I went to a party of my quite good friend and they asked their parents to get me a gluten free pizza because they were having pizza.. Yeah, so I was quite happy with that; I feel like it's good because they're making sure that I am eating what I can have and not like (big pause) cookies and biscuits and fish and stuff with like. Yeah, kind of wheat in it and stuff.* (Billy)
2. "It can either be a huge deal or not a deal at all": How others respond to my	2.1 "I don't want to make it serious": The power of labels in mitigating stigma	*Sometimes they're like ohh you've got a disease what disease is it? And they like get a bit. Weird and panicky. Because I've said the word disease and disease isn't always like a good word.* (Sam)

condition matters to me		*I just say coeliac because it just sounds like really serious I don't know if that's just to me but I feel like disease sounds like serious and I don't want to make it serious.* (Anna) *If I say coeliac disease they're like OHMYGOD you've got a DISEASE.* (Sarah) *Coeliac disease sounds a lot worse than gluten free... because it's disease. So it makes it sound as though like, you're gunna die from it when really you're not.* (Eve) *I don't normally call it coeliac disease but the people that I do tell it to I'm like before they say anything you can't get it.. because that's the first thing that comes into your mind it being a disease.* (Maya)
	2.2 "This has kind of been my life": The problem of others co-creating my identity	*A few people responded in like oh that's so rubbish or that's terrible um...then I have in my mind always, you know in the back of my mind that it is gunna be rubbish.* (Billy) *If my friends say Oh Anna is gluten free I (laughs) often correct them like no I'm actually coeliac because it's like not a choice and I feel like places like restaurants or pubs they kind of don't get that that much.* (Anna) *I haven't got an illness that stops me from doing all my hobbies that I like…It's kind of turned to, not a joke, but if its som- if it's food related it will be like, oh Maya is gluten free and it's not taken in a bad way, it's taken in a just a way that I can get past it and I don't mind if they're joking about it*. (Maya) *Everyone has always been like oh its just coeliac or my friends have been like ohh it's just coeliac and-oh I know when I was diagnosed they were just like just cut out gluten umm. And I don't- so I never really felt like (pause) I should ever be like OHMYGOD its so much more.* (Sarah)

		When you say it at first they're like oh well it can't be that bad because this is way worse or this is way worse and then you may not wanna talk about it because you worry someone else is gunna do that too. (Sam)
	2.3 "I didn't want to be different and people to think of me as like this weirdo": Attempts to avoid anticipated stigma	*I don't usually tell people generally at school so.. like I won't tell them, but if they ask I will; They might have thought that I was strange.. they might be like, oh I might not be friends with you then; When I was younger, like when people asked me why I couldn't eat things I'd say because I'm special.* (Eve) *When I was younger I definitely felt different when I was like seven to like nine tenish um.. I just felt like I was just like the odd one out like the one friend who couldn't have that specific thing; He still finds it shocking. That I can't eat KFC; They may have heard the term gluten-free but I don't think they've ever like asked like what it actually was. So they just never really learn and sometimes yeah people do they're just like oh what's that and then they understand and then they take it seriously; At first I hated it. I didn't want it at all. Because y'know I was just different from everybody else. Um. but now it's just something that y'know just kind'a I think makes me me.* (Sam) *I didn't tell my friends for ages and I can remember that. Like not telling them; Sometimes coeliac can just be another thing when you just feel a bit odd; I think I was embarrassed to tell people I didn't want to be different and people to think of me as like this weirdo.* (Sarah) *Obviously in primary school everyone wants to be the same, everyone wants to be following other people. So it's like are they gunna think I'm weird; They'll be like oh Maya with her gluten free, she can't eat anything; They're like oh that's fine and, accepting that I can't eat it and, it's not personal; When I first got diagnosed it was a bit more like well I don't wanna-I don't wanna be this.. but it was quite it was quite interesting in that it was a new thing that no one else had that made me*

		special so, that was also a side to it that- well I have this thing, I'm gluten free. you don't have it. (Maya) *The host, they just like feel bad about it and they kind of, they they make. It. Er like quite a big thing; On the rare occasions when I have been asked to go out to places to eat I just say no cus I'm just. Yeah I'm like very careful about it.* (Anna) *I don't really talk to my friends about it other than telling them that I have it...there's just better things to talk about* (Finley) *I sort of say quite a lot of gluten free food is better than food with gluten in it.* (Billy)
3. "I mean I wouldn't just speak to the random guy or girl": Who is my doctor to me?		*They may think that if they tell them like they may be angry about it because they don't have that like gentle face, they have a very serious face; I feel like going into a hospital I am able to speak less about the emotional side of things.* (Sam) *I definitely myself get nervous when I'm talking to, like, if I'm talking to doctors.. it's very intimidating when someone's sitting there and asking you is this wrong with you is that wrong with you.. When talking to an adult if you're a child and you're sitting there talking to an adult who knows everything um. and you don't know anything yourself. That's the scariest part of it; There's definitely people who have bigger issues than me. so, I don't know if it's more like oh does she need to be here checking up every year when there's people in hospital beds; The only people I would talk to about how it made me feel was the would be the doctors if they asked.* (Maya) *There's a clear divide it's like these people are here to help me for medical reasons.. .so I feel like if someone was struggling I don't think they'd even think about a doctor like being an outlet to explain their emotional like problems with it.. I just don't think like there's like a place for that in their mind; It's kind of like*

		when erm when people just ask are you alright but they kind of move on they don't actually want to know it's just kind of courtesy to do so like it's just polite; I feel like going into a hospital I am able to speak less about the emotional side of things...They're like doctors and they just wanna like- they've got a lot of people to see they just wanna do the session and move on; I feel like when the doctors are speaking err maybe there should be like an extra question like how are you feeling...every time I think they say how are you going with it I always assume they're asking me like how am I finding alternatives and like actual food I don't actually think they're asking me like emotionally like how do you feel. (Anna) *I wouldn't go to them [doctors] if I was going to talk to someone…[I would go to] my family because I know them. I don't know the doctor...I mean I wouldn't just speak to the random guy or girl; I haven't really spoken about how I feel about being gluten free...I don't really feel it's necessary...er I mean it's not something they really need to know.* (Finely) *I-I don't really feel confident enough [to tell them how I feel]..it's probably not the same doctor as like you know with like you go to not [the service] but the normal doctors you get the same doctor so they understand you like quite a lot; They don't just ask you a question and then do something else but they'll pay attention to it..they usually like turn around on their chair and push out from their computer and that just shows that they're not gunna be doing anything on the computer.* (Billy) *I didn't want people to think I'm – not weak, but I didn't want people to think I was like struggling with something which is supposedly y'know outlook and seen as so just easy; I think I sort of never felt like I should be worried about it or that I could be worried about it because it was sort of never. Everyone. It's just wheat being cut out; [the doctor] has always just sort of been like so 'hows your stomach feeling?' and stuff, it's never been like how are you feeling so like I didn't feel like it was the*

		right place for me to talk about my feelings to them. Um or that I should be feeling those feelings about something so (pause) small. (Sarah)

Appendix 2.F

Excerpts from Bracketing Interview

Q: *What areas do you think are important for you to ask about?*

.. I think asking about people's views of coeliac disease, how they interpret how other people understand coeliac disease and how they think other people see them…I also think that because it's a SIP that it's really important for me to ask about their experience of the service, um, what it's been like talking to clinicians about having coeliac and the emotional stuff that that brings up or the difficulties they might have with school or with their friends.

Q: *Tell me about your views on people's views of coeliac disease and where that comes from.*

.. I think that people who have coeliac might think that other people find it annoying or maybe treat them differently and I think for people with coeliac that might be quite difficult because, for them, they might feel like they haven't really got like a really serious condition but also that it's really difficult to cope with it because you've got to go out and see your friends and you want to fit in and so it must be hard for them to sort of, share their diagnosis with people and also not feel like sort of burdensome when they're out with their friends or not feel like they're a pain when they're trying to organise events around food... I think that probably comes from I guess, my partner has coeliac disease, and he was diagnosed when he was at uni. I know that was a big adjustment for him like it was hard to come to terms with not being able to drink beer or eat his favourite foods anymore and erm, I guess I noticed he wouldn't just sort of talk about it with others…if food was involved he would sort of not say anything and then if someone tried to make him eat something then he'd have to be like, oh no no I can't eat that..I think maybe if it was me maybe I wouldn't be so good at coping with that so I wonder whether, particularly for people who are a bit younger and

more erm at a stage where they are worried about what their peers think and about fitting in that that might be more difficult.

Q: *What are your assumptions about what people might say about talking to others at school?*

I wonder whether this will vary according to age. I wonder whether the older they get, maybe the 13,14,15,16 year olds maybe they'll find that harder…thinking about that developmental stage they'll be forming their identity and wanting to fit in and whether they want the coeliac to be part of that.. whereas I think the younger ones, the 11 12 year olds, they might not be quite at the stage where they're really worried about how it could affect how people see them. Erm. Obviously I'm aware of that and I could be wrong!.

Q: *What are your views on what they're going to say about their experience of the service?*

... I honestly don't know what I'll find with that. I think it will be good to hear if people have had good experiences. I think I might be quite surprised if they people have good experiences of talking about their emotions with clinicians because I think in my mind I have this assumption that the clinicians will like sweep it under the carpet or quickly refer it on rather than give time and space to talk about those things. Partly because clinicians are really pushed and partly because they don't really have the experience and skills to talk about things sensitively like that, so yeah. My assumption is that the experiences people have had might not be so validating or not be so good.

Q: *You have talked about your views and experiences with coeliac linked to your partner but it sounded like there are some other views around how able or not able and how comfortable or not comfortable clinicians feel about talking about these things and I'm just wondering how your views are shaped by your experiences?*

That's a really good question. I guess for me personally I've had experiences where I've gone and seen medical professionals about the impact of physical conditions on how I'm feeling where they've been quite dismissive about it or minimised the effects of it, so I guess maybe that's where those ideas come from.

Q: *and slightly linked to that, do you have any biases linked to your experiences of meeting clinicians in the service?*

So I know when I've met them before…for example, when explaining this research they've come back as quite dismissive and said I don't really understand why you're looking at psychological difficulties in people with coeliac they don't really have them, you should be looking at people with diabetes or people with IBS and stuff like that…they were really quite adamant even when I was sort of saying, oh well this is what the research suggests they were really confused about that and again, talking to them about the way I'm doing the research and them asking why is there no control group, where's the control group and not really taking on board what I was saying about that.

Q**:** so that reinforced your views?

Perhaps, but then saying that, on the other side I do have some hopes that this could change because after talking to them a few times they have come around to the purpose of the project and they are interested to know what the findings will be.

Q: *When you've been doing reading around your topic what theories or ideas have you been drawn to and why?*

I've really been drawn to stigma theory and things around that, because I feel like that maybe fits with the preconceived ideas I might have around what I might find, particularly the idea of perceived stigma where the young person feels like they might be treated differently or seen differently but actually in reality they're probably not…I think that maybe

fits quite well with, when I was talking about developmental theories around age and stage and what it means to stand out from your peer group and how threatening that can be when you're a teenager. So those are the ones I'm really drawn to at the moment.

Q: *How do you think your views, beliefs, experiences about coeliac might influence you in the research you are going to do?*

Well I guess it will bias my expectations of what I'm going to find…it might lead me to ask unintentionally more leading question around whether having coeliac makes people feel like they stand out or makes them feel like they're different…I might come across young people who say oh no it doesn't cause me any problems, I don't worry about it at all and so I have to be really mindful of that and I know one of the things that came up quite early when I was thinking about the sorts of questions I wanted to ask around that, which my supervisor pointed out -they suggested having a question around has coeliac ever made you feel really good or really positive and I thought that was an odd question because why would it make you feel good. But we had some discussion around that, around how that might lead people to thinking well actually it makes me feel special because people buy me special food and special treats and so, that really opened-up my own perception of, I guess things beyond my own biases that might come out of the interview too.

Q: *I guess what I'm hearing is that you're expecting to find quite negative things and I can see that you would think that because you're doing research to improve it. But um, yeah I think that's a really important thing to think about the positive side of it as well and that people might have positive experiences of it as well and for that not to get lost. I was interested in whether your professional experiences working with consultants had any impact on your views on them?*

I guess I've always felt with some consultants like they don't take wellbeing as seriously as maybe the medical stuff so like in MDT meetings…they'll focus on the medication and the surgery and stuff…but not the emotional impact.

Q: *And linked to what we were just talking about, do you have any positive experiences working with them?*

Yeah I definitely do and I know some Psychologists I've worked with have built up really trusting relationships with clinicians or consultants where they value their views and ask them about their opinion and they do actually actively refer people to them because they're really interested about their views and they want to help people to feel better.

Appendix 2.G

Excerpts from Reflective Journal

A reflective journal was kept to support researcher reflexivity throughout the research process. This encouraged the researcher to reflect on their preconceptions, beliefs, and assumptions which may have influenced the development of the study, facilitation of interviews, or process of data analysis.

Situating the Researcher

I am a 27-year-old White British Female without any gastrointestinal or autoimmune conditions. One of my main clinical interests is working with people with chronic health conditions. Prior to conducting this research, I have had several years of professional experience supporting both adults and children with adjusting to long term health conditions. With regards to personal experiences, my partner has type 1 diabetes and coeliac disease, both of which were diagnosed during adolescence. My experiences of living with and supporting my partner in managing these conditions led me to develop curiosity around how others may perceive certain health conditions (like diabetes) as more difficult to live with than other conditions (like coeliac disease), and how this does not always reflect lived experience. This has led me to wonder whether others' perceptions of health conditions rest more heavily on the 'biological' consequences rather than 'emotional' or 'social'. This has furthered my interest in how young people adapt to and talk about the biopsychosocial consequences of coeliac disease during adolescence. In particular, how they experience and manage disclosure and the social consequences of this at a time in their lives where 'fitting in' is important, and situations involving food are seldom within their control.

One of my assumptions when commencing this research was that most young people struggle with aspects of both the emotional and social demands placed on them by health

conditions, but that this is often missed by services due to an emphasis on the biological impact and medical management of health conditions. Concurrently, I hold the assumption that young people will perceive that consultants are not interested in hearing about these emotional and social difficulties, which will pose as a barrier to disclosure in this population. This may relate to my own experiences of medical consultations whereby my own feelings were invalidated, and my health was compared to that of someone with a much more serious condition, and this acting as a barrier to both future disclosure and access to psychological support.

Reflection on selecting IPA

IPA seeks to understand and 'give voice' both to the lived experiences of participants, and to how participants make sense of their experiences. Thus, it is a flexible and inductive approach, which seeks to uncover the meanings which experiences hold for participants. In this, there is a focus on the individual and patterns across participants, enabling discussion on convergence and divergence (Smith et al., 2009). This approach enabled participants to reflect on the meaning of their experiences, and it was hoped that this allowed the young people to identify the factors that make disclosure of psychological difficulties easier or more difficult, what could be done differently and what changes could be made to the service to facilitate disclosure of psychological difficulties.

Alternative qualitative methodologies were considered in the development of this project. Grounded theory was considered, which aims to develop an explanatory theory of basic social processes (Starks & Trinidad, 2007). It was agreed amongst the research team that this approach was not appropriate, as the aim of the present study aimed to understand young people's experiences of disclosure of psychological difficulties in medical appointments, rather than to generate a model of this. Moreover, grounded theory requires ongoing data collection until data saturation, which was beyond the scope of this time-

limited project (Glaser & Strauss, 2017). A further considered approach was discourse analysis, which aims to understand how people use language to create and enact identities and activities (Parker, 1992). However, this approach requires naturally occurring data from a range of sources, which was not appropriate within the context of a service-improvement project. A further approach considered was discursive psychology, which is concerned with how people make use of cultural resources and how people talk about their experiences (Smith, 2007). It was agreed that this approach was not suitable as the present study was concerned with the content of young people's narratives when speaking about their experiences over and above how people talk about their experiences. Finally, thematic analysis was also considered, as this too offers an understanding of people's perspectives, however, the researchers viewed this as lacking the depth of interpretation required for the ideographic focus of the present study.

Based on my own research and reading, alongside discussion with supervisors, I concluded that IPA was deemed the most appropriate method to explore young people's lived experiences. I felt both excited and daunted by conducting research using IPA, as I had never conduced qualitative research before. I have previously carried out quantitative research in paediatric health and I was motivated to carry out a qualitative project in order to learn more about this research approach whilst I had the opportunity to be supervised by Clinical Psychologists with experience in this area. I faced some negativity around the use of qualitative methods from the service multi-disciplinary team (MDT). They voiced concerns that the research would lack validity and generalisability due to the small sample size and reliance on interpretation. However, the more I read and learnt about IPA, the more I felt it suited my motivation to understand the lived experiences of others whilst also suiting the aims of the current project and I was able to explain this to the MDT with confidence. I also explained that I hoped that this small-scale project would lead to larger mixed methods or quantitative research projects in this area.

Extracts from the Period of Project Development

Following the research fayre held at the Oxford Institute for Clinical Practice and Training, I felt quite certain that I wanted to build on my previous research experiences in paediatric health. I was initially interested in exploring stigma associated with conditions and the impact on mental health and quality of life. I was particularly drawn to working with my external supervisor due to her existing involvement in carrying out research exploring stigma in inflammatory bowel disease and functional conditions such as irritable bowel syndrome. However, subsequent iterative discussions between me and my external supervisor led to us 'shaping up' the project in the context of it being a service improvement project (SIP). In between these discussions, I carried out a scoping search of the literature and my supervisor carried out an audit of condition-specific referrals to psychological medicine. From this, we discovered that the number of coeliac referrals to psychological medicine were significantly fewer than the prevalence of psychological difficulties experienced by young people with coeliac proposed in the literature. This led me to feel curious about young people's experiences of disclosure of psychological difficulties to medical consultants in the service, and whether perceived stigma may be a mediating factor.

This initial idea was further developed following discussions with the MDT during a team meeting, which involved consultant paediatricians, paediatric gastroenterologists, specialist paediatric nurses and paediatric dieticians. Prior to the meeting, my external supervisor had explained that the consultants are all very psychologically minded and pro-research. However, during this meeting, I was struck by how the consultants appeared disappointed that my focus of research was on the coeliac population. They made suggestions such as focusing on groups such as those with diabetes, who they felt would experience more psychological difficulties. When I explored this further, they described a

tendency to view CD as a condition which is 'easier' to adjust to and live with than other gastrointestinal conditions, and consequently did not ask directly about psychological wellbeing during appointments.

Following this, myself and my supervisors considered interviewing the paediatric gastroenterology consultants to understand their experience of the young people reporting difficulties and their experience of trying to identify those with a psychological need. However, there are only three paediatric gastroenterology consultants in the team so this would have involved going outside of the service, which would have fallen outside the scope of the SIP. In addition, I felt more motivated to explore the lived experiences of the young people themselves, as based on the sparse literature in this area, I felt their stories are often 'unvoiced'. Therefore, we agreed that it would be helpful to focus on the experiences of barriers to disclosure of psychological difficulties within the service. We spent time in supervision thinking about how this could lead to service improvements. For example, by prompting training on perceived stigma in young people to medical consultants. Or by introducing a psychological wellbeing screening tool in coeliac clinics, to identify those who may benefit from psychology input.

It was agreed that one-to-one interviews would be the preferred method of data collection in the present study, as this would ensure a detailed and extensive investigation into clinical experience (Smith et al., 2009). In line with IPA principles of maintaining a rich idiographic account of each individual whilst allowing convergence and divergence of themes across transcripts, it was agreed that the present study would aim for approximately 7 -10 participants aged 11-16 years to limit heterogeneity of the sample regarding life stage (Smith et al., 2009). The service database was consulted in the development of exclusion/inclusion criteria, to ensure that criteria was inclusive enough to represent the demographics and clinical characteristics of young people under the care of the service, whilst concurrently ensuring that criteria was refined enough to ensure that a rich

idiographic account of each participant could be obtained. It was agreed that young people at a similar life stage (secondary school) would ensure homogeneity in understanding participant experiences. Moreover, it was felt that participants would need to have had the diagnosis for at least 3 months and have had the opportunity to experience at least one follow-up appointment to ensure that they had experiences to draw upon in during the interview. Young people with co-morbid gastro-intestinal conditions were excluded following discussions with the MDT, which highlighted a distinction between their approach to consultations with young people with coeliac vs young people with other gastrointestinal conditions. Similarly, young people currently receiving psychological support from any psychological service were excluded as the research question was particularly interested in barriers to disclosure.

On Friday 31st May 2019, a project approval session was held whereby the research proposal was presented to a course tutor and a research tutor. The purpose of the session was to get feedback on the research proposal at an advanced stage, to ensure methodological rigour and consideration of ethical issues. I experienced this to be a very supportive process, which enabled me to further my own understanding of the importance of quality criteria when carrying out a qualitative project.

Extracts from Developing the Interview Schedule

In planning this project and considering questions to explore with young people, a consultation was held with a 14-year-old male with a diagnosis of coeliac disease, under the care of the service. Advice was sought from him about the topics that may be important to include in the interview schedule, and the wording of questions to ensure they were age appropriate. This involved reviewing a list of words such as ‘wellbeing’ and ‘disclosing’ to assess their understanding of these words. Consultation was also held with two supervising Clinical Psychologists. Feedback on draft interview schedules led to changes in the wording

of questions, the order in which they were asked, additional prompts, and topic areas whilst adhering to guiding principles of IPA methodology. A bracketing interview established my prior assumptions and biases, to attempt to set these aside throughout the research process.

As I developed the research schedule, I found it quite difficult to keep my questions open and not make them too leading. In particular, I was interested in hearing about a broad range of experiences at home, school and over time and I had to ensure that I didn't lose sight of the aims and objectives of the SIP. Supervision was particularly helpful in this respect. It was helpful to consult with those who were less familiar with the condition, such as my internal supervisor and peers as they made suggestions which helped me to reflect on my own biases. For instance, my supervisor suggested having a question around whether coeliac has ever made the young person feel good or positive. I remember thinking that was a strange question and that led me to reflect on my own biases of it being 'all bad'. Following an iterative process of discussions with supervisors, I felt quite happy with my final list of questions which broadly ask about both young people's experiences of living with the condition more broadly, and their experiences of the service more specifically.

Extracts from Seeking Project Approvals

Obtaining course approval through a project approval session was relatively straightforward and an altogether positive experience for me. I found it quite nerve-wracking to present my project to the course team, but found it really reassuring and motivating to receive positive feedback about the project. It was also really helpful of them to suggest that I spend more time reflecting on quality checks for qualitative analysis. Putting the project through the REC was a more difficult experience. I found it took far longer than I had expected, which frustrated me because I had put a lot of work into getting the project going quickly so that I would have more time to recruit. I found the process of presenting the project at a REC meeting anxiety provoking and was not expecting to hear some of the

concerns raised, such as their worry that I would recruit very quickly and have to turn some young people away. I was frustrated that I had to do some amendments and re-submit as I did not feel it was a particularly ethically controversial topic and I knew of colleagues doing very similar topics, who did not need to go through full NHS ethics. Despite this, it was very rewarding when I did get my approvals in place and I learnt a lot about the process of seeking ethical approval in the NHS.

Extracts from the Process of Recruitment

Despite waiting a long time to get ethics approvals in place, I found that recruiting through the team was slow to start. I feel this is perhaps a reflection of the business of services and the fact that I was not in the team and able to steer clinicians towards actively recruiting on my behalf. I found that this experience enabled me to develop confidence in my own assertiveness and it taught me the importance of clearly setting out expectations early on.

Unfortunately, one month into recruiting, the pandemic started in the UK and the Trust placed a freeze on all research activity. I spent a lot of time trying to find a way to amend the project in a way that would enable me to continue recruiting e.g., through not using Trust resources, but I faced what felt like relentless barriers. This led me to feel very hopeless and frustrated that the project may not be able to continue. This experience enabled me to empathise with qualified Clinical Psychologists who often feel reluctant to continue research once qualified, due to the complexity of the ethics process and the time it takes to do this, alongside the unpredictable barriers that can arise. I felt that the most challenging aspect of this experience was that I was doing it in a time-restricted context to be able to submit it as part of my thesis. I feel like if I did not have this pressure, I wouldn't feel as frustrated by the barriers. There were points where I wondered whether it would be more productive to put this project to one side and start a new one which did not involve

recruiting through the Trust, however I have always been very passionate about the purpose of this project and felt like I owed the completion of this project to those who had already taken part.

I was initially really pleased with how the pilot interview went and really surprised by how quickly time went. I felt that I had left with a good depth and breadth of information. However, upon listening to the interview back the same evening, I felt slightly disappointed at the 'missed' opportunities to explore interesting ideas further. Reflecting on this in supervision was very helpful in reassuring me that I would likely never capture everything I wanted to in an interview. As I started to transcribe the first pilot interview, I realised that there had been situations where I hadn't quite used the same words as the participant, or had been slightly leading in my questions. Discussion with my internal supervisor around how to develop my interview style helpfully enabled me to adapt my approach for the second interview, in keeping more closely with the practice of IPA.

Sarah's interview (Example of coming away feeling pleased following an interview):

I felt quite anxious during the first interview, which I carried out straight after a clinical session on placement. It felt difficult making a shift from 'clinician' to 'researcher' and I felt I had to be particularly mindful during the interview to ensure I maintained this positioning. During the interview, I found myself feeling surprised that the young person was so open with how coeliac makes them feel and with how eloquently they were able to describe their thoughts and feelings without much prompt from myself. I surprised myself too by feeling quite moved at points during the interview, particularly when the participant was talking about worrying that coeliac wasn't an important enough reason to see a psychologist or how something so small could make her feel so bad. I was struck by how brave she was in telling me about that. I was really impressed by the way that the participant was able to talk about wanting health care to be more integrated in the context of my own

awareness of how this relates to the NHS long term plan. On one hand, I was left wondering whether Sarah was able to talk so openly due to her experiences of having psychology input. However, I was surprised that she described feeling that the Psychologist would judge her too and I think this perhaps made me steer away from asking more questions about that. I need to be more aware of that bias and explore that more because it could be really important.

Eve's interview (Example of coming away feeling anxious and deflated following an interview):

I came away from this interview feeling more anxious and deflated. I was worried that I had upset Eve with my questions, as halfway through the interview, she started to fiddle with toys and made less eye contact, whilst looking sad. Seeking supervision on this and making contact with Eve the following day to ensure she was ok was helpful in putting my own worries to rest. The interview was also a lot shorter, and Eve provided very brief answers to questions. I recognised that there was a part of me that felt disappointed by the participants' difficulties expressing her thoughts and feelings, even though, logically, this is completely normal for a girl of her age and developmental stage! When reflecting more on this, I had the thought that another interesting outcome to the SIP could be that young people with coeliac actually don't want to talk about their feelings or don't think about them very much. There were still some interesting things that she said about worrying about being 'odd' or people not wanting to be friends with her. I wonder whether there is something about being diagnosed really young and it forming part of your identity as you grow, as this is perhaps what sets her apart from others.

Extracts from the Process of Analysis

I really enjoyed the process of analysing transcripts. I found the process of transcribing and listening back to interviews very helpful in enabling me to think about what

was said without the pressure of having to formulate a response ‘in the room’. This process already started to prompt ideas and notes for coding and I tried to type these onto the transcripts as I listened. It took me a while to get used to the different codes ‘descriptive’, ‘linguistic’ and ‘conceptual’ and I often felt like I was getting them mixed-up. I found supervision helpful in enabling me to take a ‘birds-eye’ view of analysis and think about the meaning and sense-making behind the words used by the individual.

I found the process of drawing out emerging themes from each transcript challenging and very uncertain. I ended up with 66 emergent themes for my first transcript, which led me to feel quite overwhelmed and question how I could possibly make some kind of sense of the data. I initially started working on the computer using an excel sheet, however I found that this was limiting and found that writing down emergent themes on post-it notes and clustering emerging themes into categories first, before transferring this to the computer much easier. I noticed how some themes could fall into several categories and others didn’t really fit into any category. I was worried that I would lose some of the depth in analysis by clustering and this made me very cautious in pulling together themes. I also questioned whether I was at times trying to fit quotes and emerging themes ‘into a box’ by neatly clustering them and supervision was helpful in exploring this.

Whilst identifying emerging themes for P03 (Eve), I found it hard to classify the quote “When I was younger, like when people asked me why I couldn’t eat things I’d say because I’m special” – 9. Initially, I coded this as ‘diagnosis as a gift’, but when compiling my themes, I noticed that Eve did not have the understanding or the language to disclose the condition to others and whether this was her way of ‘finding her own language’ to do this, in a context of wanting to ‘fit in’ with others. I felt this was probably more accurate given that she spoke about not being allowed to have certain foods and feeling jealous etc.

I initially clustered emerging themes using abstraction, by grouping similar themes together and coming up with a cluster title. There were some emergent themes that felt really relevant, such as ‘how to disclose is important’ and this became a cluster label through the process of subsumption. I noticed some polarization within themes, for example, ‘coeliac defines me vs I am more than my coeliac (it does not define me)’. I developed this into the cluster ‘relationship between the diagnosis and the self’. I also used contextual grouping by considering life stage and how this may impact the perception of ‘difference’ and perceived stigma. I did not group themes according to ‘function’ quite so much, which I found interesting to reflect on as I had received initial feedback from supervisors to suggest that I was coding transcripts in a way that bordered on ‘discourse analysis’ by focusing very heavily on the use of language and impact of this on the positioning of the participant. This made me wonder whether I had intentionally directed myself away from this method of connecting emergent themes, in an effort to ‘do IPA’.

Extracts from Clarifying Themes and Clusters with Supervisors

During a supervision session with my external supervisor, we had a helpful discussion about my clustered emerging themes for participant 6 (Anna). Specifically relating to attending appointments and her interactions with professionals:

We thought about the powerless position of attending hospital (not just the power dynamic between patient and doctors, but how power can come from a different level of the system). For example, the participant indicates an awareness that the NHS is under-resourced, with long waiting times and over-worked staff. I reflected on my own positioning and tendency to align with the patient through wanting to make things better for them, and how I had noticed that my initial reaction was not wanting to acknowledge how the wider hospital system may be impacting on doctors’ approach to consultations because this may take away a sense of responsibility in them for doing things differently. This discussion was

helpful in enabling me to be mindful of bracketing this and not letting it bias my themes and clusters.

We discussed how the participant talks about poor communication between herself and the doctors she has seen and considered how this may link to doctors inadequately adapting their communication style in response to different developmental stages (particularly when considering that young people of different ages process information in different ways). From this, we thought about sharing with the doctors the need to re-explain the condition and purpose of appointments in different ways at different stages of the same patients' development.

During a further supervision session with my internal supervisor, we reviewed how well the master themes and cluster labels for P07 (Billy) fit with the raw data in the transcript. We spoke about how the clustered themes 'the importance of others sharing responsibility for the diet' and 'importance of feeling that I'm not on my own with it' may both be underpinned by core stigma models and the concept of 'broadcasting' whereby the young person seeks not only to tell people about his diagnosis, but to educate them and bring them 'onto his side' so that they 'follow him' (and literally eat his gluten free food with him). We discussed the importance not only in him perceiving that others validate his experiences, but in how they respond to his disclosure by 'journeying alongside' him. This led me to think about how superordinate themes may come together across transcripts, underpinned by models of disclosure and stigma.

We then spoke about the clustered theme of doctors creating safety to facilitate disclosure. I thought about how he used the term 'put words into their mouth' in a positive way and we thought about how this young person is trying to communicate that "I find having cues helpful". This could possibly take the form of having pictures of faces or emotion word lists. We also considered the importance of the message: it's important that

professionals create space to talk about feelings. This also led me to think about the powerful position that doctors hold as ‘experts’ and take in leading the session and setting the agenda.

We thought about the cluster label ‘importance of shared understanding in facilitating disclosure’ and the indicative quote not quite fitting with this emerging theme label. We thought about how the quote “it depends like who-it depends whether they understand it or not cus there’s probably some professionals that don’t understand it as well as other professionals and don’t really-not-not-not they don’t care but that they don’t really, like, give-give much attention, not attention, like give much (big pause) erm like feedback”. 32-33) may be a reflection of him being careful about what he says to me about professionals. We thought about how it may be his way of communicating that some professionals don’t have the same level of knowledge as others about the condition, perhaps telling us something about team cohesiveness.

When I first took ideas for superordinate and subordinate themes to supervision, it was helpful to receive the feedback that I needed to think more theoretically about what each theme is saying by ‘stepping beyond’ the text. It was helpful to spend time reflecting on relevant development, disclosure, and stigma theory and existing literature to think about what each theme was trying to convey. This time in supervision bolstered my confidence in ‘trusting the process’ of IPA and enabled me to repeat the process of clustering master themes to explore other interpretations of data.

Extracts from Writing Up

Prior to beginning the write-up, I was mindful that I tend to go over word counts with ease. I found it helpful setting myself limits for each section of the write-up in ensuring I wrote succinctly. I felt apprehensive about writing the results section, having never done an IPA write-up before. However, I found that using the order of superordinate themes to

structure the results/discussion section helpful and the process of choosing quotes and weaving an interpretative narrative around these rewarding, following the long and time-consuming process of analysis.

During the process of writing, I found that the subordinate theme, ‘"They start explaining it and then I’m like, oh I get the question now": The need for adults to scaffold disclosure’ began to fade into the theme, ‘"Why can't I just have one?..Will it really have an effect on me?": The need for adults to scaffold my understanding over time’. This led me to discard the former theme and consider how quotes may relate to the need for adults to scaffold information, understanding and disclosure more broadly.

Appendix 2.H

Lay Summary of Findings

Dear [name of participant],

Thank you again for taking part in our study on understanding what it is like for young people to have coeliac disease and understanding why young people do or do not tell their doctor at the Paediatric Gastroenterology service about how having coeliac disease makes them feel. Here is a summary of what the study found.

What We Did



We wanted to understand what it is like for young people to have coeliac disease. We were interested to find out why young people may nor may not tell their doctor about how having coeliac disease makes them feel during their appointments. We used interviews to ask questions about things like how young people talk about coeliac disease to friends, family, and healthcare professionals. We also asked about how having coeliac disease makes young people feel in different situations, who they talk to about it, and what makes it easier or more difficult to do this.



Who Took Part

Seven young people took part in this study. These were five girls and two boys, aged between 11 and 16 years. Everyone who took part had a diagnosis of coeliac disease and had attended appointments for this at the Oxford Children's Hospital.

What We Found

Young people were very good at speaking about their experiences. They told the interviewer about their experience of having coeliac disease and what talking about it to others was like for them. There were three main themes that people talked about. These were:

1. It is common for young people to feel lots of different emotions related to having coeliac disease:



- Young people described feeling sad, angry, confused, annoyed, frustrated, jealous, left out, unique, and special, amongst other things, in response to having coeliac disease and having to eat a gluten free diet. Some young people who were diagnosed with coeliac disease when they were very young found that they did not miss foods containing gluten, because they could not remember eating them. Some young

people who were diagnosed when they were older found that they did miss foods containing gluten and eating a gluten free diet took time to get used to. Some young people described that this got easier over time, as they got used to the condition and the diet and it started feeling 'more normal'.

- Young people described having parents and doctors who could explain the condition and diet to them in a way that they could understand as very important. Some young people said that they did not remember everything they were told when they were first diagnosed, and it was helpful for adults to repeat this information over time. This included information about the types of food that they can and cannot eat, why blood tests are done and what the results mean, and how coeliac disease can impact the body and cause different symptoms. Lots of young people talked about wanting to be able to understand these things so that they were able to explain them to other people.



2. Young people described that it was important that their friends and family were supportive, provided them with safe foods, and didn't draw additional attention to them when they told them about having coeliac disease:

- Young people spoke a lot about friends being really important. They talked about how it was helpful when their friends made sure that there was going to be gluten free food available at parties and sleepovers. Some young people described finding it hard when their friends offered them food that was not gluten free, or asked them what gluten was, because they did not always know what to say.



- Some young people explained that it can be hard to decide what to call the condition when talking to their friends. For example, some young people felt that the word 'disease' could make the condition seem more dangerous or life-threatening than it is. Some young people said that calling it coeliac 'disease' can also make other people react in a way that is unhelpful.

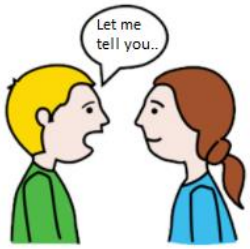


- Lots of young people spoke about wanting other people to take the condition seriously and provide safe foods, whilst also wanting to avoid drawing lots of attention to themselves which they worried could make them appear 'different' to others.

- Lots of young people said when they told their friends about the condition, they responded to this in a helpful way by providing food and support, or by just accepting it and moving on. Some young people said that others responded in an unhelpful way, by making comparisons to other conditions e.g., lactose intolerance, making a 'big deal' out of it, or guessing what they could and couldn't eat without checking first.



- Young people coped with these challenges in lots of different ways. Some people avoided telling others about their coeliac disease, or avoided situations that involved food. Others talked to others about the benefits of having coeliac disease and how it made them unique and special. Some young people decided that educating others about the condition helped them to understand it more and take it more seriously.



3.There are lots of reasons why young people might find it easier or harder to talk to their doctor about how they feel about coeliac disease:

- Some young people described thinking doctors are serious or intimidating and wondered whether they would understand what to do with emotions if they are spoken about. Young people said that these beliefs could make it harder for them to talk to their doctor about how they are feeling emotionally.



- A lot of young people questioned whether their feelings were something the doctor wanted to hear about and whether their feelings were serious enough to talk to doctors about.

- Lots of young people felt it could be helpful for doctors to ask them specific questions about how they were feeling to show them that this is something that they could talk about during appointments.

<u>What This Means</u>



We have learned some new things about young people with coeliac disease from these findings. Healthcare professionals who work with young people with coeliac disease can read this. It might help them to understand what can be challenging about having coeliac disease and what young people would like support with. It might give them ideas about how to help, like giving more information about how to talk about the condition and diet with friends, or by asking a question about how the young person is feeling. This will go in a report to be published in a journal.



If you would like to talk to me or my supervisors about this, or have any more questions, you can contact us using the details below. Thank you very much for your help.

Miranda Wheeler, Chief Investigator
Email: Miranda.Wheeler@hmc.ox.ac.uk

Dr Juliet Kennedy, Senior Clinical Psychologist
Email: Juliet.Kennedy@ouh.nhs.uk



Appendix 3.A

MRP Instructions for Authors

To assist the reader, all tables and figures have been integrated into the main text, as

opposed to submitted separately.

COVID-19 impact on peer review
As a result of the significant disruption that is being caused by the COVID-19 pandemic we understand that many authors and peer reviewers will be making adjustments to their professional and personal lives. As a result they may have difficulty in meeting the timelines associated with our peer review process. Please let the journal editorial office know if you need additional time. Our systems will continue to remind you of the original timelines but we intend to be flexible.

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Please ensure that all research reported in submitted papers has been conducted in an ethical and responsible manner, and is in full compliance with all relevant codes of experimentation and legislation. All papers which report in vivo experiments or clinical trials on humans or animals must include a written statement in the Methods section. This should explain that all work was conducted with the formal approval of the local human subject or animal care committees (institutional and national), and that clinical trials have been registered as legislation requires. Authors who do not have formal ethics review committees should include a statement that their study follows the principles of the Declaration of Helsinki.

Consent

All authors are required to follow the ICMJE requirements on privacy and informed consent from patients and study participants. Please confirm that any patient, service user, or participant (or that person's parent or legal guardian) in any research, experiment, or clinical trial described in your paper has given written consent to the inclusion of material pertaining to themselves, that they acknowledge that they cannot be identified via the paper; and that you have fully anonymized them. Where someone is deceased, please ensure you have written consent from the family or estate. Authors may use this Patient Consent Form, which should be completed, saved, and sent to the journal if requested.

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Please include all relevant safety precautions; and cite any accepted standard or code of practice. Authors working in animal science may find it useful to consult the International Association of Veterinary Editors' Consensus Author Guidelines on Animal Ethics and Welfare and Guidelines for the Treatment of Animals in Behavioural Research and

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Queries

Should you have any queries, please visit our Author Services website or contact us here.

Appendix 3.B

MRP Ethical Approvals for Primary Studies

NHS
Health Research Authority
NRES Committee London - Camden & Islington
North East REC Office
Room 002
TEDCO Business Centre
Rolling Mill Road
Jarrow
Tyne & Wear
NE32 3DT

Telephone: 0191 4283545

08 April 2014

Prof Glyn Humphreys
Watts Professor and Head of Department of Experimental Psychology
University of Oxford
Department Of Experimental Psychology
Tinbergen Building
OX1 3UD

Dear Prof Humphreys

Study title:	**Improving diagnosis of cognitive problems: validation and reliability of computerized cognitive screening tools**
REC reference:	**14/LO/0648**
IRAS project ID:	**132754**

The Proportionate Review Sub-Committee of the NRES Committee London - Camden & Islington reviewed the above application on 07 April 2014.

Decision - Provisional Opinion

The Sub-Committee was content to give a favourable ethical opinion of the research, subject to the following changes being made to the documentation for study participants:

1. Members noted the initial two cognitive tests which are to be carried out no more than five days apart however it was unclear whether the participants' would be re-consented for the second test in light of them having a brain injury and further clarification is required.

2. To amend the writing in the PIS so it reads in lay language to allow it to be better understood by the participants'.

When submitting your response, please send the revised documentation underlining or otherwise highlighting the changes you have made and giving revised version numbers and dates.

The sub-committee delegated authority to confirm its final opinion on the application to the Chair.

Please let me know if you need any further clarification or would find it helpful to discuss the changes required with the lead reviewer.

The Committee will confirm the final ethical opinion within 7 days of receiving a full response.

Documents reviewed

The documents reviewed were:

Document	*Version*	*Date*
Covering Letter	email correspondence, Nele Demeyere	31 March 2014
Investigator CV	Glyn Humphreys	30 November 2011
Investigator CV	Nele Demeyere	
Participant Consent Form	v1.0	10 January 2014
Participant Information Sheet	v1.0	10 January 2014
Protocol	v1	12 February 2014
Questionnaire: Oxford Cognitive Screening	version A	
REC application	IRAS 3.5 132754/587295/1/18	28 March 2014

Membership of the Committee

The members of the Committee who were present at the meeting are listed on the attached sheet.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

14/LO/0648 Please quote this number on all correspondence

Yours sincerely

pp

Ms Heidi Chandler
Alternate Vice Chair

Email: prescommittee.london-camdenandislington@nhs.net

Enclosures: *List of names and professions of members who took part in the review*

Copy to: Ms Heather House, Oxford University Hospitals NHS Trust

NRES Committee London - Camden & Islington

Attendance at PRS Sub-Committee of the REC meeting on 07 April 2014

Committee Members:

Name	*Profession*	*Present*	*Notes*
Ms Heidi Chandler (Chair)	Research Co-ordinator	Yes	
Dr Janice Christie	Senior Lecturer in Public Health & Primary Care / Programme Director	Yes	
Dr Andy Petros	Consultant Paediatric Intensivist	Yes	

Also in attendance:

Name	*Position (or reason for attending)*
Mrs Hayley Henderson	REC Manager

NHS
Health Research Authority
NRES Committee London - Camden & Islington
North East REC Office
Room 002
TEDCO Business Centre
Rolling Mill Road
Jarrow
Tyne & Wear
NE32 3DT

Telephone: 0191 4283545

10 April 2014

Prof Glyn Humphreys
Watts Professor and Head of Department of Experimental Psychology
University of Oxford
Department Of Experimental Psychology
Tinbergen Building
OX1 3UD

Dear Prof Humphreys

Study title:	**Improving diagnosis of cognitive problems: validation and reliability of computerized cognitive screening tools**
REC reference:	**14/LO/0648**
IRAS project ID:	**132754**

Thank you for your e-mail correspondence of 9th April 2014, responding to the Proportionate Review
Sub-Committee's request for changes to the documentation for the above study.

The revised documentation has been reviewed and approved by the sub-committee.

We plan to publish your research summary wording for the above study on the NRES website, together with your contact details, unless you expressly withhold permission to do so. Publication will be no earlier than three months from the date of this favourable opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to withhold permission to publish, please contact the REC Manager at nrescommittee.london-camdenandislington@nhs.net

Confirmation of ethical opinion
On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised.

Ethical review of research sites
The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see **"Conditions of the favourable opinion" below).**

Conditions of the favourable opinion
The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission ("R&D approval") should be sought from all NHS organisations *involved in the study in accordance with NHS research governance arrangements.*

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance *should be sought from the R&D office on the information it requires to give permission for this activity.*

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations.

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non clinical trials this is not currently mandatory.

If a sponsor wishes to contest the need for registration they should contact Catherine Blewett (catherineblewett@nhs.net), the HRA does not, however, expect exceptions to be made. Guidance on where to register is provided within IRAS.

You should notify the REC in writing once all conditions have been met (except for site approvals from host organisations) and provide copies of any revised documentation

with updated version numbers. The REC will acknowledge receipt and provide a final list of the approved documentation for the study, which can be made available to host organisations to facilitate their permission for the study. Failure to provide the final versions to the REC may cause delay in obtaining permissions.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Approved documents

The documents reviewed and approved by the Committee are:

Document	*Version*	*Date*
Covering Letter	email correspondence, Nele Demeyere	31 March 2014
Covering Letter	E-mail correspondence	09 April 2014
Investigator CV	Glyn Humphreys	30 November 2011
Investigator CV	Nele Demeyere	
Participant Consent Form	1.1	08 April 2014
Participant Information Sheet	1.1	08 April 2014
Protocol	1 (Clean/Tracked)	12 February 2014
Questionnaire: Oxford Cognitive Screening	version A	
REC application	IRAS 3.5 132754/587295/1/18	28 March 2014
Response to Request for Further Information		09 April 2014

Statement of compliance
The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review
Reporting requirements

The attached document 'After ethical review – guidance for researchers' gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

Feedback

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

Further information is available at National Research Ethics Service website > After Review

14/LO/0648	Please quote this number on all correspondence

We are pleased to welcome researchers and R & D staff at our NRES committee members' training days – see details at http://www.hra.nhs.uk/hra-training/

With the Committee's best wishes for the success of this project.

Yours sincerely

pp

Ms Heidi Chandler
Alternate Vice Chair

Email: nrescommittee.london-camdenandislington@nhs.net

Enclosures: ***"After ethical review – guidance for researchers"*** *[SL-AR2]*

Copy to: Ms Heather House, Oxford University Hospitals NHS Trust

NHS
Health Research Authority

South Central - Oxford C Research Ethics Committee
Level 3, Block B
Whitefriars Building
Lewins Mead
Bristol
BS1 2NT

Telephone: 0207 104 8241

Please note: This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

02 November 2018

Dr Nele Demeyere
Dept. of Experimental Psychology
New Radcliffe House
Oxford
OX2 6GG

Dear Dr Demeyere

Study title:	**OCS-Recovery: Investigating the longer term natural history of cognitive domains after stroke.**
REC reference:	**18/SC/0550**
Protocol number:	**PID 13803**
IRAS project ID:	**248483**

The Research Ethics Committee reviewed the above application at the meeting held on 26 October 2018. Thank you for attending, along with Ms Margaret Moore, to discuss the application.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this favourable opinion letter. The expectation is that this information will be published for all studies that receive an ethical opinion but should you wish to provide a substitute contact point, wish to make a request to defer, or require further information, please contact hra.studyregistration@nhs.net outlining the reasons for your request. Under very limited circumstances (e.g. for student research which has received an unfavourable opinion), it may be possible to grant an exemption to the publication of the study.

Ethical opinion

The members of the Committee present gave a favourable ethical opinion of the above research on the basis described in the application form, protocol and supporting documentation, subject to the conditions specified below.

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

1. **Changes to the PIS:**
   A. Add the correct REC name to the PIS.
   B. **Under the section, 'What will happen to me if I take part?' change "A member of the research team will train you" to "you will be trained to complete the Oxford Cognitive Screen's Hearts Cancellation Task."**
   C. **Amend "In parts of the computerised tests, you** may be voice-**recorded."**
2. **Changes to the ICF:**
   A. **Change to follow clause "I understand that data collected during the study may** be looked at by individuals from University of Oxford, from regulatory authorities and local NHS Trust, where it is relevant **to my taking part in this research" to "I** understand that data collected during the study may be looked at by individuals from the University of Oxford, from regulatory authorities and local NHS Trust."
   B. Update the Carers Consent Form with the correct version of the Information Sheet.
3. **Changes to the Letter of Invitation to Patient:**
   A. **Make sure the correct date is used in the line "I am writing to confirm that I will** be visiting you at your home on Tuesday 15th August at 4:00pm. The **appointment should last for about 1 hour."**
   B. Add a line thanking the participant for their contribution in the study.
4. **Changes to Letter of Invitation to Carer:**
   A. **Update the following sentence "You received** a pack with 6 assessments, for **which we would like you to do one very two weeks" to "of which we would like you to do one every two weeks."**

Recommendation:

The Committee recommended that once the researchers have received a Favourable Opinion, a Substantial Amendment is submitted to add adults who lack capacity.

You should notify the REC once all conditions have been met (except for site approvals from host organisations) and provide copies of any revised documentation with updated version numbers. Revised documents should be submitted to the REC electronically from IRAS. The REC will acknowledge receipt and provide a final list of the approved documentation for the study, which you can make available to host organisations to facilitate their permission for the study. Failure to provide the final versions to the REC may cause delay in obtaining permissions.

Management permission must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for HRA and HCRW Approval (England and Wales)/ NHS permission for research is available in the Integrated Research Application System, at www.hra.nhs.uk or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought *from the R&D office on the information it requires to give permission for this activity.*

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations.

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database. This should be before the first participant is recruited but no later than 6 weeks after recruitment of the first participant.

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact hra.studyregistration@nhs.net. The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from the HRA. Guidance on where to register is provided on the HRA website.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

NHS Sites

The favourable opinion applies to all NHS sites taking part in the study taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of **the study (see "Conditions of the favourable opinion" below).**

Extract of the meeting minutes

The Committee welcomed the Chief Investigator, Dr Nele Demeyere, along with Ms Margaret Moore to the meeting.

- **Recruitment arrangements and access to health information, and fair participant selection**

  The Committee noted that the researchers have referenced "capacity" in the Brief Overview section and queried the reason adults lacking capacity have been excluded from the study, because adults who lack capacity following stroke may be those patients most in need of specific and targeted cognitive interventions.

Dr Nele Demeyere explained that this project had initially excluded adults that lack capacity due to difficult previous experiences in submitting applications involving research on adults who lack capacity.

The Committee thanked Dr Nele Demeyere for her response and recommended that once approval has been obtained a Substantial Amendment is submitted to include adults who lack capacity.

- **Informed consent process and the adequacy and completeness of participant information**

  The Committee noted a number of minor changes to the supporting documentation which will be included in the opinion letter.

Other ethical issues were raised and resolved in preliminary discussion before your attendance at the meeting.

Please contact the REC Manager if you feel that the above summary is not an accurate reflection of the discussion at the meeting.

Approved documents

The documents reviewed and approved at the meeting were:

Document	*Version*	*Date*
Covering letter on headed paper [CoverLetter_ProportionateReview]		05 September 2018
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Insurance cover letter]		10 July 2018
IRAS Application Form [IRAS_Form_31082018]		31 August 2018
IRAS Checklist XML [Checklist_21092018]		21 September 2018
Letter from sponsor [Sponsor Letter]		22 August 2018
Letters of invitation to participant [Reminder email for Carer]	0.1	15 August 2018
Letters of invitation to participant [Appointment Confirmation]	0.2	08 August 2018
Participant consent form [Consent Form patient]	0.3	16 August 2018
Participant consent form [Consent form Carer]	0.1	09 July 2018
Participant information sheet (PIS) [PIS Patient]	0.3	16 August 2018
Participant information sheet (PIS) [PIS Carer]	0.2	07 August 2018
Research protocol or project proposal [Protocal OCS-Recovery]	0.4	16 August 2018
Summary CV for Chief Investigator (CI) [CV Chief Investigator Nele Demeyere]	0.1	11 May 2017
Summary CV for student [CV Margaret Moore]		14 March 2018
Summary CV for student [CV Elise Milosevich]	1	28 August 2018
Summary CV for supervisor (student research) [CV Nele Demeyere]	0.1	11 May 2017
Validated questionnaire [Barthel]	0.1	16 August 2018
Validated questionnaire [HADS]	0.1	16 August 2018
Validated questionnaire [Overall Quality of Life]	0.1	16 August 2018
Validated questionnaire [Stroke Impact Scale]	0.1	16 August 2018

Membership of the Committee

The members of the Ethics Committee who were present at the meeting are listed on the attached sheet.

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/

HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at http://www.hra.nhs.uk/hra-training/

18/SC/0550	**Please quote this number on all correspondence**

With the Committee's best wishes for the success of this project.

Yours sincerely



PP
Professor Nigel Wellman
Chair

E-mail: nrescommittee.southcentral-oxfordc@nhs.net

Enclosures: *List of names and professions of members who were present at the meeting and those who submitted written comments*

Copy to: *OUHFT, Oxford University Hospitals NHS Foundation Trust*

South Central - Oxford C Research Ethics Committee

Attendance at Committee meeting on 26 October 2018

Committee Members:

Name	*Profession*	*Present*	*Notes*
Dr Leonard Brookes	Consultant to the Pharmaceutical Industry	Yes	
Dr Linda Cartwright	Retired Consultant Epidemiologist	No	
Dr Ben Caswell	Accountant	Yes	
Dr Nicholas Coupe	PhD Student	No	
Dr James Gavin (Co-Optee)	Academic (Exercise Physiology)	Yes	
Mrs Vivienne Laurie	Barrister	No	
Mrs Susan Lousada (Vice Chair)	Company Director (Property) & Non-legal member of first-tier tax tribunal	Yes	
Mr Trevor Olding (Co-Optee)	Clinical Coordinator	Yes	
Dr Lee Potiphar (Alternate Vice Chair)	Lecturer in Adult Nursing (Evidence Based Practice)	Yes	
Ms Anna Rathmell	Medical Manager - GI	Yes	
Dr Sabeena Sharma	Consultant Anaesthetist	No	
Dr Surjeet Singh	Clinical Trials Coordinator	No	
Ms Kayleigh Stanbury	Trial Coordinator and Developmental Assessor	No	
Ms Susan Tonks (Co-Optee)	Senior Research Support Associate	Yes	
Professor Nigel Wellman (Chair)	Professor of Health and Human Sciences	Yes	

Also in attendance:

Name	*Position (or reason for attending)*
Miss Charlotte Ferris	REC Manager

Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

NHS
Health Research Authority

Dr Nele Demeyere
Dept. of Experimental Psychology
New Radcliffe House
Oxford
OX2 6GG

Email: hra.approval@nhs.net
Research-permissions@wales.nhs.uk

03 December 2018
Reissued 30 January 2019 to correct document list

Dear Dr Demeyere

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title:	**OCS-Recovery: Investigating the longer term natural history of cognitive domains after stroke.**
IRAS project ID:	**248483**
Protocol number:	**PID 13803**
REC reference:	**18/SC/0550**
Sponsor	**University of Oxford**

I am pleased to confirm that HRA and Health and Care Research Wales (HCRW) Approval has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

How should I continue to work with participating NHS organisations in England and Wales?
You should now provide a copy of this letter to all participating NHS organisations in England and Wales, as well as any documentation that has been updated as a result of the assessment.

Following the arranging of capacity and capability, participating NHS organisations should formally confirm their capacity and capability to undertake the study. How this will be confirmed is detailed in the "*summary of assessment*" section towards the end of this letter.

You should provide, if you have not already done so, detailed instructions to each organisation as to how you will notify them that research activities may commence at site following their confirmation of capacity and capability (e.g. provision by you of a 'green light' email, formal notification following a site initiation visit, activities may commence immediately following confirmation by participating organisation, etc.).

IRAS project ID	248483

It is important that you involve both the research management function (e.g. R&D office) supporting each organisation and the local research team (where there is one) in setting up your study. Contact details of the research management function for each organisation can be accessed here.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland? (if applicable)

HRA and HCRW Approval does not apply to NHS/HSC organisations within the devolved administrations of Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) has been sent to the coordinating centre of each participating nation. You should work with the relevant national coordinating functions to ensure any nation specific checks are complete, and with each site so that they are able to give management permission for the study to begin.

Please see IRAS Help for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations? (if applicable)
HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?
The document "*After Ethical Review – guidance for sponsors and investigators***", issued with your REC** favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

I am a participating NHS organisation in England or Wales. What should I do once I receive this letter?
You should work with the applicant and sponsor to complete any outstanding arrangements so you are able to confirm capacity and capability in line with the information provided in this letter.

The sponsor contact for this application is as follows:

Name: Heather House
Email: ctrq@admin.ox.ac.uk

IRAS project ID	248483

Who should I contact for further information?
Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **248483**. Please quote this on all correspondence.

Yours sincerely

Isobel Lyle | Senior Assessor
Health Research Authority
T: 0207 972 2496
HRA, Holland Dr, Newcastle upon Tyne NE2 4NQ
Hra.approval@nhs.net or Isobel.lyle@nhs.net
www.hra.nhs.uk

Copy to: *Heather House, Sponsor contact,* Oxford University
ouhtma@ouh.nhs.uk, R&D, Oxford University Hospitals NHS Foundation Trust

IRAS project ID	248483

List of Documents

The final document set assessed and approved by HRA and HCRW Approval is listed below.

Document	*Version*	*Date*
Covering letter on headed paper [CoverLetter_ProportionateReview]		05 September 2018
Covering letter on headed paper [REC Response]		23 November 2018
Covering letter on headed paper [HRA response]		23 November 2018
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Insurance cover letter]		10 July 2018
HRA Schedule of Events [HRA assessed]	1.0	08 October 2018
HRA Statement of Activities [HRA assessed]	1.0	08 October 2018
IRAS Application Form [IRAS_Form_31082018]		31 August 2018
Letter from funder [Stroke Award Letter]	0.1	16 August 2018
Letter from sponsor [Sponsor Letter]		22 August 2018
Letters of invitation to participant [Appointment Confirmation]	0.2	08 August 2018
Letters of invitation to participant [Reminder email for Carer]	0.2	23 November 2018
Other [Funding Letter 2]	0.1	29 March 2017
Other [Appointment Confirmation Template]	0.3	23 November 2018
Participant consent form [Consent Form patient]	0.4	23 November 2018
Participant consent form [Consent form Carer]	0.2	23 November 2018
Participant information sheet (PIS) [PIS Patient]	0.5	23 November 2018
Participant information sheet (PIS) [PIS Carer]	0.3	23 November 2018
Research protocol or project proposal [Protocal OCS-Recovery]	0.4	16 August 2018
Summary CV for Chief Investigator (CI) [CV Chief Investigator Nele Demeyere]	0.1	11 May 2017
Summary CV for student [CV Margaret Moore]		14 March 2018
Summary CV for student [CV Elise Milosevich]	1	28 August 2018
Summary CV for supervisor (student research) [CV Nele Demeyere]	0.1	11 May 2017
Validated questionnaire [Barthel]	0.1	16 August 2018
Validated questionnaire [HADS]	0.1	16 August 2018
Validated questionnaire [Overall Quality of Life]	0.1	16 August 2018
Validated questionnaire [Stroke Impact Scale]	0.1	16 August 2018

IRAS project ID	248483

Summary of assessment

The following information provides assurance to you, the sponsor and the NHS in England and Wales that the study, as assessed for HRA and HCRW Approval, is compliant with relevant standards. It also provides information and clarification, where appropriate, to participating NHS organisations in England and Wales to assist in assessing, arranging and confirming capacity and capability.

Section	Assessment Criteria	Compliant with Standards?	Comments
1.1	IRAS application completed correctly	Yes	No comments
2.1	Participant information/consent documents and consent process	Yes	No comments
3.1	Protocol assessment	Yes	No comments
4.1	Allocation of responsibilities and rights are agreed and documented	Yes	A statement of activities has been submitted and the sponsor is not requesting and does not expect any other site agreement to be used.
4.2	Insurance/indemnity arrangements assessed	Yes	No comments
4.3	Financial arrangements assessed	Yes	No funding is being provided to participating NHS organisations.
5.1	Compliance with the Data Protection Act and data security issues assessed	Yes	No comments
5.2	CTIMPS – Arrangements for compliance with the Clinical Trials Regulations assessed	Not Applicable	No comments
5.3	Compliance with any applicable laws or regulations	Yes	No comments
6.1	NHS Research Ethics Committee favourable opinion	Yes	No comments

IRAS project ID	248483

Section	Assessment Criteria	Compliant with Standards?	Comments
	received for applicable studies		
6.2	CTIMPS – Clinical Trials Authorisation (CTA) letter received	Not Applicable	No comments
6.3	Devices – MHRA notice of no objection received	Not Applicable	No comments
6.4	Other regulatory approvals and authorisations received	Not Applicable	No comments

Participating NHS Organisations in England and Wales

This provides detail on the types of participating NHS organisations in the study and a statement as to whether the activities at all organisations are the same or different.
All organisations will be undertaking the same activity (i.e. there is only one 'site-type'). This involves all activities as indicated in the Statement of Activities. The Chief Investigator or sponsor should share relevant study documents with participating NHS organisations in England and Wales in order to put arrangements in place to deliver the study. The documents should be sent to both the local study team, where applicable, and the office providing the research management function at the participating organisation. Where applicable, the local LCRN contact should also be copied into this correspondence. If chief investigators, sponsors or principal investigators are asked to complete site level forms for participating NHS organisations in England and Wales which are not provided in IRAS or on the HRA or HCRW websites, the chief investigator, sponsor or principal investigator should notify the HRA immediately at hra.approval@nhs.net, or HCRW at Research-permissions@wales.nhs.uk. We will work with these organisations to achieve a consistent approach to information provision.

Principal Investigator Suitability

This confirms whether the sponsor's position on whether a PI, LC or neither should be in place is correct for each type of participating NHS organisation in England and Wales, and the minimum expectations for education, training and experience that PIs should meet (where applicable).
Currently, the Chief Investigator is acting as Principal Investigator at a single NHS participating NHS organisation. Further clarification is being sought from the Sponsor should more NHS sites be added. This will be confirmed in the Outcome of the Application for HRA Approval. GCP training is not a generic training expectation, in line with the HRA/HCRW/MHRA statement on training expectations.

IRAS project ID	248483

HR Good Practice Resource Pack Expectations

This confirms the HR Good Practice Resource Pack expectations for the study and the pre-engagement checks that should and should not be undertaken.
Access will be required for Students involved in data collection and processing for this study. Further clarification is being sought from the Sponsor and the type of access required from the NHS will be confirmed in the Outcome of the Application for HRA Approval.

Other Information to Aid Study Set-up

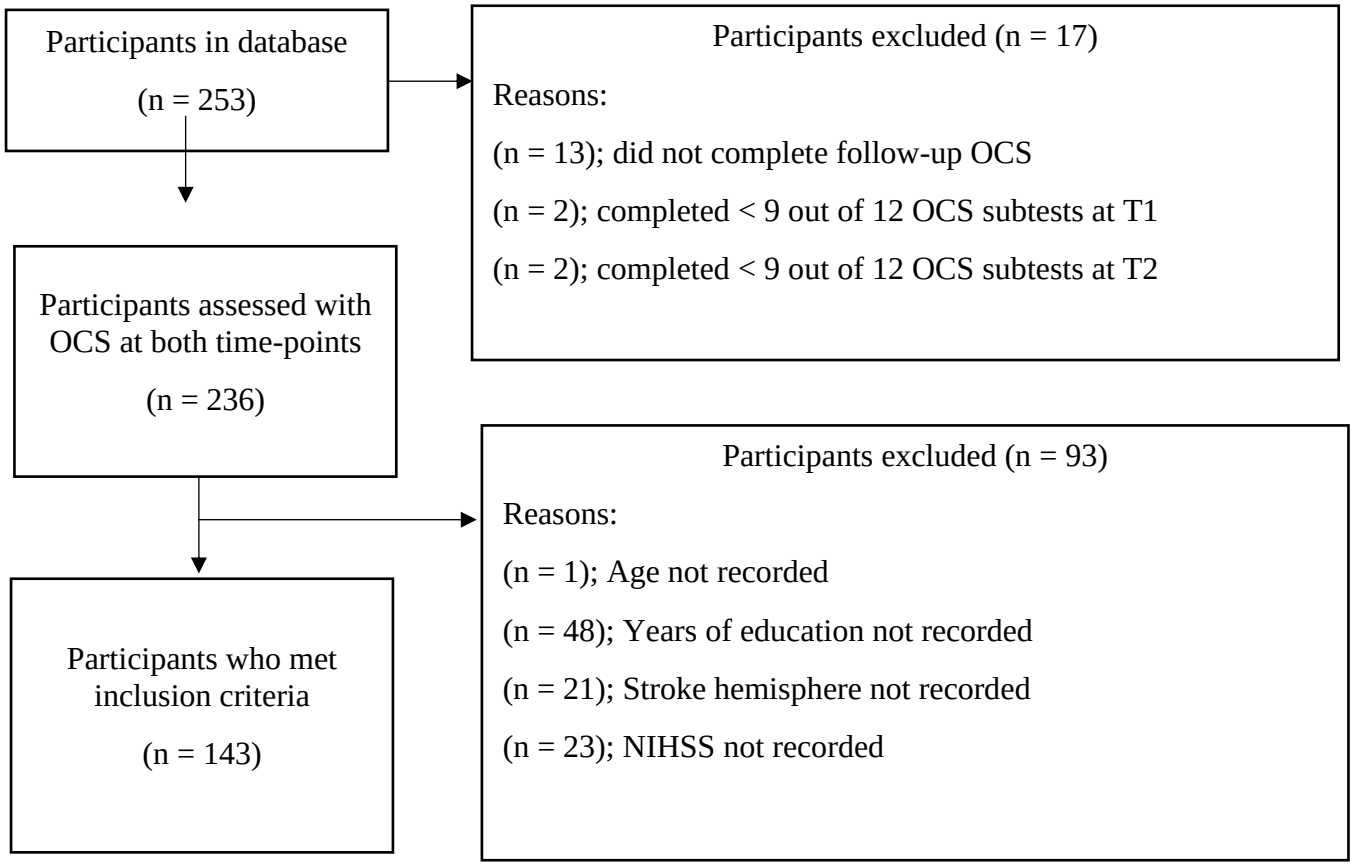
This details any other information that may be helpful to sponsors and participating NHS organisations in England and Wales in study set-up.
The applicant has indicated that they intend to apply for inclusion on the NIHR CRN Portfolio. The applicant expects this research could lead to the development of a new product/process or the generation of intellectual property

Appendix 3.C

Participant Inclusion/ Exclusion Flow Diagram

Figure A1

Participant Flow Diagram



Appendix 3.D

A Priori Power Calculations

Analysis 1

Based on data from Table 3 in Boosman et al. (2014) a large effect size of $d = 1.18$ was calculated for this analysis. The power analysis for the two-tailed independent sample t-test yielded a power of 0.99 with an $\alpha = 0.007$.

t tests - Means: Difference between two independent means (two groups)

Analysis:	Compromise: Compute implied α & power		
Input:	Tail(s)	=	Two
	Effect size d	=	1.18
	β/α ratio	=	1
	Sample size group 1	=	23
	Sample size group 2	=	120
Output:	Noncentrality parameter δ	=	5.1840422
	Critical t	=	2.7186598
	Df	=	141
	α err prob	=	0.007378172
	β err prob	=	0.007378172
	Power (1-β err prob)	=	0.9926218

Analysis 2

Based on data from Tables 2 and 3 in Carroll and Coetzer (2011), an estimated slope of -0.31 was used for this analysis. The power analysis for the two-tailed simple regression model yielded a power of 0.99 with an $\alpha = 0.01$.

t tests - Linear bivariate regression: One group, size of slope

Analysis:	Compromise: Compute implied α & power		
Input:	Tail(s)	=	Two
	Slope H1	=	-0.3067143
	β/α ratio	=	1
	Total sample size	=	143
	Slope H0	=	0
	Std dev σ_x	=	5.6
	Std dev σ_y	=	4.52
Output:	Noncentrality parameter δ	=	-4.9126548
	Critical t	=	-2.5879187
	Df	=	141
	α err prob	=	0.0106665
	β err prob	=	0.0106665
	Power (1-β err prob)	=	0.9893335

Analysis 3

To the best of the authors knowledge, there is no existing effect size for the moderation analysis within the literature base. It is understood that interaction effects tend to be less powerful than linear effects with the same effect size (Perugini et al., 2018). In line with this, a small-medium effect size ($f^2 = 0.09$) was estimated (Cohen, 2013). The power analysis for the multiple regression model yielded a power of 0.95 with an $\alpha = 0.05$.

F tests - Linear multiple regression: Fixed model, R^2 increase

Analysis:	Compromise: Compute implied α & power		
Input:	Effect size f^2	=	0.09
	β/α ratio	=	1
	Total sample size	=	143
	Number of tested predictors	=	1
	Total number of predictors	=	3
Output:	Noncentrality parameter λ	=	12.8700000
	Critical F	=	3.8305514
	Numerator df	=	1
	Denominator df	=	139
	α err prob	=	0.0523301
	β err prob	=	0.0523301
	Power (1-β err prob)	=	0.9476699

Appendix 3.E

Computing Variables

1. Recoding Data to Make the OCI Scores at T1 and T2

Data was encoded as 'spared' (0) or 'impaired' (1) in line with the cut-offs provided in A2 (Demeyere et al., 2015). Scores below the cut-off were scored as 'impaired'.

Figure A2

Normative Data and Cut-Offs Indicating Impairment for OCS Subtests

Table 1
Normative Data and Cut Offs for Impairment (Scores on Subtests Lower Than 5th Centile and Higher Than 95th Centile Denote an Impairment)

Task name	Measure	Control sample *N*	Median score on task	Min task score	Max task score	5th centile	95th centile
Picture naming	Overall accuracy	140	4	2	4	3	
Semantics	Overall accuracy	140	3	3	3	3	
Orientation	Overall accuracy	140	4	2	4	4	
Visual field	Overall accuracy	140	4	4	4	4	
Sentence reading	Overall accuracy	140	15	12	15	14	
Number writing	Overall accuracy	140	3	2	3	3	
Calculation	Overall accuracy	140	4	2	4	3	
Broken hearts	Overall accuracy	140	48	34	50	42	
	Space Asym (left inattention > 0, right < 0)	140	0	−5	4	−2	3[a]
	Obj Asym (left inattention > 0, right < 0)	140	0	−2	2	0[b]	0[b]
Imitation	Overall accuracy	140	11	6	12	8	
Recall and recognition	Verbal recall	115	2	0	4	0	
	Verbal memory recall and recognition	115	4	1	4	3	
	Episodic recognition	64	4	1	4	3	
Executive task	Mixed	138	12	1	13	7	
	Exec score acc (sum of accuracy in single tasks versus mixed)	138	0	−3	9		4

[a] For a more consistent approach: Use cut off of absolute value >2. [b] For a more conservative approach: Use cut off of 1.

Note. Table reproduced from (Demeyere et al., 2015); highlighted tests included in analyses.

In order to calculate the percentage of OCS test failed at chronic, the number of missing subtests were summed for each participant. The OCI score was subsequently computed as: total number of failed tests/ (12 – number of missing tests) × 100.

2. Generating the Cognitive Change Score

Overall OCI at T1 and overall OCI at T2 were calculated for each participant. Cognitive change scores were computed as $OCI_{T2} – OCI_{T1}$ (positive scores indicate cognitive decline).

3. Generating the Awareness Score

CFQ raw scores and OCI at T2 were transformed to Z-scores and OCI *Z*-scores were subtracted from the CFQ *Z*-scores ($Z_{CFQ} - Z_{OCI_T2}$). The answer was subsequently transformed into a *T*-score to represent awareness: Awareness *T*-score = (($Z_{CFQ} - Z_{OCI_T2}$) x 10) + 50.

4. Recoding OCI at T2 into 'Mild-No Cognitive Impairment' vs 'Moderate-Severe Cognitive Impairment'

Participants who obtained a OCI_{T2} Z-score of < 1 were scored as 'mild-no cognitive impairment' and participants who obtained a OCI_{T2} Z-score $\geq$ 1 were scored as 'moderate-severe cognitive impairment'.

Appendix 3.F

Minimising Bias

Table A1

Forced-entry Logistic Regression Analyses of the Predictive Value of Baseline Characteristics of Patients on Completed follow-up at 6-months (yes/no)

Variable	*b*	SE *b*	Wald	*p*
Constant	-1.20	1.25	0.92	.337
Age	-0.02	0.02	1.68	.195
Constant	-1.93	0.86	5.06	.025
Sex	-0.71	0.62	1.34	.247
Constant	-3.06	1.67	3.35	.067
Education (years)	-0.03	0.13	0.06	.810
Constant	-2.78	0.81	11.81	.001
Stroke Hemisphere	-0.04	0.47	0.006	.937
Constant	-4.71	1.04	20.68	.000

NIHSS Total	0.03	0.08	0.14	.708
Constant	-2.78	1.07	6.75	.009
BI Total	0.001	0.06	0.00	.983
Constant	-3.06	0.45	46.60	.000
OCI (%) T1	0.005	0.01	0.19	.663

Benjamini and Hochberg (1995) False Discovery Rate

The Benjamini and Hochberg (1995) false discovery rate procedure was carried out to decrease the likelihood of type-1 error due to multiple comparisons, whilst maintaining power to detect differences where they exist at the 0.5 level.

SPSS Syntax:

```
DATA LIST free / p (F5.3).
BEGIN DATA
.247 .195 .810 .937 .708 .983 .663
END DATA.
SORT CASES by p (a).
COMPUTE i=$casenum.
SORT CASES by i (d).
COMPUTE q=.05.
COMPUTE m=max(i,lag(m)).
COMPUTE crit=q*i/m.
COMPUTE test=(p le crit).
COMPUTE test=max(test,lag(test)).
FORMATS i m test(f8.0) q (f8.2) crit(f8.6).
VALUE LABELS test 1 'Significant' 0 'Not Significant'.
LIST.
```

No variables were significant.

Table A2

Forced-entry Logistic Regression Analyses of the Predictive Value of Baseline Characteristics on Exclusion Due to Missing Data (yes/no)

Variable	*b*	SE *b*	Wald	*p*
Constant	0.27	0.71	0.15	.699
Age	0.000	.009	.000	.988
Constant	0.04	0.39	0.01	.911
Sex	0.15	0.26	0.34	.558
Constant	1.14	0.62	3.39	.066
Education (years)	-0.01	0.05	0.06	.808
Constant	0.29	0.40	0.54	.463
Stroke Hemisphere	0.24	0.23	1.09	.296
Constant	1.26	0.28	20.28	.000
NIHSS Total	0.04	0.03	1.53	.217

Constant	0.73	0.53	1.93	.165
BI Total	-0.02	0.03	0.30	.582
Constant	0.34	0.19	3.01	.083
OCI (%) T1	-0.002	0.005	0.26	.612

Benjamini and Hochberg (1995) False Discovery Rate

The Benjamini and Hochberg (1995) false discovery rate procedure was carried out to decrease the likelihood of type-1 error due to multiple comparisons, whilst maintaining power to detect differences where they exist at the 0.5 level.

SPSS Syntax:

```
DATA LIST free / p (F5.3).
BEGIN DATA
.558 .988 .808 .296 .217 .582 .612
END DATA.
SORT CASES by p (a).
COMPUTE i=$casenum.
SORT CASES by i (d).
COMPUTE q=.05.
COMPUTE m=max(i,lag(m)).
COMPUTE crit=q*i/m.
COMPUTE test=(p le crit).
COMPUTE test=max(test,lag(test)).
FORMATS i m test(f8.0) q (f8.2) crit(f8.6).
VALUE LABELS test 1 'Significant' 0 'Not Significant'.
LIST.
```

No variables were significant.

Appendix 3.G

Univariate Analyses to Identify Covariates

Table A3

Univariate Analyses Exploring the Association Between Demographic and Stroke Characteristic Variables and Awareness to Identify Covariates

Variable	Statistic	*p*
Age	- .14[a]	.105
Education (years)	.07[a]	.408
NIHSS	-.13[b]	.133
BI	.14[b]	.090*
Sex	-.58[c]	.563
Stroke Hemisphere	.59[d]	.556

Note. [a]Pearson's *r*; [b]Spearman's ρ (r_s); [c] t-test (*t*); [d]ANOVA (*F*); * $p < .1$.

Table A4

Univariate Analyses Exploring the Association Between Demographic and Stroke Characteristic Variables and HADS to Identify Covariates

Variable	Statistic	*P*
Age	.08[a]	.324
Education (years)	-.14[a]	.107
NIHSS	.01[b]	.908
BI	-.37[b]	.000**
Sex	.37[c]	.713
Stroke Hemisphere	1.18[d]	.310

Note. [a]Pearson's *r*; [b]Spearman's ρ (r_s); [c] t-test (*t*); [d]ANOVA (*F*); * $p < .1$

Appendix 3.H

Statistical Assumptions for Hypothesis One

Variable Type

The dependent variable (DV) and covariate variable were continuous. The independent variable (IV) consisted of two categorical, independent groups.

Independence of Observations

Different participants were in each group, with no participant being in more than one group.

Absence of Significant Outliers

Boxplots indicated that there were no 'extreme' outliers. CFQ Z-scores and OCI at T2 Z-scores were further examined for extreme outliers. One would expect to see $\leq$ 5% of participants with values greater than $\pm$ 1.96. Outcomes indicated that 95.8% of awareness CFQ Z-scores and 94.4% of OCI at T2 Z-scores were within the 'normal' range (Z-score < + 1.96), respectively.

Normal Distribution of Residuals

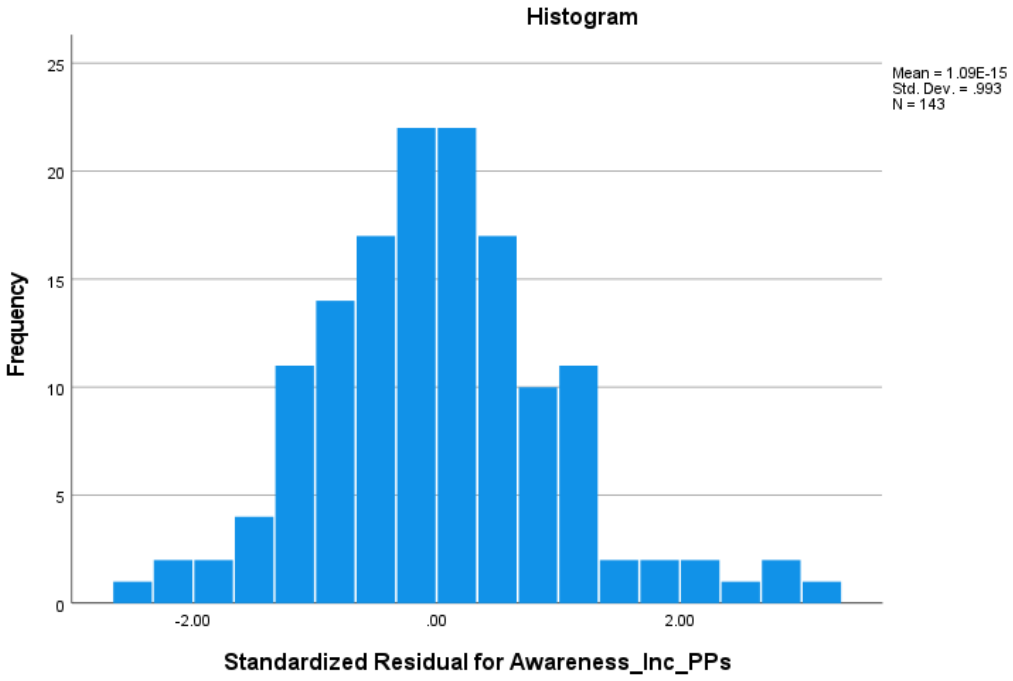
Overall model fit:

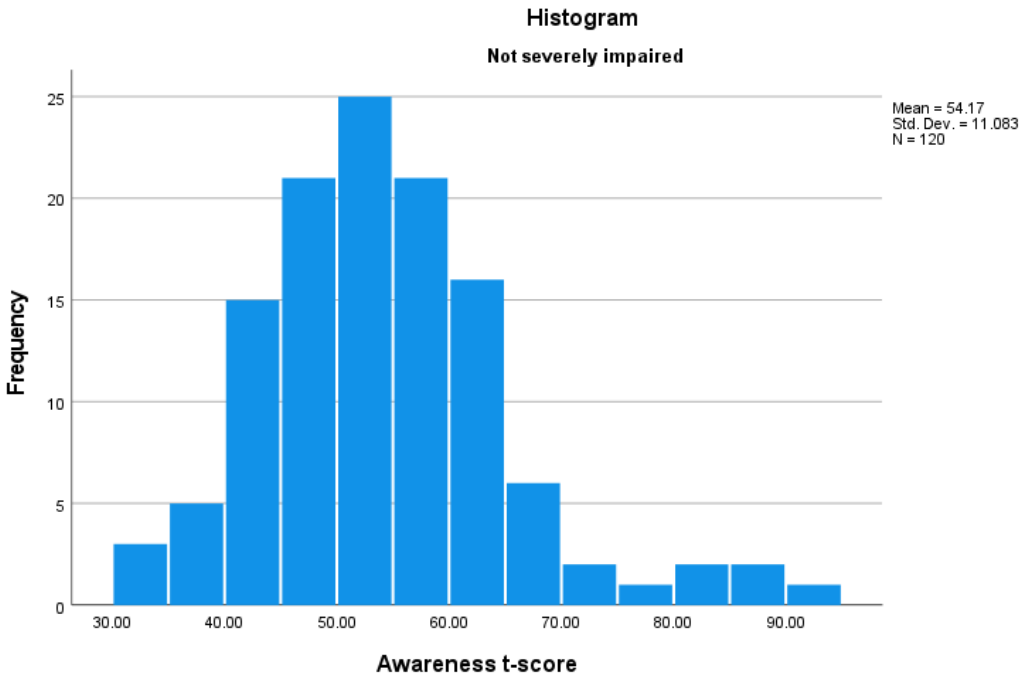
	Shapiro-Wilk		
	Statistic	df	Sig.
Standardized Residual for Awareness	.978	143	.019

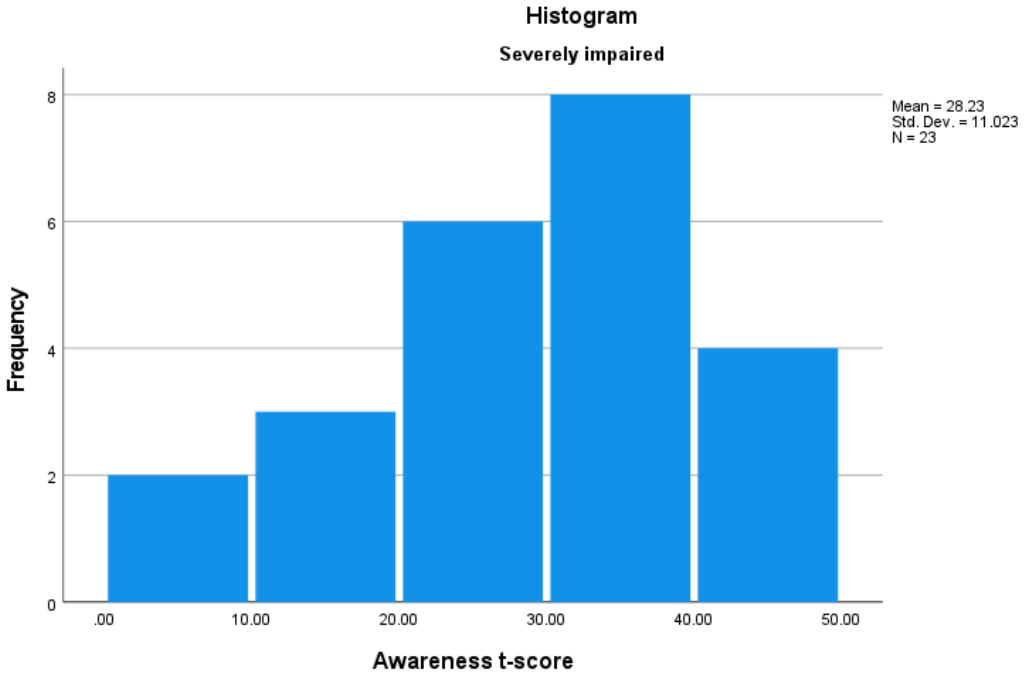
Within-group residuals:

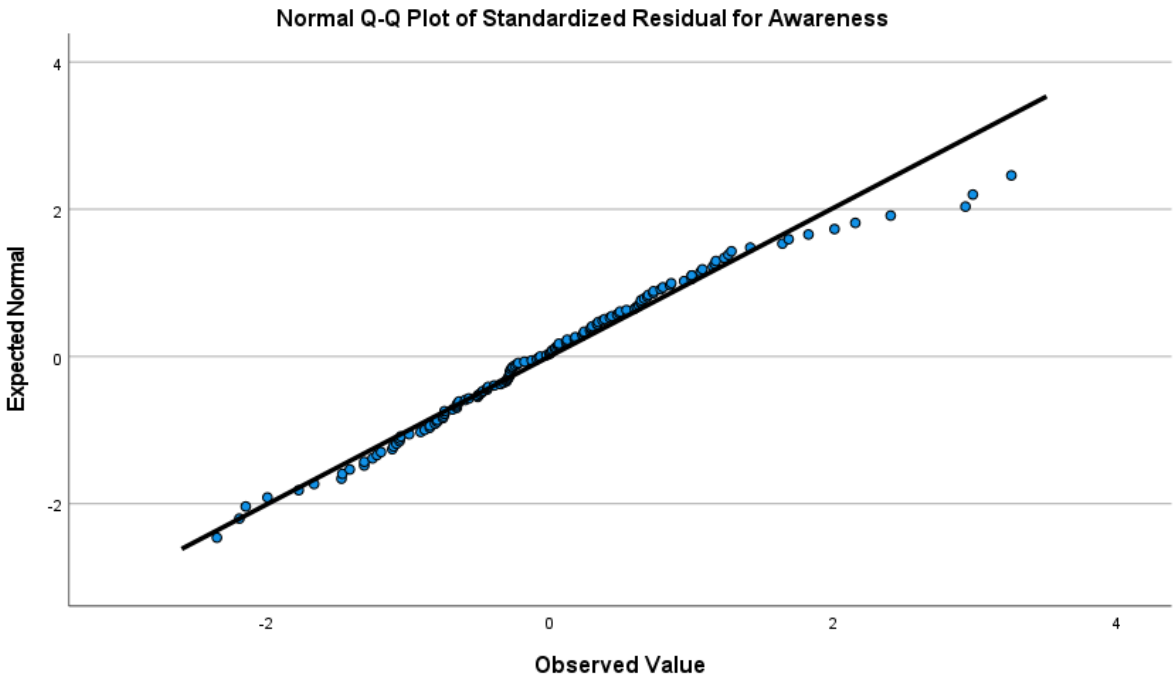
		Shapiro-Wilk		
	OCOGNITIVE IMPAIRMENTT2 Group	Statistic	df	Sig.
Awareness t-score	Mild-no cognitive impairment	.953	120	.000
	Moderate-severe cognitive impairment	.973	23	.762

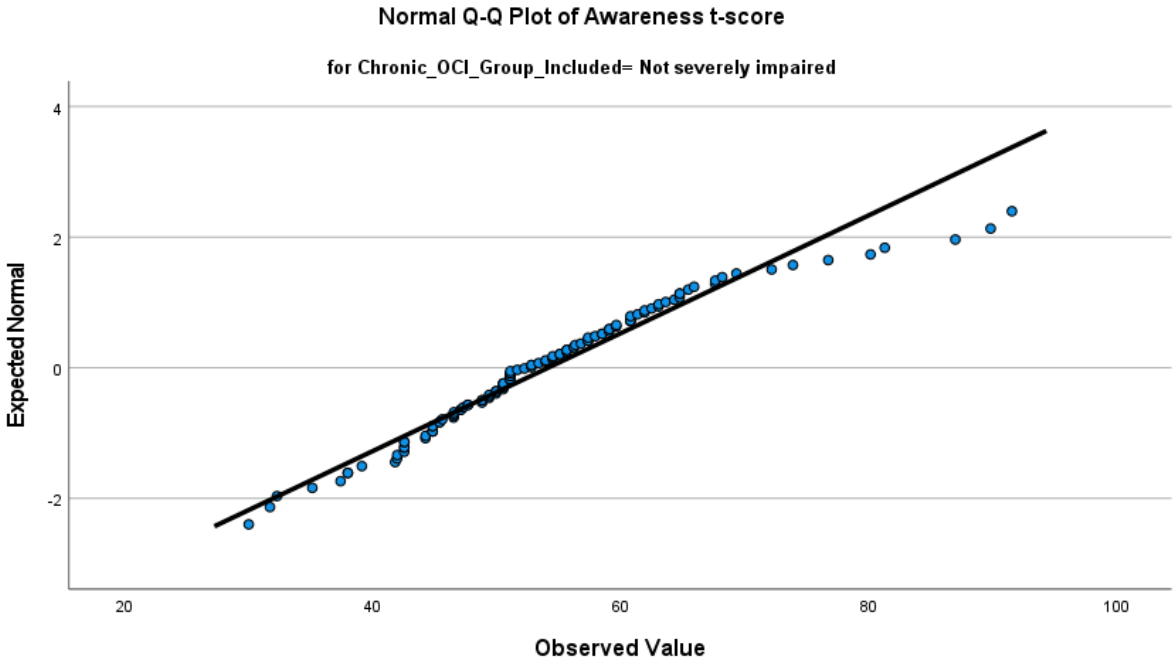
The Shapiro-Wilk test indicates that the standardised residuals for the overall model and the within-group residuals are not normally distributed. However, the statistics are .978 and .953 respectively, whereby 1 is a perfect normal distribution. It is known that the Shapiro-Wilk test can be overly-sensitive to minor deviations from a perfect normal distribution in a large sample, whilst ANCOVA is quite robust to violations of normality, so it is unlikely that this assumption is violated in this case (Field, 2013). This is further demonstrated by the histograms and Q-Q plots, which show small acceptable deviation. Therefore, it was decided that the assumption of normality was met.

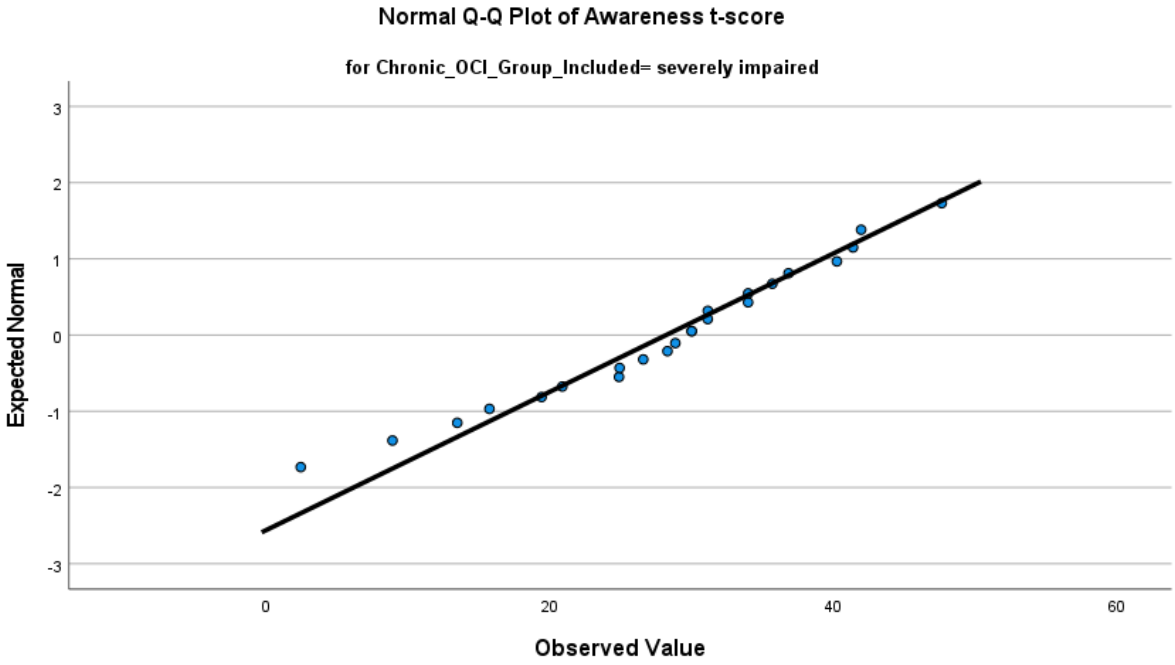



Histogram
Not severely impaired
Mean = 54.17
Std. Dev. = 11.083
N = 120
Frequency
25
20
15
10
5
0
30.00
40.00
50.00
60.00
70.00
80.00
90.00
Awareness t-score


Histogram
Severely impaired
Mean = 28.23
Std. Dev. = 11.023
N = 23
Frequency
8
6
4
2
0
.00
10.00
20.00
30.00
40.00
50.00
Awareness t-score


Normal Q-Q Plot of Standardized Residual for Awareness
Expected Normal
4
2
0
-2
-2
0
2
4
Observed Value


Normal Q-Q Plot of Awareness t-score
for Chronic_OCI_Group_Included= Not severely impaired
Expected Normal
4
2
0
-2
20
40
60
80
100
Observed Value

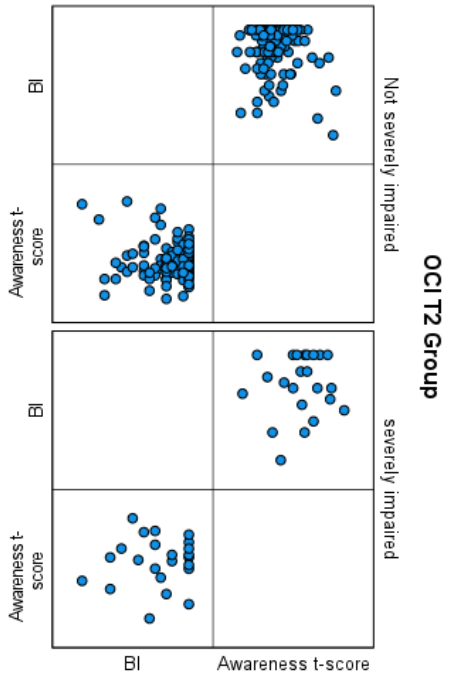


Homogeneity of Variances

Levene's test indicated that data met the assumption for homogeneity of variance (p = .962).

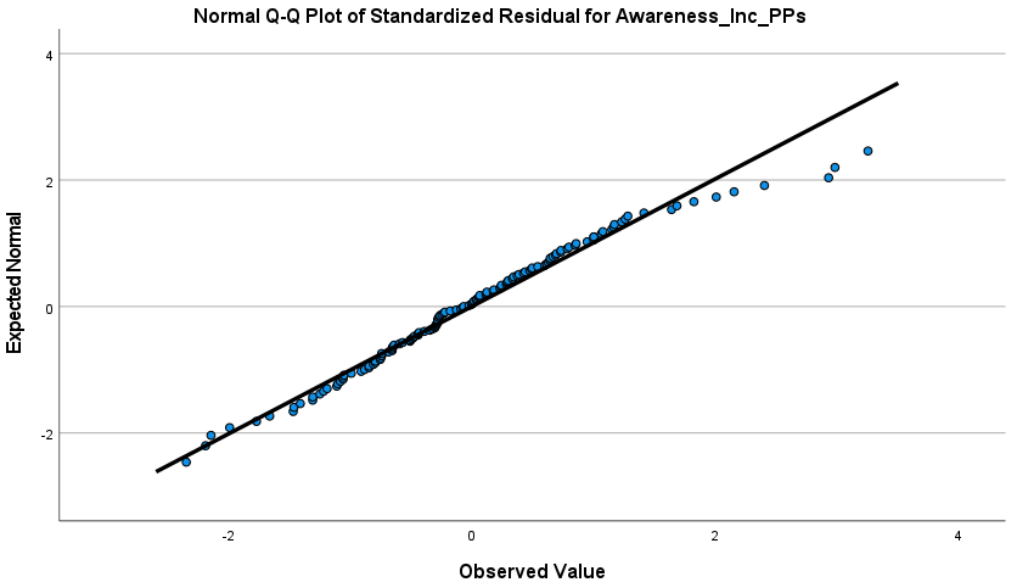
Linearity

The covariate should be linearly related to the dependent variable at each level of the independent variable. A matrix scatterplot of the relationship between residuals for BI scores and residuals for awareness scores against the residuals for each level of the predictor variable revealed that there was a violation of the assumption that a linear relationship exists between the covariate and the dependent variable.



Homoscedasticity

A scatterplot of the standardised residuals and the predicted values was created. This suggests that the observed residual values lie closely to the line which represents a normal distribution.



Homogeneity of Regression Slopes

A univariate analysis indicated that the covariate, BI, had a significantly different relationship with the moderate-severe cognitive impairment group vs the mild-no cognitive

impairment group $F(2, 140) = 40.27$, $p < .001$. This indicates that the assumption of homogeneity of regression slopes was violated.

Robust ANCOVA code

SPSS code:

```
BEGIN PROGRAM R.
library(WRS2)
mySPSSdata = spssdata.GetDataFromSPSS(factorMode = "labels")
ancova(Awareness_Inc_PPs ~ Chronic_OCI_Group_Included + Barthel, data =
mySPSSdata)
END PROGRAM.
Call:
ancova(formula = Awareness_Inc_PPs ~ Chronic_OCI_Group_Included +
    Barthel, data = mySPSSdata)

             n1 n2    diff     se lower COGNITIVE IMPAIRMENTupper COGNITIVE
IMPAIRMENT statistic p-value
Barthel = 13 13 22 22.4358 3.9752  10.7323  34.1393    5.6440   1e-04
Barthel = 16 27 19 20.5287 2.6708  12.8723  28.1851    7.6863   0e+00
Barthel = 19 68 17 25.1111 2.6868  17.3284  32.8937    9.3461   0e+00
Barthel = 20 61 16 25.4051 2.7071  17.4808  33.3294    9.3848   0e+00
```

Appendix 3.I

Statistical Assumptions for Hypothesis Two

Variable Type

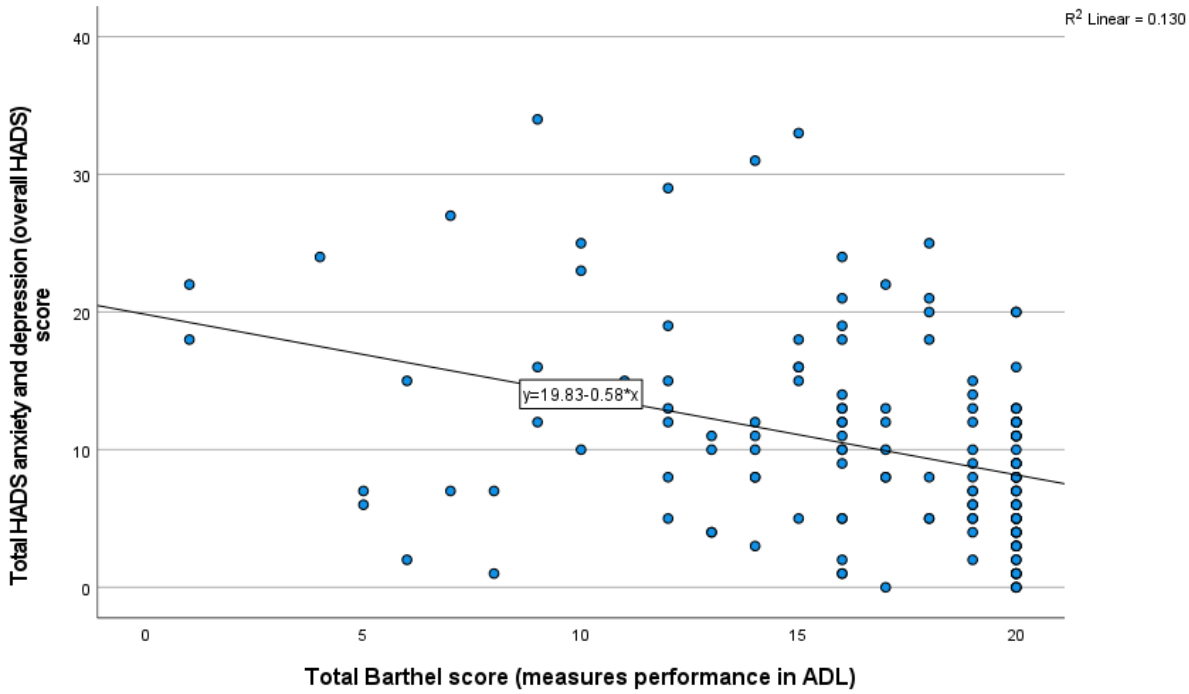
The DV was continuous. There were two continuous IVs.

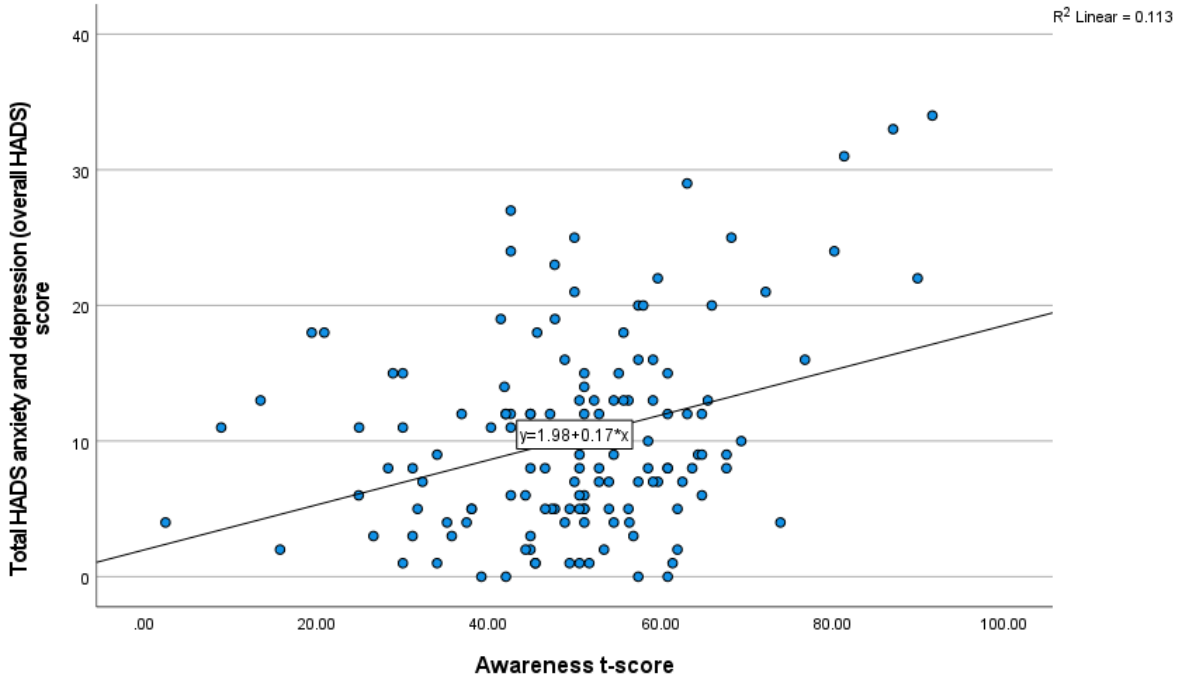
Independence of Residuals

The Durbin-Watson statistic was 2.03, indicating that the values of the residuals are independent, and this assumption has been met.

Linearity

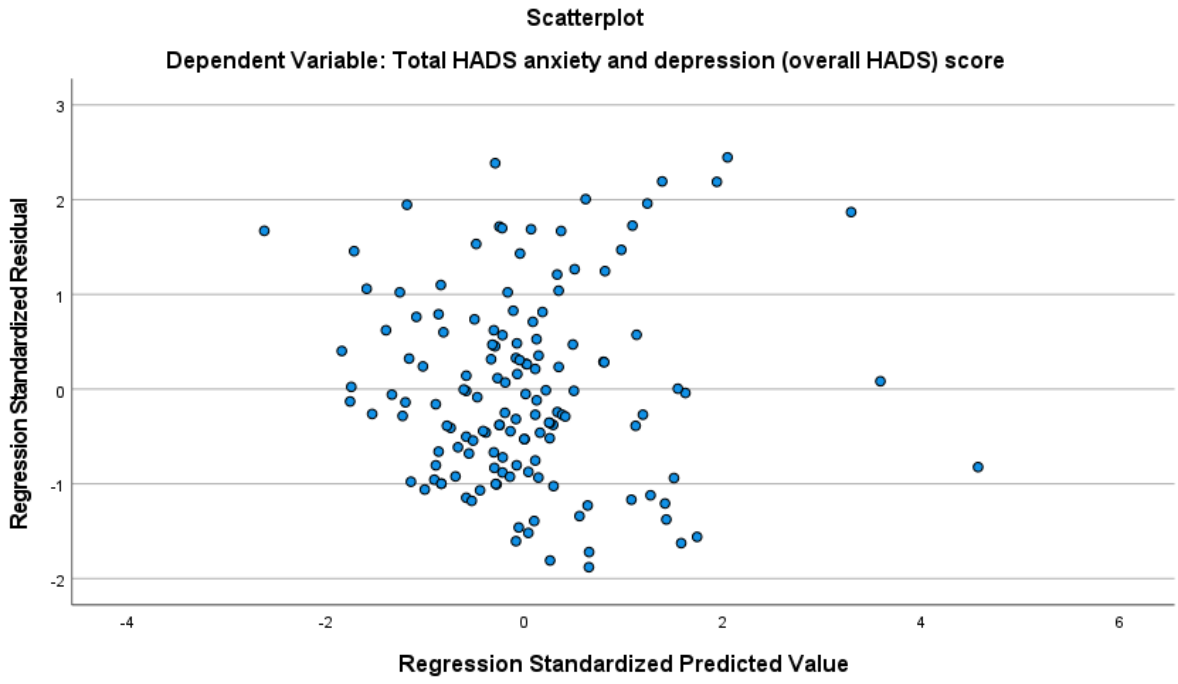
Scatterplots were used to examine the relationship between the IVs and the DV and show that this assumption was met.





Homoscedasticity

The variance of the standard residuals appears constant, in that the plot of standardised residuals vs standardised predicted values showed no obvious signs of funnelling, suggesting the assumption of homoscedasticity was met.



Absence of Multicollinearity

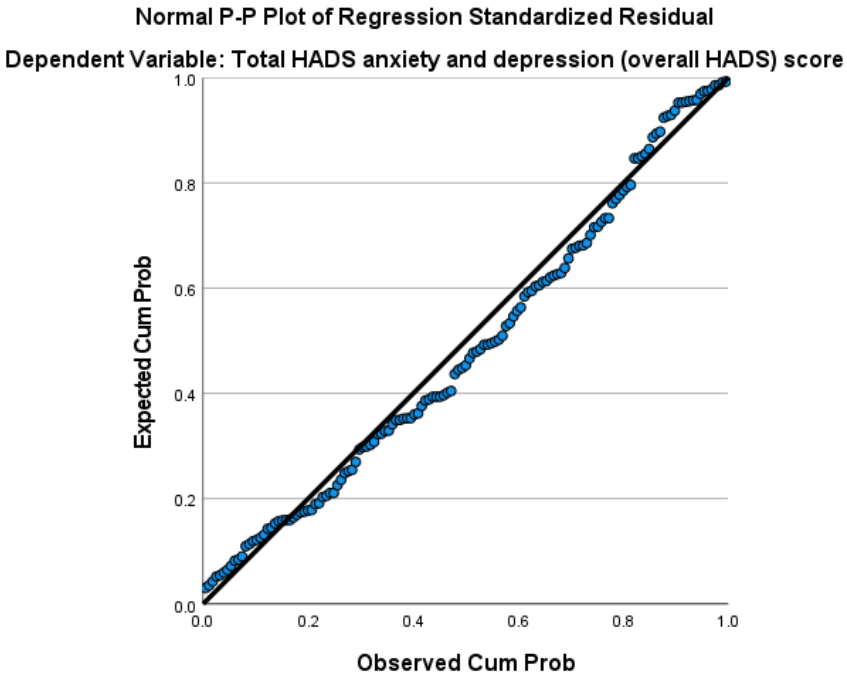
Correlation coefficients and Tolerance/VIF values were inspected for possible multicollinearity. Correlation coefficients were <0.8. VIF scores were well below 10, and tolerance scores were >0.2 (statistics = 1.00 and .994 respectively). Thus, the assumption was met.

Absence of Significant Outliers

Cook's Distance values were all <1, suggesting that individual cases were not unduly influencing the model, thus this assumption was met.

Normal Distribution of Residuals

A P-P plot shows that there is a reasonably good linear relationship between residuals, suggesting that they are normally distributed, and this assumption is met.



Appendix 3.J

Statistical Assumptions for Hypotheses Three

Assumptions for the moderator analysis were examined by firstly centering the means for the predictor and moderator variable and computing an interaction variable by multiplying scores of the centred variables cognitive change and OCI T1. A forced-entry regression with the centred predictor, centred moderator, the interaction variable, and BI and Awareness as predictors in order to inspect sources of bias in the model. Assumptions are the same as for a multiple linear regression.

Variable Type

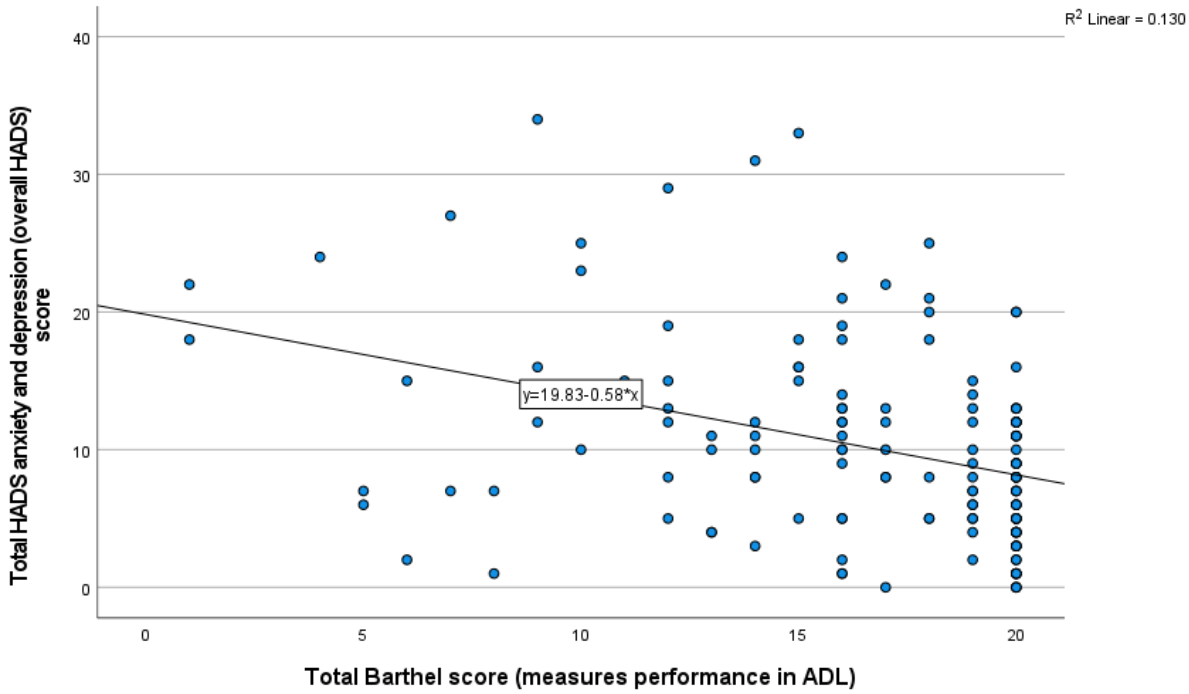
The DV was continuous. There were two continuous IVs.

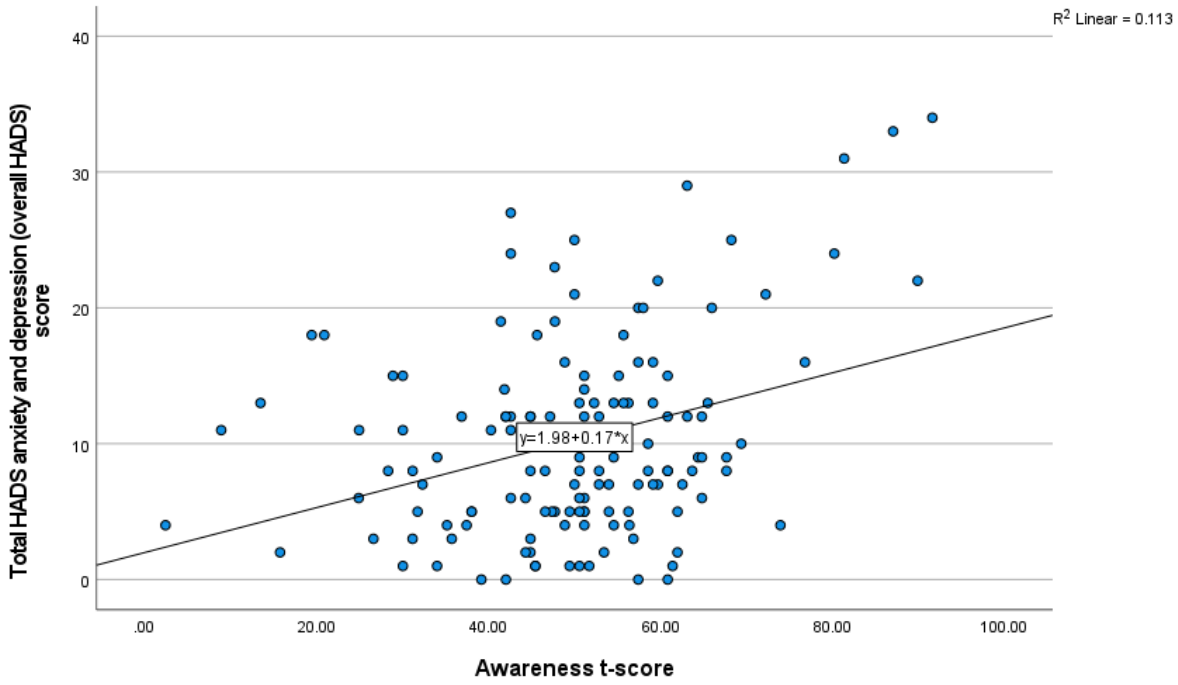
Independence of Residuals

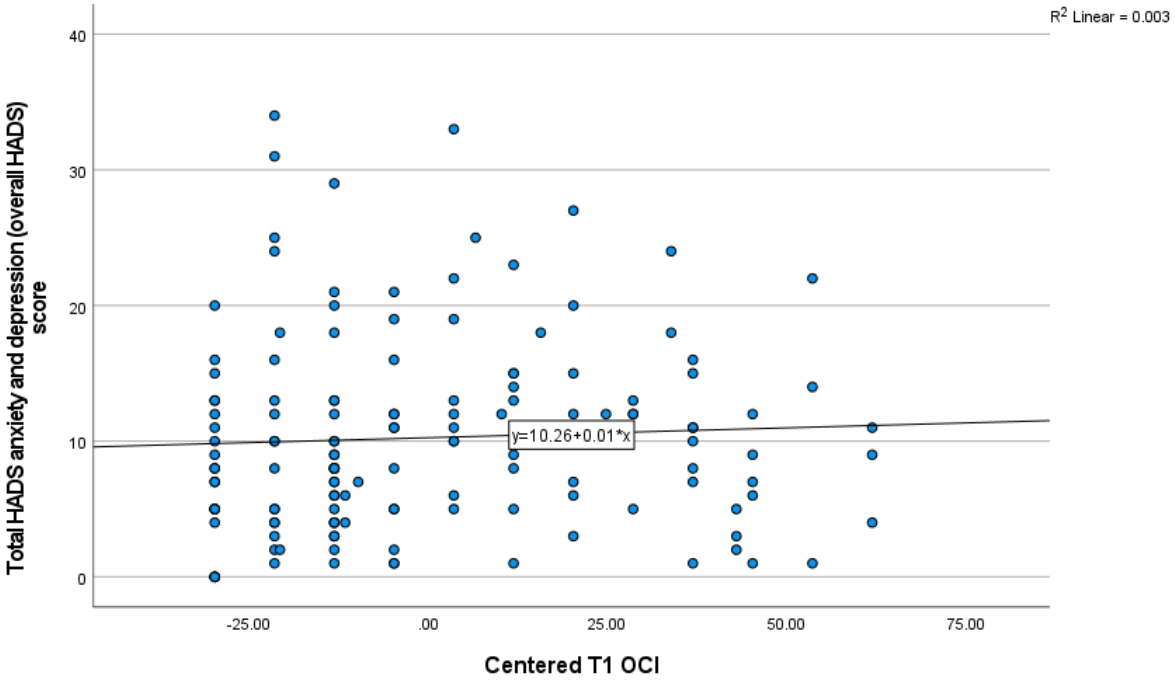
The Durbin-Watson statistic was 2.1, indicating that the values of the residuals are independent, and this assumption has been met.

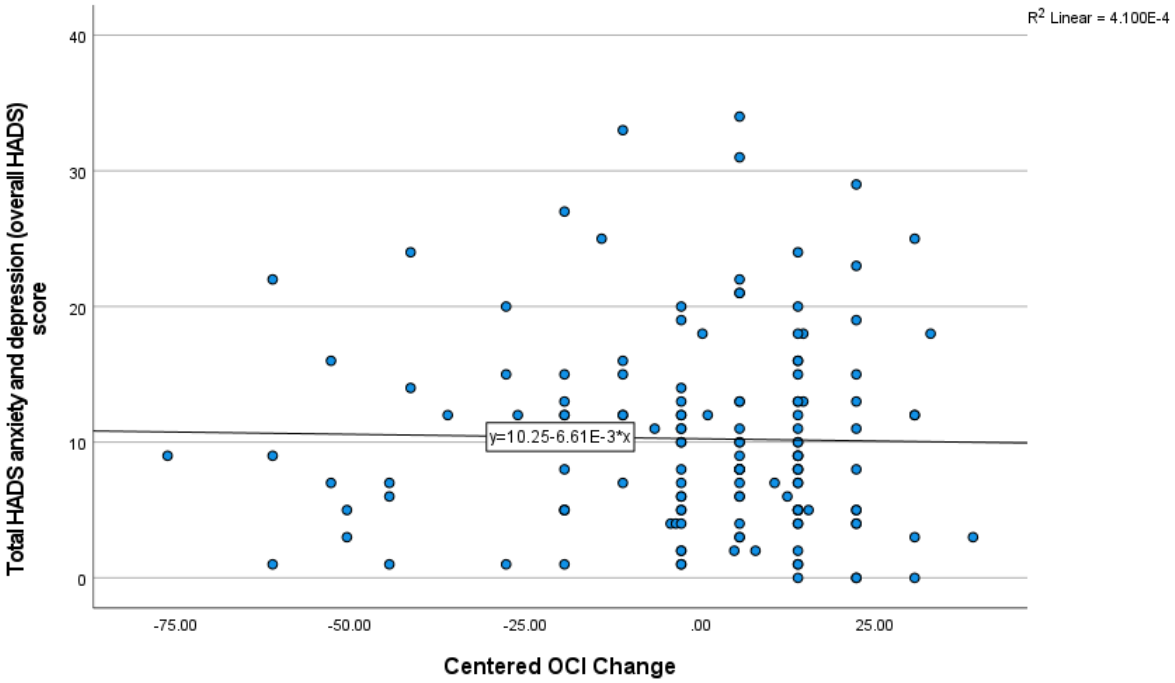
Linearity

Scatterplots were used to examine the relationship between the IVs and the DV and show that this assumption was met.



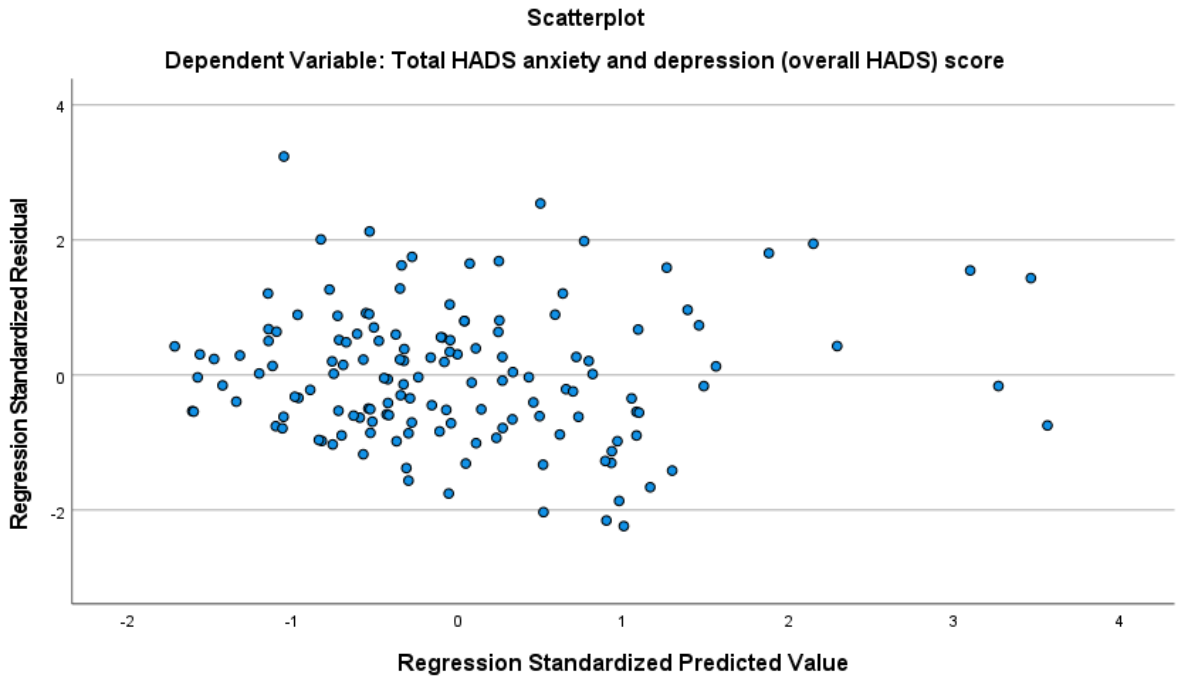



R2 Linear = 0.003
Total HADS anxiety and depression (overall HADS) score
40
30
20
10
0
y=10.26+0.01*x
-25.00
.00
25.00
50.00
75.00
Centered T1 OCI


R2 Linear = 4.100E-4
Total HADS anxiety and depression (overall HADS) score
40
30
20
10
0
y=10.25-6.61E-3*x
-75.00
-50.00
-25.00
.00
25.00
Centered OCI Change

Homoscedasticity

The variance of the standard residuals appears constant, in that the plot of standardised residuals vs standardised predicted values showed no obvious signs of funnelling, suggesting the assumption of homoscedasticity was met.



Absence of Multicollinearity

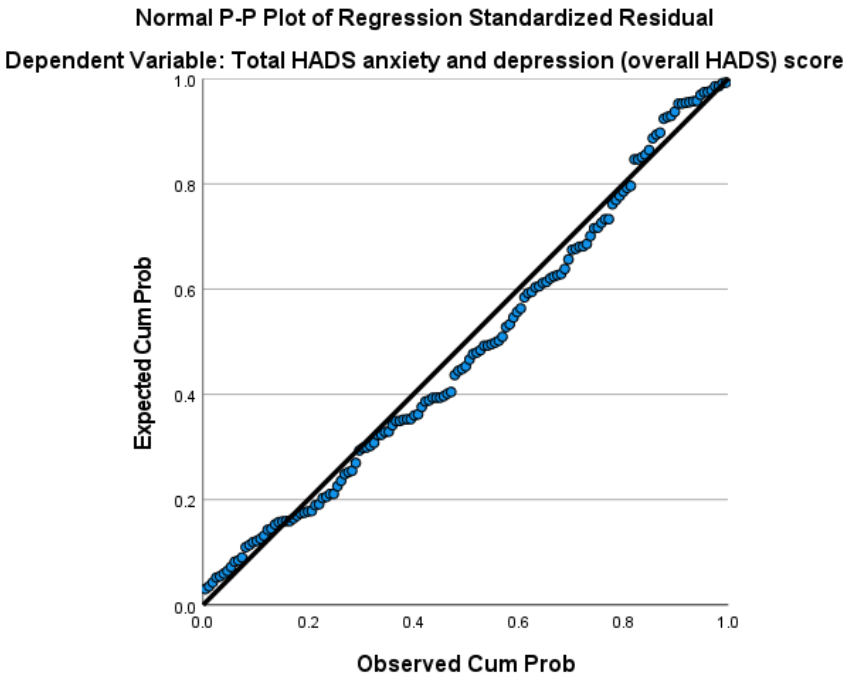
Correlation coefficients and Tolerance/VIF values were inspected for possible multicollinearity. The correlation coefficient was -.77 for the relationship between centred cognitive change and centred OCI at T1, suggesting possible multicollinearity. Correlation coefficients were <0.8 for the remaining variables. VIF scores were <10, and Tolerance scores were >0.2 (statistics = 1.2 – 6.0 and 0.2 – 0.87 respectively). Thus, it was decided that the assumption had been met.

Absence of Significant Outliers

Cook's Distance values were all <1, suggesting that individual cases were not unduly influencing the model, thus this assumption was met.

Normal Distribution of Residuals

A P-P plot shows that there is a reasonably good linear relationship between residuals, suggesting that they are normally distributed, and this assumption is met.



Appendix 3.K

Moderation Analysis Code for the PROCESS Tool

The PROCESS plug-in transformed predictors using grand mean centring and created the interaction variable (cognitive change × OCI_{T1}). The plug-in then runs a forced entry regression with the covariates, the centred predictor variable, the centred moderator variable, and the interaction of the two centred variables respectively.

```
Run MATRIX procedure:

***************** PROCESS Procedure for SPSS Version 3.5 *****************

          Written by Andrew F. Hayes, Ph.D.       www.afhayes.com
    Documentation available in Hayes (2018). www.guilford.com/p/hayes3

**************************************************************************
Model  : 1
    Y  : HADS
    X  : Cog_cge
    W  : OCI_sAc

Covariates:
 Barthel  Aware

Sample
Size:  143

**************************************************************************
OUTCOME VARIABLE:
 HADS

Model Summary
          R       R-sq        MSE          F        df1        df2          p
      .6251      .3908    32.5402    17.5747     5.0000   137.0000      .0000

Model
              coeff         se          t          p       LLCOGNITIVE IMPAIRMENT
ULCI
constant     -.4909     3.4291     -.1431      .8864    -7.2718     6.2900
Cog_cge       .1913      .0535     3.5734      .0005      .0855      .2972
OCI_sAc       .2226      .0461     4.8338      .0000      .1316      .3137
Int_1         .0020      .0009     2.2091      .0288      .0002      .0038
Barthel      -.4212      .1158    -3.6364      .0004     -.6502     -.1921
Aware         .3705      .0495     7.4883      .0000      .2727      .4683

Product terms key:
 Int_1    :        Cog_cge  x        OCI_sAc

Covariance matrix of regression parameter estimates:
          constant    Cog_cge    OCI_sAc      Int_1    Barthel      Aware
constant    11.7590     -.1142     -.1136      .0000     -.2767     -.1399
Cog_cge      -.1142      .0029      .0021      .0000      .0015      .0016
OCI_sAc      -.1136      .0021      .0021      .0000      .0019      .0017
Int_1         .0000      .0000      .0000      .0000      .0000      .0000
Barthel      -.2767      .0015      .0019      .0000      .0134      .0012
Aware        -.1399      .0016      .0017      .0000      .0012      .0024

Test(s) of highest order unconditional interaction(s):
       R2-chng          F        df1        df2          p
X*W      .0217     4.8802     1.0000   137.0000      .0288
----------
    Focal predict: Cog_cge  (X)
```

```
          Mod var: OCI_sAc  (W)

Conditional effects of the focal predictor at values of the moderator(s):

    OCI_sAc     Effect         se          t          p       LLCOGNITIVE
IMPAIRMENT      ULCI
   -21.4198      .1484      .0632     2.3458      .0204      .0233      .2734
    -9.7531      .1718      .0574     2.9951      .0033      .0584      .2852
    33.8832      .2593      .0512     5.0658      .0000      .1581      .3606

Moderator value(s) defining Johnson-Neyman significance region(s):
      Value    % below    % above
   -28.7805    15.3846    84.6154

Conditional effect of focal predictor at values of the moderator:
    OCI_sAc     Effect         se          t          p       LLCOGNITIVE
IMPAIRMENT      ULCI
   -29.7531      .1316      .0682     1.9314      .0555     -.0031      .2664
   -28.7805      .1336      .0676     1.9774      .0500      .0000      .2672
   -25.1698      .1408      .0654     2.1537      .0330      .0115      .2702
   -20.5865      .1500      .0628     2.3897      .0182      .0259      .2742
   -16.0031      .1592      .0604     2.6385      .0093      .0399      .2786
   -11.4198      .1684      .0581     2.8983      .0044      .0535      .2833
    -6.8365      .1776      .0561     3.1665      .0019      .0667      .2886
    -2.2531      .1868      .0543     3.4392      .0008      .0794      .2942
     2.3302      .1960      .0528     3.7113      .0003      .0916      .3005
     6.9135      .2052      .0516     3.9766      .0001      .1032      .3073
    11.4969      .2144      .0507     4.2284      .0000      .1141      .3147
    16.0802      .2236      .0501     4.4597      .0000      .1245      .3228
    20.6635      .2328      .0499     4.6640      .0000      .1341      .3315
    25.2469      .2420      .0500     4.8365      .0000      .1431      .3409
    29.8302      .2512      .0505     4.9741      .0000      .1513      .3511
    34.4135      .2604      .0513     5.0758      .0000      .1589      .3618
    38.9969      .2696      .0524     5.1428      .0000      .1659      .3732
    43.5802      .2788      .0538     5.1781      .0000      .1723      .3852
    48.1635      .2880      .0555     5.1856      .0000      .1782      .3978
    52.7469      .2972      .0575     5.1700      .0000      .1835      .4108
    57.3302      .3064      .0597     5.1357      .0000      .1884      .4243
    61.9135      .3156      .0620     5.0870      .0000      .1929      .4382

Data for visualizing the conditional effect of the focal predictor:
Paste text below into a SPSS syntax window and execute to produce plot.

DATA LIST FREE/
   Cog_cge    OCI_sAc    HADS       .
BEGIN DATA.
   -18.7434   -21.4198     3.5681
     6.2566   -21.4198     7.2772
    15.3172   -21.4198     8.6215
   -18.7434    -9.7531     5.7267
     6.2566    -9.7531    10.0211
    15.3172    -9.7531    11.5775
   -18.7434    33.8832    13.8006
     6.2566    33.8832    20.2838
    15.3172    33.8832    22.6335
END DATA.
GRAPH/SCATTERPLOT=
 Cog_cge  WITH     HADS     BY       OCI_sAc  .

************************ ANALYSIS NOTES AND ERRORS ************************

Level of confidence for all confidence intervals in output:
  95.0000

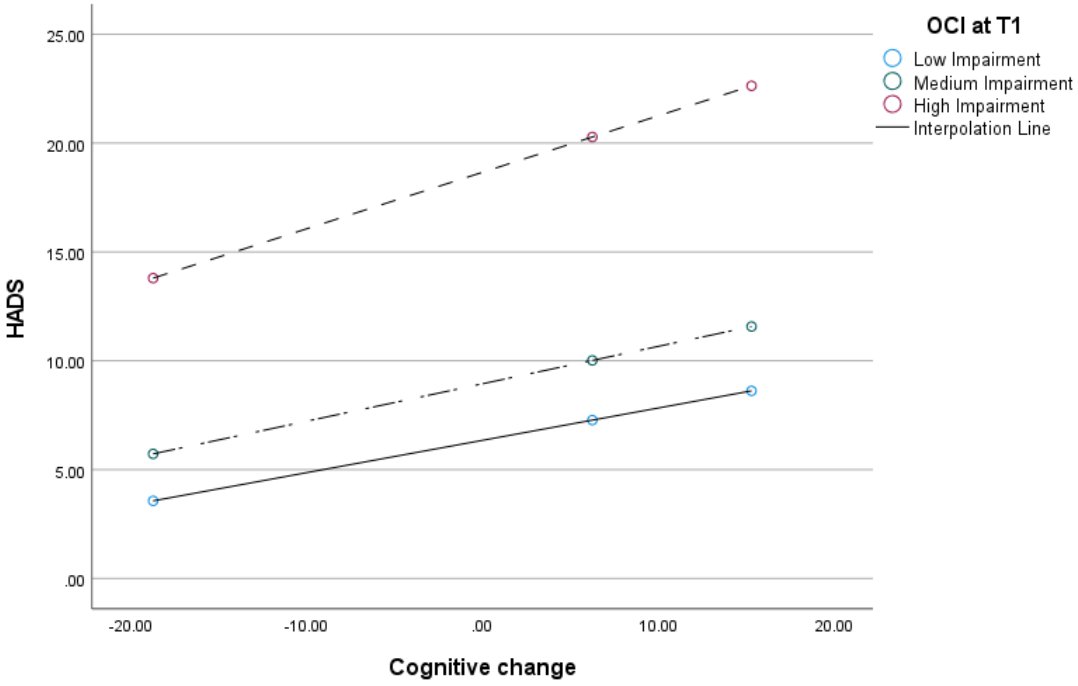
W values in conditional tables are the 16th, 50th, and 84th percentiles.

NOTE: The following variables were mean centered prior to analysis:
          OCI_sAc  Cog_cge

------ END MATRIX -----
```

Figure 3.K.1

Relationship Between Cognitive Change and HADS at Low, Medium, and High levels of OCI at T1



Appendix 3.L

Statistical Assumptions for the Exploratory Analyses

The logistic regression model was built systematically to choose the most parsimonious model as the final one. The final model indicated that BI and awareness significantly contributed to the model, but adding an interaction term had virtually no effect on the fit of the model, so the final model did not include this.

Variable Type

The DV was dichotomous. The IVs were continuous.

Independence of Observations

Different participants were in each group, with no participant being in more than one group.

Linearity

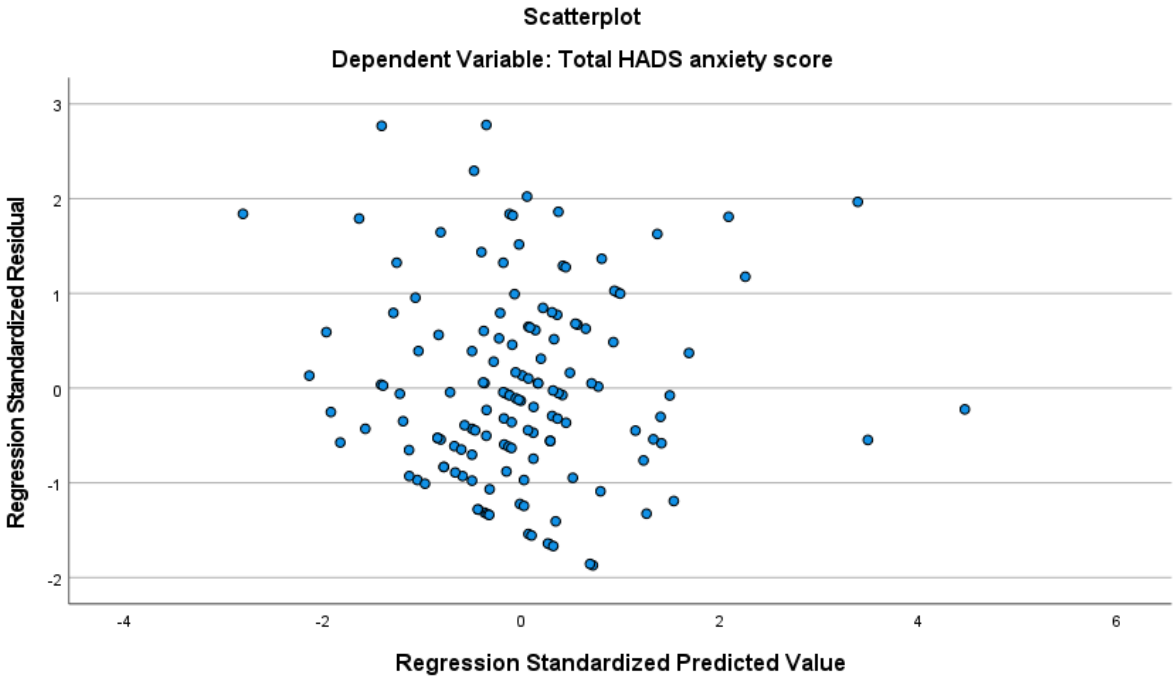
There needs to be a linear relationship between any continuous IVs and the logit transformation of the DV. New variables that are the log of the original variables for each of the predictor variables was computed. A forced entry binary logistic regression including interaction effects between the original and log of the original variables was carried out (e.g. Barthel × Ln(Barthel)). None of the interaction effects reached significance in either model, so the assumption of linearity of the logit was met.

Absence of Multicollinearity

Tolerance/VIF values were inspected for possible multicollinearity. VIF scores were well below 10, and tolerance scores were above 0.2 (statistics = 1.0 and 0.99 respectively). Thus, the assumption was met.

Independence of Errors

A scatterplot was used to explore independence of residuals. Most points fell between +3 and -3 on each axis, suggesting that this assumption is met. The Durbin-Watson statistic is 1.85, further indicating that the assumption is met.



Appendix 3.M

Additional Findings from the Exploratory Analyses

The anxiety model explained 18% (Nagelkerke R^2) of the variance in symptoms of anxiety. The model correctly classified 72.7% of cases, however it was more accurate at classifying cases not reporting clinically anxious symptoms (93.2% correct) than cases reporting clinically anxious symptoms (20%). The depression model explained 32% (Nagelkerke R^2) of the variance in symptoms of depression. The model correctly classified 83% of cases, however, it was more accurate at classifying cases not reporting clinical symptoms of depression (97.3% correct) than cases reporting clinical symptoms of depression (34%).