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Abstracts

Critical Review of the Literature

Autistic children's social skills do not always align with those of their neurotypical peers and research suggests that this can negatively impact quality of life. This review aimed to assess the effectiveness of Extended Reality interventions in helping autistic children to enhance their social skills. Five electronic databases were systematically searched and twenty-one studies were identified. The majority targeted social-emotional reciprocity and were of relatively low quality. There was insufficient evidence to determine whether effects were generalisable, sustained or important to autistic people. Research in this field is in its infancy and evidence of effectiveness should be viewed with caution. Future studies should aim for high-quality, theory-driven research, and involve autistic people to ensure meaningful outcomes. PROSPERO ID: CRD42021229442.

Key Words: Autism, Social Skills, Augmented Reality, Virtual Reality, Assistive Technology

Service Improvement Project

‘Patient activation’, defined as a patient’s knowledge, skills and confidence in managing their health condition, is essential for collaborative neurorehabilitation. It is therefore in the interest of neurorehabilitation services to monitor and seek to improve this. The process of increasing patient activation at a Community Neurorehabilitation Service in Buckinghamshire (CNRS) was hereby explored. This research used a mixed methods design to investigate whether patient activation increases throughout intervention with CNRS and if so, what factors contribute to this. Quantitative analysis showed that activation did increase significantly and this was not impacted by demographic factors. Qualitative interviews with eight former patients and four therapists suggested that the nature of the therapeutic relationship and wider social support may have been important in contributing to the change. Negative past experiences of services and cognitive difficulties seemed to be associated with lower activation. As a result, CNRS embedded a number of service improvements including increased opportunities for peer support and greater consideration of the patient’s wider health journey and emotional difficulties. Future research should seek to measure the impact of these improvements, as well as investigating the potential causal role of the factors identified in this qualitative research.

Key Words: Patient Activation, Neurological Conditions**,** Neurorehabilitation, Self-Management, Behaviour Change

Theoretically Driven Research Project

Understanding cognitive impairment and emotional difficulties after stroke are key priorities for research and clinical practice. However, cognitive screening tools are not always sensitive to impairments after stroke, and psychological distress in this population remains poorly understood, despite high prevalence and secondary implications. Part one of this research aimed to determine the sensitivity and clinical validity of a new stroke-specific, cognitive screening tool, the OCS-Plus, compared with the MoCA, a more established screening tool. Part two sought to better understand why cognitive impairment is associated with greater psychological distress after stroke, by exploring the role of illness appraisals. Eighty participants were consecutively recruited from Oxfordshire stroke wards over a 15-month period and completed a battery of assessments of mood, cognition and illness appraisals which were analysed cross-sectionally. The OCS-Plus was found to have good convergent validity and sensitivity when compared with the MoCA. Cognitive impairment significantly predicted depressive symptoms but was not predictive of anxiety when controlling for depression. The relationship with low mood was shown to be mediated by perceptions of control in this sample. Specifically, this research showed that higher levels of cognitive impairment were significantly associated with lower perceived control and accounted for the relationship between cognitive impairment and mood. Perceptions of threat and illness coherence were also predictive of low mood but were not predicted by degree of cognitive impairment. Illness appraisals may therefore be a target for intervention in the early stages after stroke, although prospective research is required to establish causality.

Key Words: Stroke, Cognitive Impairment, Cognitive Screening, Post-Stroke Depression, Illness Perceptions

Critical Review of the Literature

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Journal and Rationale: Review Journal of Autism and Developmental Disorders (Published April 2022 – Appendix N). This was chosen due to its publication of critical reviews of topics relating to autism spectrum and developmental disorders (submission guidelines included in Appendix A).

Abstract

Autistic children's social skills do not always align with those of their neurotypical peers and research suggests that this can negatively impact quality of life. This review aimed to assess the effectiveness of Extended Reality interventions in helping autistic children to enhance their social skills. Five electronic databases were systematically searched and twenty-one studies were identified. The majority targeted social-emotional reciprocity and were of relatively low quality. There was insufficient evidence to determine whether effects were generalisable, sustained or important to autistic people. Research in this field is in its infancy and evidence of effectiveness should be viewed with caution. Future studies should aim for high-quality, theory-driven research, and involve autistic people to ensure meaningful outcomes. PROSPERO ID: CRD42021229442.

Key Words: Autism, Social Skills, Augmented Reality, Virtual Reality, Assistive Technology

Are Extended Reality Interventions Effective in Helping Autistic Children Enhance their Social Skills? A Systematic Review

Autism and Social Differences

Effective social communication depends on a range of *social skills*, a term which is complex and has been variably defined across the literature, but is broadly considered to encompass verbal and non-verbal behaviours that enable positive social interactions (Gresham & Elliott, 1984). This multifaceted concept includes innate skills, such as eye tracking and a preference for faces, as well as more complex, learned behaviours, such as adjusting behaviour to suit particular social contexts (Spence, 2003). An exhaustive list of skills considered to be *social* is difficult to establish, due to the number of cognitive and behavioural processes drawn upon in any given social situation.

Autistic[1] children's social skills do not always align with those of neurotypical people. Their skills are different in terms of social-emotional reciprocity (e.g. social approach and initiation of interactions), nonverbal communicative behaviours (e.g. eye contact, and use of facial expressions), and the development, maintenance and understanding of relationships (e.g. reduced interest in peers and difficulties making friends); Diagnostic and Statistical Manual of Mental Disorders, 5th ed.; DSM-5; American Psychiatric Association [APA], (2013). These differences manifest differently for each individual (Happé et al., 2006) and can have negative secondary effects on wellbeing, learning and relationships. For example, social skills which do not align with

[1] Identity first language is used within this article as this is the preferred language of many adults on the autism spectrum, i.e. 'autistic individual' rather than 'individual with autism' (Kenny et al., 2016). However, there is a lack of information about the preferred language of autistic children.

neurotypical people have been shown to predict social anxiety (Bellini, 2006) and are associated with social rejection from peers (Ochs et al., 2001). This is not through lack of interest in social contact (Jaswal & Akhtar, 2019); autistic young people often express a desire for social interaction and report being more lonely than their neurotypical peers (Bauminger & Kasari, 2000). Furthermore, social differences in autistic people can have a negative effect on academic and occupational outcomes (Howlin, 2003; Welsh et al., 2001).

Theories of Social Difference in Autism

A number of theories might explain the social differences observed in autism. One theory proposes that autistic people can find it harder to make predictions about others' mental states (Theory of Mind) and so may find it difficult to encode socially relevant information (Baron-Cohen et al., 1985). Alternatively, it may be that difficulty understanding others' interpretations of the world is due to an underlying difference in executive functioning, such as rule learning (J. Russell, 1997). This can account for the finding that some autistic children struggle with Theory of Mind style tasks, even when the social element is removed (J. Russell et al., 1991). In particular, predictive processing might be different in autism. Pellicano and Burr (2012) propose that autistic people perceive the world more accurately, as they are less biased by prior experience when making predictions. At times this may be a strength, but could make some elements of social interaction more difficult, such as predicting when another person will speak (Stark et al., 2021). Another cognitive theory of autism is 'Weak Central Coherence' (Frith, 1989) which suggests that autistic people have a preference for local processing rather than striving for overall meaning. This may lead to differences in the interpretation of social behaviours. However, more recent evidence suggests that this theory sits alongside social differences, rather than explains them (Happé & Frith,

2006). These cognitive theories are in line with evidence that social desire is not affected by autism (Happé & Frith, 1996).

In contrast, Social Motivation Theory proposes that autistic people have differences in the psychological dispositions and biological mechanisms that bias neurotypical individuals to orient towards social stimuli, seek and derive pleasure from social interactions, and work to maintain social bonds (Chevallier et al., 2012). However, this contrasts with many accounts of autistic people, overlooks the role of other people's perceptions and responses, and ignores the many other explanations of social differences in autism (Jaswal & Akhtar, 2019). Furthermore, this theory has led to differences, such as limited eye contact, being prioritised as intervention targets, despite evidence for the positive effects of gaze aversion on concentration and regulation of emotions (Robledo et al., 2012).

Social Skills Interventions

A number of interventions have sought to teach social skills to autistic children, due to the secondary benefits on wellbeing and quality of life shown by previous research (McConnell, 2002). A range of outcomes have been targeted, such as turn taking, initiating interaction, recognising emotions and understanding Theory of Mind (Begeer et al., 2011; Matson et al., 2007; Rieth et al., 2014). Well-established interventions include Social Stories ™ (Karkhaneh et al., 2010), video modelling (Sng et al., 2014), pivotal response training (Koegel et al., 2016), social skills groups (Reichow et al., 2012) and peer mediated behavioural interventions (Laushey & Heflin, 2000). There is increasing evidence that, at least in some contexts, these interventions can help autistic children to utilise neurotypical social skills but the duration, generalisability and precise mechanism of change remains unclear (McConnell, 2002).

Furthermore, little acknowledgement has been given to the fact that it may be effortful, and perhaps emotionally draining for autistic people to operationalise these skills.

When considering social skills interventions, it is important to acknowledge that social differences in autism do not necessarily equate to a 'difficulty'. Many argue that difficulties arise due to a lack of accommodation of autistic differences in a neurotypical society (Brownlow, 2010). Social skills training can perpetuate the idea that autistic people need to adapt, rather than wider society accommodating for neurodiversity. However, many autistic people already find ways to overcome social challenges and seek support in doing this (Cresswell et al., 2019). Social skills interventions are one way in which services can support people to overcome these challenges, particularly in circumstances where it is not possible to adapt the environment e.g. public places.

Extended Reality Technologies

Over the last three decades, social skills interventions have begun to capitalise on the potential advantages of digital technologies. Computerised approaches can support social skills *in situ* (Vygotsky & Luria, 1994) or improve traditional training approaches, due to the attentional and motivational advantages of interactive technologies (Golan & Baron-Cohen, 2006). More recently, these advantages have been exploited using Extended Reality (XR), i.e. the merging of physical and virtual realities (Riva et al., 2016). This encompasses Augmented Reality (AR), in which virtual information is overlaid onto the real world, Virtual Reality (VR), in which users are fully immersed in a simulated environment and Mixed Reality (MR), in which digital and real-world objects interact in real-time. By merging physical and virtual realities, XR could reduce the salience of potentially distressing stimuli, making interactions less overwhelming for some autistic people, thus providing an effective space to experiment

with social skills (Thye et al., 2018). In virtual social settings, there is no risk of embarrassment in front of one's peers, yet the degree of realism may increase the likelihood of generalisability (Strickland et al., 1996).

Extended reality interventions have been successfully implemented in a number of therapeutic contexts including stroke rehabilitation (Laver et al., 2017) and exposure therapies for anxiety disorders (Bouchard et al., 2017; Vincelli et al., 2003). Applications for autistic people are also emerging, although the majority of interventions to date have used Virtual Learning Environments (VLEs), a non-immersive technology in which users interact with simulated social situations using a desktop computer (e.g. Didehbani et al., 2016). More immersive technologies tend to require a Head-Mounted Display (HMD) or Smartglasses, computer glasses that change what the wearer sees. Despite the potential challenges for those with tactile hypersensitivities, they have been shown to be well-tolerated, enjoyable and engaging in this population (Keshav et al., 2017; Newbutt et al., 2016), and effective in teaching other skills, such as taking public transport (Simoes et al., 2018).

Aims of this Review

Previous reviews have shown that XR interventions are feasible and sometimes beneficial for teaching autistic children a range of skills, although recognise that the quality of the literature is varied (e.g. Berenguer et al., 2020; Mesa-Gresa et al., 2018). However, no review to date has investigated the specific potential of XR for social skills. Furthermore, previous reviews of VR interventions have tended to include non-immersive technologies, such as VLEs. This review takes a focused approach by reviewing the evidence base for interventions in which one's perception of reality is altered. Social skills interventions have most commonly been implemented in children

but we chose to include young adults, based on evidence that brain maturation, particularly that of the prefrontal cortex which coordinates the goal directed behaviour needed to implement social skills, continues to develop into a person's early twenties (Johnson et al., 2009). This is also in line with current plans to extend children's mental health services in England up to 25 years (NHS Long Term Plan, 2019). Due to the infancy of this research area, we chose not to restrict inclusion on the basis of study design.

It was considered important to understand the effectiveness of these interventions because of the secondary benefits social skills can have on indicators of wellbeing, such as increased peer interactions (Mandelberg et al., 2014) and reduced anxiety and low mood (Hillier et al., 2011; Schohl et al., 2014). Secondly, the realism of immersive VR offers a unique potential to improve upon existing social skills interventions which do not always generalise to real contexts (Bellini et al., 2007). Thirdly, XR is at a point of rapid expansion, with VR headsets becoming increasingly widespread. If social skills interventions are to capitalise on this expansion, it is important to consider whether there is any good quality evidence for their effectiveness and whether investment in these technologies will actually lead to meaningful benefits for autistic young people.

To this end, this review aims to answer the following questions:

1. Is there any evidence that Extended Reality interventions are effective in helping autistic children and young people to enhance their social skills?
2. What is the quality of the evidence for observed effects?
3. To what extent have observed effects been shown to lead to meaningful change for autistic people?

Method

This quantitative systematic review uses synthesis without meta-analysis methodology and information is reported in line with Cochrane guidance (Garritty et al., 2021). The protocol was pre-registered with PROSPERO (ID: CRD42021229442; available at https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=229442). Five online databases (PsychInfo, Medline, Pubmed, EMBASE, CINAHL) were searched using key terms listed in Appendix B. All retrieved datasets were downloaded into a Zotero library and exported to excel for first author screening by title and abstract, then by full text. Another author (EM) screened 23% of studies at initial screening and 24% of studies at full text screening. All texts accepted at final screening were forward and backward citation searched using Scopus.

Screening and Selection

Studies were screened for eligibility according to the following inclusion criteria:

- The paper was published in English, in a peer reviewed journal to ensure a minimum quality level.
- The paper was published after 1990, when AR was developed and VR headsets became widely commercially available.
- The paper included an active intervention using XR. Studies described as VR but which used a desktop computer or motion based video game were not included for review as they do not to extend one's perception of reality.
- The study targeted one or more social skill.

- The paper reported a quantitative measure of social skills, at least pre and post intervention. This included *in vivo* outcomes, as well as validated and idiosyncratic measures of social skills.
- Study participants were children and young adults up to the age of 25.
- Study participants were autistic.

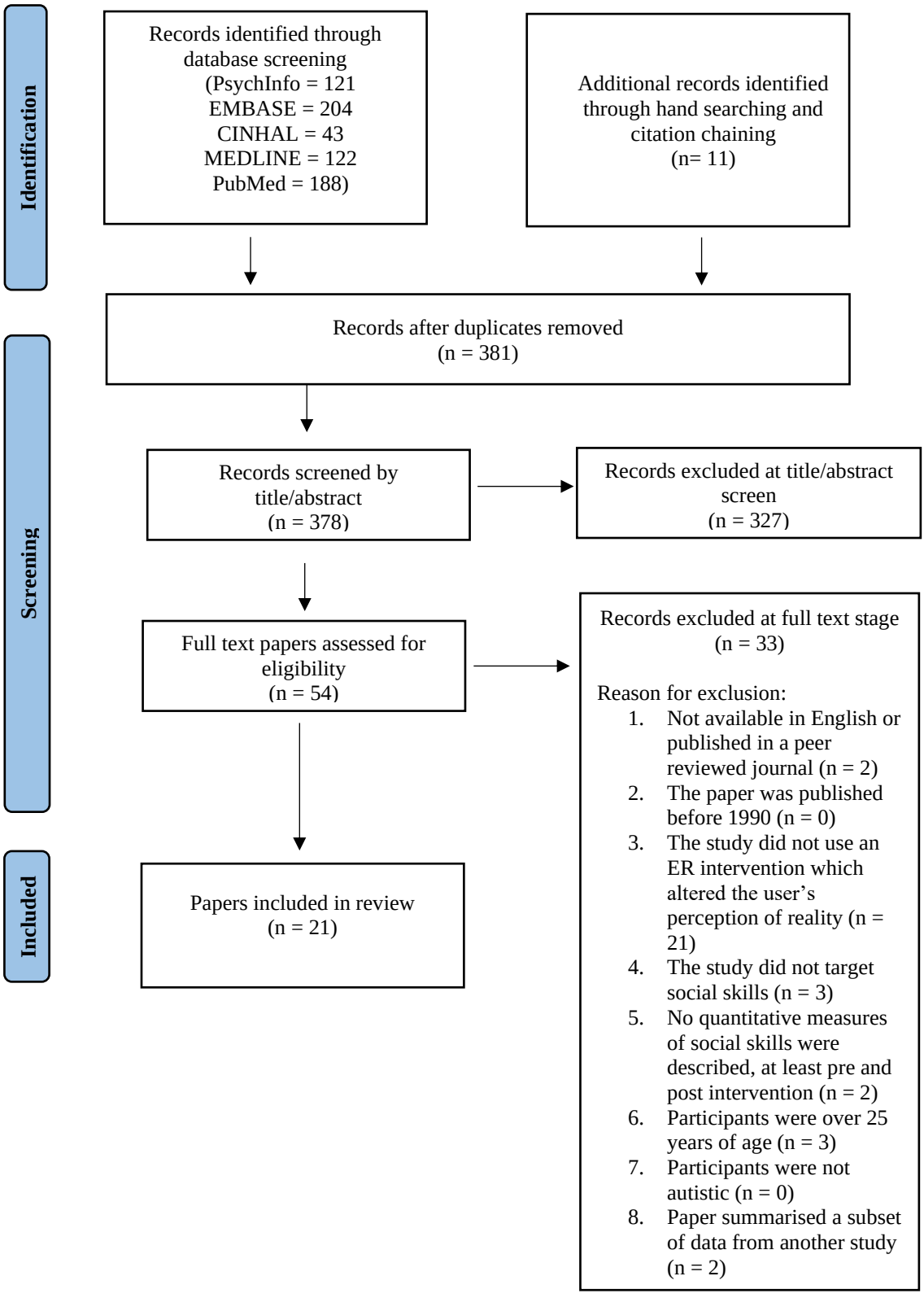
Where papers reported on more than one study, they were included based on the study which met the inclusion criteria. Borderline cases were discussed amongst the researchers.

Study Selection

The initial search identified 689 records, 381 of which were retained after duplicates were removed (Figure 1). 327 studies were excluded at the abstract stage, with a 96.5% inter-rater reliability (kappa = .87). Thirty-three studies were excluded at the full text screen, with a 91.7% inter-rater reliability (kappa = .83). Disagreements were resolved through further screening at the full text stage and discussion with other authors. Eleven additional records were identified through hand searching and citation chaining (Scopus search conducted August 8, 2022). A total of 21 studies were included in the review and a number of data items were extracted from each study. Where it was not possible to extract data directly, study authors were contacted, or available information was used to calculate an effect size. 43% of data extraction was duplicated by a second reviewer (EM).

Figure 1

Study Selection Process



Data Synthesis

Data were synthesised and organised according to the social communication criteria of DSM-5's autism diagnostic criteria (APA, 2000), as the effect of XR on specific social skills was the primary area of interest. In line with Siddaway et al. (2019), findings were integrated and critiqued, not merely summarised. The high number of single case studies meant that it was not possible to conduct a meta-analysis but key information is summarised in tables, with a summary of effect (Cohen's *d*) reported where enough information was provided in the paper or could be obtained through correspondence with the study authors. All studies were included for synthesis, irrespective of quality, but due to the range of study quality, summary plots were not developed, to avoid misrepresentation of the evidence base.

Quality Assessment

The quality and risk of bias of each study was evaluated using a quality checklist for healthcare intervention studies, developed by Downs and Black (1998). The tool was selected due to its applicability to both randomised and non-randomised studies. All papers were rated by the first author and 24% were duplicated by another author (EM), with an interrater reliability of 90.4% (kappa = 0.86). Discrepancies were resolved through discussion. Some items were not applicable to small-N research and so a summary score relative to the number of items assessed was generated for each paper to allow for direct comparison across studies. This was calculated by dividing the score obtained by the total possible score according to study design. A risk of bias score was generated by the same method, using only items assessing bias (14 to 26). A full summary of the assessment approach is provided in Appendix C. Given that a number of items were not applicable to single case and small-N studies, the Single Case

Experimental Design (SCED) Scale (Tate et al., 2008) was also used to qualitatively assess these studies (Appendix D).

Results

Study Characteristics

Across the 21 studies (summarised in Table 1), 357 participants (15.7% female) aged 2 to 16 were included. Sample sizes ranged from one to 94 participants (M = 17.00, SD = 19.92). All participants were autistic however, only 14 studies reported that this was a clinical diagnosis according to DSM and only four confirmed diagnosis using a screening tool: Ip et al. (2018) using the Childhood Autism Spectrum Test (Williams et al., 2005); Liu et al. (2017) using the Social Communication Questionnaire (Chandler et al., 2007); Lorenzo et al. (2019) using the Autism Spectrum Inventory (Rivière, 2002); and Frolli et al. (2022) using the Autism Diagnostic Observation Schedule (ADOS-2, Lord et al., 2012). Twelve studies specified an Intelligence Quotient (IQ), with a mean or lower limit of at least 70. The studies used a range of designs, although a majority (11) were small-N, with either multiple baseline or pre-post measures. Only seven studies used a control (Crowell et al., 2020; Frolli et al., 2022; Herrero & Lorenzo, 2020; Ip et al., 2018; Lorenzo et al., 2016, 2019; Wedyan et al., 2021). Seven studies explicitly reported being at the preliminary or feasibility stage of the research.

Fifteen unique interventions were evaluated (eight AR; six VR; one MR) and are described in Table 2. Nine adapted existing social skills interventions, such as video modelling (Chen et al., 2016), collaborative game play (Mora-Guiard et al., 2017) and Social Stories ™ (Herrero & Lorenzo, 2020). Rationale for the use of XR, included distraction reduction; the potential for engagement and sustained attention; freedom to practice social interactions without risk; and customisability i.e. the capacity to

emphasise social stimuli and adapt difficulty level. Interventions most often took place in schools, and ranged from one 15 minute session (Crowell et al., 2020) to eighty 25 minute sessions (Lorenzo et al., 2013). Some studies gave rationale for these design decisions, such as short sessions due to attentional capacity, but the majority gave no justification for the frequency or duration of XR use. Adverse outcomes were not routinely assessed but one study commented that some children had difficulties with restlessness and tolerance of the headset (Ravindran et al., 2019) and another used ‘reinforcement candy’ to encourage ‘emotional stability’ during the intervention (Cheng et al., 2015).

Table 1

Summary of Included Studies

Study	Design	Country of Study	Name of Intervention (AR/VR/MR)	Autism Diagnosis and Assessment Criteria	Age Range of Participants (years)	IQ	Total Sample Size included in analysis (% female)	Social Skills Targeted	Measure of Social Skills (Validated?)
Liu et al. (2017)	Single Case Series, Pre-Post	United States	Brain Power System - Face Game and Emotion Game (AR)	Clinical diagnosis according to DSM-5	8 - 9	Not specified	2 (0%)	**Social Emotional Reciprocity & Non-Verbal Communication –** Social withdrawal and non-verbal communicative behaviours e.g. eye contact	ABC (✓) and Caregiver Report (x)
Sahin et al. (2018)	Single Case Study	United States	Empowered Brain – Face2Face Module (AR)	Clinical diagnosis but criteria not specified	13	Not specified	1 (0%)	**Social Emotional Reciprocity –** Social awareness, cognition, communication, motivation and restricted interests	SRS-2 (✓)
Vahabzadeh et al. (2018)	Single Case Series, Pre-Post	United States	Empowered Brain - Face2Face Module (AR)	Clinical diagnosis according to DSM-IV or DSM-V	6.7 - 8.8	Not specified	4 (0%)	**Social Emotional Reciprocity –** A range of behaviours indicating social reciprocity	ABC (✓)

Study	Design	Country of Study	Name of Intervention (AR/VR/MR)	Autism Diagnosis and Assessment Criteria	Age Range of Participants (years)	IQ	Total Sample Size included in analysis (% female)	Social Skills Targeted	Measure of Social Skills (Validated?)
Lee (2020)	Multiple Baseline Experimental Design	Taiwan	Real Time Skeletal Tracking System (AR)	Clinical diagnosis according to DSM-IV-TR	7 – 9	M = 102.33	3 (33.3%)	**Social Emotional Reciprocity –** Understanding of social greetings	Novel situation based questions (x); Idiosyncratic ratings of behaviours (x)
Lee et al. (2018)	Multiple Baseline Experimental Design	Taiwan	Augmented Reality Concept Map Training System (AR)	Clinical diagnosis according to DSM-IV-TR	8 – 9	M = 93.3	3 (33.3%)	**Social Emotional Reciprocity –** Understanding of social greetings	Novel situation-based questions (x); Idiosyncratic ratings of behaviours (x)
Lorenzo et al. (2019)	Case controlled, Pre-Post (Non-randomised control)	Spain	Quiver Vision (AR)	Clinical diagnosis according to DSM-V and score >30 on IAS	2 – 6	7 participants had a diagnosis of Developmental Delay, Intellectual Disability or Language Disorder	11 (9.1%)	**Social Emotional Reciprocity –** Unclear target behaviour but domains of the IDEA include 'social relationship'; 'joint reference skills'; 'intersubjective and mentalistic skills'	IDEA (validation unknown due to publication in Spanish)
Cheng et al. (2015)	Multiple Baseline Experimental Design	Taiwan	3D-SU System (VR)	Diagnostic criteria not specified	10 - 12	M = 82	3 (0%)	**Social Emotional Reciprocity –** Understanding of neurotypical social initiations	Novel situation based questions (x)

Study	Design	Country of Study	Name of Intervention (AR/VR/MR)	Autism Diagnosis and Assessment Criteria	Age Range of Participants (years)	IQ	Total Sample Size included in analysis (% female)	Social Skills Targeted	Measure of Social Skills (Validated?)
Herrero & Lorenzo (2020)	Randomised Controlled Trial (Parallel Group Design)	Spain	HMD Immersive VR System (VR)	Diagnostic criteria not specified	8 – 15	Not specified	14 (14.3% of experimental group, control not specified)	**Social Emotional Reciprocity & Non-verbal Communication** - Social reciprocity, flexibility to change and non-verbal communication skills	Idiosyncratic ratings of *in situ* social reciprocity (x)
Ip et al. (2018)	Randomised Controlled Trial (Parallel Group Design)	Hong Kong	Cave Automatic Virtual Environment (CAVE; VR)	Clinical diagnosis but diagnostic criteria unknown (mean score on Childhood Autism Spectrum Test above cut off, 19.1)	7 – 10	95 (non-verbal)	72 (11.1%)	**Social Emotional Reciprocity & Understanding Relationships** – Social reciprocity; emotion recognition; emotion regulation and everyday social skills e.g. maintaining relationships	PEP-3 (✓); Adapted Faces test (x); Eyes test (✓); ABAS-II (✓)
Lorenzo et al. (2013)	Quasi-experimental, Pre-Post (No control)	Spain	Immersive Virtual Reality System (VR)	Asperger's Syndrome, diagnostic criteria not specified	8 - 15	'Average Intelligence Levels' stated but not specified	20 (20%)	**Social Emotional Reciprocity –** No one primary outcome measure	Idiosyncratic ratings of *in situ* performance (x); Teacher ratings of classroom behaviours (x)

Study	Design	Country of Study	Name of Intervention (AR/VR/MR)	Autism Diagnosis and Assessment Criteria	Age Range of Participants (years)	IQ	Total Sample Size included in analysis (% female)	Social Skills Targeted	Measure of Social Skills (Validated?)
Lorenzo et al. (2016)	Randomised Controlled Trial (Parallel Group Design)	Spain	Immersive Virtual Reality System (VR)	Diagnosis according to DSM-V criteria	8– 12	Not specified	40 (27.5%)	**Social Emotional Reciprocity** – A range of behaviours indicating social reciprocity	Idiosyncratic ratings of *in situ* performance (x); Teacher ratings of classroom behaviours (x)
Ravindran et al. (2019)	Single Case Series, Pre-Post	United States	Floreo's Joint Attention Module (VR)	Diagnosis confirmed by a family member	9 – 16	Not specified	12 (16.7%)	**Social Emotional Reciprocity & Non-verbal Communication –** Frequency of interactions and eye contact	Joint Attention Assessment (x)
Crowell et al. (2020)	Randomised Controlled Trial (Crossover Design)	Spain	Lands of Fog (MR)	Clinical diagnosis according to DSM -IV-TR	8 – 12	>70	18 (16.7% assumed)	**Social Emotional Reciprocity** – Social initiations and responses in a collaborative game	Frequency of *in situ* social initiations and responses (x)
Mora-Guiard et al. (2017)	Quasi-experimental, Repeated Measures (No control)	Spain	Lands of Fog (MR)	Clinical diagnosis according to DSM-IV-TR	10 – 14	>70	10 (0%)	**Social Emotional Reciprocity** – Social initiations, acts and responses in a collaborative game	Frequency of *in situ* social initiations, acts and responses (x)

Study	Design	Country of Study	Name of Intervention (AR/VR/MR)	Autism Diagnosis and Assessment Criteria	Age Range of Participants (years)	IQ	Total Sample Size included in analysis (% female)	Social Skills Targeted	Measure of Social Skills (Validated?)
Sayis et al. (2022)	Randomised Controlled Trial (Crossover Design)	Spain	Lands of Fog (MR)	Clinical Diagnosis made using the ADOS	8 - 12	>70	36 (16.7% assumed)	**Social Emotional Reciprocity** – Social initiations, responses and externalisations in a collaborative game	Frequency of *in situ* social initiations, responses and externalisations (x)
Tsai et al. (2020)	Multiple Baseline Experimental Design	Taiwan	Cave Automatic Virtual Environment (CAVE; VR)	Clinical diagnosis according to DSM-5 criteria	7 – 9	M = 87	3 (0%)	**Social Emotional Reciprocity** – Understanding of social initiations in response to others' expressions	Novel situation-based questions (x); Idiosyncratic rating of role play behaviours (x)
Pérez-Fuster et al. (2022)	Multiple Baseline Experimental Design	Spain	The Pictogram Room (AR)	Clinical diagnosis according to DSM-5 criteria	3-8	All participants reported to have 'intellectual and language impairment)	6 (16.7%)	**Non-verbal Communication –** Eye-gaze responses to joint attention attempts	ADOS-2 (✓) ; ESCS (✓); Idiosyncratic measure of responses to joint attention (x)
Chen et al. (2015)	Multiple Baseline Experimental Design	Taiwan	Augmented Reality Self-Facial Modelling Learning System (AR)	Clinical diagnosis according to DSM-IV-TR	10 - 13	M = 101	3 (33.3%)	**Understanding Relationships –** Aimed to improve emotion recognition	Idiosyncratic measure of ability to identify facial expressions and emotions (x)

Study	Design	Country of Study	Name of Intervention (AR/VR/MR)	Autism Diagnosis and Assessment Criteria	Age Range of Participants (years)	IQ	Total Sample Size included in analysis (% female)	Social Skills Targeted	Measure of Social Skills (Validated?)
Chen et al. (2016)	Multiple Baseline Experimental Design	Taiwan	AR with Video Modelling Storybook Learning System (AR)	Clinical diagnosis according to DSM-IV-TR	11 - 13	M = 103.66	6 (17%)	**Understanding Relationships –** Aimed to improve emotion recognition	Idiosyncratic measure of ability to identify facial expressions and emotions (x)
Frolli et al. (2022)	Randomised Controlled Trial (Parallel Group Design)	Italy	VR Emotional Literacy Training (VR)	Clinical diagnosis according to DSM-5 criteria	9 - 10	M = 103	60 (15%)	**Understanding Relationships –** Aimed to improve emotion recognition	Idiosyncratic measure of ability to recognise emotions and related situations (x)
Wedyan et al. (2021)	Quasi-experimental, Pre-Post	Australia	Facial Expressions Training Application (AR)	Clinical diagnosis according to DSM-5 criteria	6 – 9	M = 125	30 (43.4%)	**Understanding Relationships –** Aimed to improve emotion recognition	Idiosyncratic measure of ability to identify facial expressions (x)

Table 2.

Description of AR, VR and MR Interventions and Their Reported Purpose

Intervention	Description
AR	
AR based Self-Facial Modelling Learning System	This system is designed for children to interact with while wearing masks which represent particular emotions. AR superimposes facial expressions onto a participant's view of themselves, to show the emotion most appropriate to the situation. This augmentation of the participant's view of themselves is thought to promote learning.
AR Video Modelling With Storybook Learning System	A tablet computer is used to augment a physical storybook with short video clips, which interactively emphasise social details. This draws attention to social elements of the story and is thought to be more engaging than traditional video modelling approaches.
Real Time Kinect Skeletal Tracking System	This system augments a virtual character with a user's body gestures and superimposes a range of real life backgrounds. This enables children to practice a range of body gestures which might be appropriate to different situations e.g. shaking hands or waving arms. The primary benefit is that it is real time but without the anxiety associated with real world interactions.
AR Concept Map Training System	A tablet computer is used to augment a physical concept map with 3D greeting animations and graphics to indicate relationships between people and degrees of closeness in relationships. The purpose is to help children to visually learn about basic social relations and appropriate greeting behaviours.
Empowered Brain/ Brain Power System	Children wear a pair of Smartglasses which can evaluate looking behaviours and superimpose digital text and images onto the world, so that social cues are given in real time according to the user's ability. It includes a number of applications: *Face Game (also termed Face2Face)* uses an AR game to draw attention to human faces with visual overlays such as arrows and cartoon masks which gradually fade. Users score points by directing their gaze towards their caregiver's face. *Emotion Game* asks children to identify the facial expression of the person in front of them by tilting their head towards the correct emoticon. Points are gained for correct answers.
Quiver Vision	A smartphone application is used to augment real life pictures so that they are perceived as 3D objects. In Lorenzo et al. (2019), participants engage in activities which involve interacting with the AR components alongside the therapist, such as touching different AR objects according to the therapist's instructions.
The Pictogram Room	This system augments a virtual character with a user's body movements which can be used to play a number of games. In Pérez-Fuster et al. (2022), users play the 'Gaze Following' game which involves selecting a window which the virtual character looks at. The software is thought to improve body awareness and scaffold learning of joint attention skills through the blending of a real-world experience with augmented elements.

Intervention	Description
Facial Expressions Training Application	Facial expressions are presented to the user as augmentations of reality, first as a graphical facial expression, then as a real facial expression. The user then identifies the correct expression with the support of their caregiver. This allows children to identify facial expressions in an environment that is reported to be safe, engaging and portable.
VR	
3D-SU System	Users view and interact with animated social events using an immersive VR headset. Based on their responses to questions about the events, users are rewarded with a message and applause or instructed to try again. Using a headset is thought to reduce distraction and encourage sustained attention towards social events.
Cave Automatic Virtual Environment (CAVE) VR	Users are immersed in a number of social scenarios within a 'cave' of multiple large projection screens. This allows children to practice their responses free from risk of embarrassment. Ip et al. (2018) employ this technology to allow children to practice responses in a range of situations. They are supported by a trainer if needed and a relaxation scenario is available if the experience becomes distressing. Tsai et al. (2020) enhance the CAVE with motion control cameras. Users are encouraged to interact with avatars within the environment in specific role playing games.
Floreo - Joint Attention Module	A smartphone is fitted to a Google Cardboard headset, creating a VR experience. Users work through a number of 'learning cards' which require the demonstration of joint attention skills. A virtual avatar initiates and responds to joint attention bids and can prompt where necessary. A monitoring therapist can observe using a tablet computer and provide further support.
Immersive Virtual Reality System	Users are immersed in a number of scenarios using two large projection screens and movement detection software. Lorenzo et al. (2013) use this system to allow children to rehearse their responses to a range of classroom based tasks such as initiating conversation with a classmate. In Lorenzo et al. (2016), participants follow an evaluator's guidance on how to behave in a range of social situations e.g. attending a birthday party. This approach is thought to be beneficial due to its realism and the capacity to present information clearly without irrelevant distractors.
HMD Immersive VR System	Users are immersed in two school environments which are viewed through a head mounted display. Children can socially interact with a number of avatars, whose responses are controlled by the researchers. The system is reported to be highly immersive and flexible to allow for adaptation and promote real world generalisability.
VR Emotional Literacy Training	Users are presented with two sequences of scenes referring to primary and secondary emotions, within a 3D virtual reality environment. The user in then presented with photos from the sequences and asked to recognise the emotion depicted in each. The approach is thought to be effective due to the user being immersed in the social context when learning the emotions.
MR	
Lands of Fog	Users collaborate with a partner to hunt for insects in a dense layer of virtual fog using a physical butterfly net. Explorative and collaborative efforts are rewarded with an upbeat tune or new feature. The approach is thought to be effective in encouraging collaboration as users can clearly see the actions of themselves and others, unlike in traditional collaborative game play.

Outcomes

Social-Emotional Reciprocity

Sixteen studies targeted social-emotional reciprocity: six using AR, seven using VR and three using MR. Specific targets included identifying and responding to social greetings; motivation to communicate with a conversation partner; understanding and use of social initiations; and sharing emotions and affect with others. Given the multiple countries in which these studies took place, it is possible that reciprocal behaviours considered socially appropriate differed according to cultural norms. Participants ranged from 6 to 16 years, although the majority of participants were primary school aged.

AR interventions included Smartglasses, which gave real-time prompts in social situations (Liu et al., 2017; Sahin et al., 2018; Vahabzadeh et al., 2018), a motion based game, which provided opportunities to practice reciprocal behaviours (Lee, 2020), and a concept map enhanced with AR to aid learning (Lee et al., 2018). One study used an AR smartphone app to enhance child-therapist interactions, but the specific effects of AR were unclear (Lorenzo et al., 2019). VR interventions were less varied, with the majority simulating social situations in which participants could practice specific reciprocal behaviours (Cheng et al., 2015; Herrero & Lorenzo, 2020; Ip et al., 2018; Lorenzo et al., 2013, 2016; Ravindran et al., 2019). One study utilised a specific role playing game within the VR (Tsai et al., 2020). Conversely, the MR intervention (Crowell et al., 2020; Mora-Guiard et al., 2017; Sayis et al., 2022) used mixed reality objects to digitally emphasise the benefits of collaboration with a partner.

Outcomes were most commonly measured by presenting participants with a range of social situations (sometimes in the form of Social Stories™) and asking them to describe, and/or demonstrate, how they might respond (Cheng et al., 2015; Herrero & Lorenzo, 2020;

Lee, 2020; Lee et al., 2018; Tsai et al., 2020). These studies all concluded that social-emotional reciprocity could be improved using their specific XR intervention however, the criteria for rating responses was not validated or tested for age appropriateness using a non-autistic sample. Furthermore, one study reported that participants were given prompts when responding but did not specify a protocol for this (Cheng et al., 2015). Another reported improvements in social reciprocity but assessed behaviours using a novel and unvalidated joint attention assessment (Ravindran et al., 2019). A number of studies measured outcomes by observing changes in behaviours during the XR intervention. Again, it was unclear whether the coded behaviours were age-appropriate, or had any relation to social skills in the real world. Of studies measuring *in situ* behaviours, two concluded that behaviours improved after the intervention (Herrero & Lorenzo, 2020; Mora-Guiard et al., 2017) and two found no impact of XR on social reciprocity when compared to an active control (Crowell et al., 2020; Sayis et al., 2022). Others did not set clear hypotheses regarding primary outcome measures, making it difficult to draw conclusions on their efficacy (Lorenzo et al., 2013, 2016). Only four of the studies targeting social-emotional reciprocity utilised validated outcome measures (Ip et al., 2018; Liu et al., 2017; Sahin et al., 2018; Vahabzadeh et al., 2018): the Aberrant Behaviour Checklist (ABC; Aman et al., 1985), Social Responsiveness Scale (SRS-2; Constantino & Gruber, 2012) and PEP-3 Psychoeducational Profile (Schopler et al., 2005). All of these studies showed an improvement in scores before and after the intervention, with a maximum follow up of 2 weeks (Ip et al., 2018). However, only in one instance was this in comparison to a control (Ip et al., 2018). One measure was unsuitable for the age of the study participants (Ip et al., 2018) and another indicated a 100% reduction in social difficulties (Liu et al., 2017) despite theoretical understandings of autism as a lifelong neurodevelopmental difference characterised by differences in social communication. One study measured outcomes using the Autism Spectrum Inventory (IDEA; Rivière, 2002). It is unclear whether

this is validated as the measure is in Spanish, but no improvements were observed when compared with an active control, and again, the theoretical justification for use of this measure was limited (Lorenzo, 2019).

Overall, a number of studies have targeted elements of social-emotional reciprocity and almost all claim evidence of effectiveness (see Table 3). However, there is significant risk of bias in how outcomes were measured and the better quality studies tended to be single case. It is therefore difficult to draw conclusions about the efficacy of XR for improving this aspect of social communication. Furthermore, despite some qualitative reports, the studies did not attempt to determine generalisability and lack of longer-term follow-up makes it difficult to determine the extent to which improvements were sustained.

Non-verbal Communicative Behaviours

One AR study targeted non-verbal communication as a primary outcome and three studies (two VR and one AR) included non-verbal behaviours amongst other outcome measures. Pérez-Fuster et al. (2022) aimed to improve responses to joint attention attempts, as measured using the ADOS-2 (Lord et al., 2012), the Early Social Communication Scales (ESCS, Mundy et al., 2003) and idiosyncratic measures. All six participants responded more frequently to joint attention attempts by eye gaze shifting after the AR intervention, although this was rated by the study's lead author. Herrero and Lorenzo (2020) used an idiosyncratic measure of non-verbal behaviours (e.g. use of facial cues, imitation and gestures) to suggest that use of their VR intervention was beneficial for 7- to 12-year-olds. However, all but one of the participants were rated as having 'fair' or 'good' non-verbal communication prior to the intervention and the measure used was not validated. Two studies concluded that their respective interventions increased eye contact: Ravindran et al. (2019) coded the amount of

eye contact used by the 9 to 16-year-old participants during a joint attention assessment and Liu et al. (2017) utilised an idiosyncratic caregiver measure.

Given the limited number of studies which have investigated non-verbal communicative behaviours, it is difficult to draw conclusions about XR's efficacy for this domain of social communication. This is compounded by potential bias in assessment methods, small sample sizes and unclear theoretical rationale for anticipated improvements in non-verbal behaviours. Only one study measured generalisability to interactions outside of the research environment (caregiver report).

Table 3

Summary of Results

Study	Quality Rating	Risk of Bias Rating	Effect of Intervention on Social Skills (+ indicates improvement and – indicates lack of improvement)
Liu et al. (2017)	0.61	0.67	Observed reduction in social withdrawal after using the Brain Power System (+) Carer reported increases in non-verbal communicative behaviours post-intervention (+)
Sahin et al. (2018)	0.56	0.83	Observed reduction in ratings of social skills difficulties measured by the SRS-2 after using Empowered Brain (+)
Vahabzadeh et al. (2018)	0.61	0.67	Observed reduction in social withdrawal after using Empowered Brain, at two time points (+)
Lee (2020)	0.58	0.5	Statistically significant mean improvement in understanding of social greetings (+)
Lee et al. (2018)	0.53	0.33	Statistically significant mean improvement in understanding of social greetings (+)
Lorenzo et al. (2019)	0.67	0.62	No significant difference between pre- and post- scores across all dimensions of the IDEA (-) No significant difference in post-intervention ratings between the intervention group and non-digital control for all items compared (-)
Cheng et al. (2015)	0.56	0.5	Observed increases in ability to understand social initiations across baseline, intervention and maintenance (+)
(Herrero & Lorenzo, 2020)	0.31	0.23	Observed improvements in social reciprocity and inflexibility to change post-intervention for all participants (+) Observed improvements in non-verbal communication post-intervention for 6 of 7 participants (+)
Ip et al. (2018)	0.48	0.54	Statistically significant improvement social reciprocity post-intervention (+) (*d*=0.50) Statistically significant improvement in emotion regulation post-intervention (+) (*d*=0.39) No significant difference in emotion recognition (Faces Test) post-intervention (-) (*d*=0.26) No significant difference in emotion recognition (Eyes Test) post-intervention (-) (*d*=0.19) No significant difference in everyday social skills post-intervention (-) (*d*=0) Statistically signification effect of group (intervention vs waitlist control) on improvements in emotion regulation (+) (*d*=0.54) and social reciprocity (+) (d=0.67)

Study	Quality Rating	Risk of Bias Rating	Effect of Intervention on Social Skills (+ indicates improvement and – indicates lack of improvement)
Lorenzo et al. (2013)	0.5	0.17	Observed increase in percentage of children who understood the intervention tasks (+); Observed marginal increase in teacher ratings of classroom behaviours (+)
Lorenzo et al (2016)	0.37	0.46	No clear primary outcome measure. Study reported greater overall improvements in the intervention group, compared with the non-immersive control (+)
Ravindran et al. (2019)	0.42	0.33	Observed increases in frequency of interactions and amount of eye contact after using the VR system (+)
Crowell et al. (2020)	0.46	0.33	No significant difference in social initiations using MR, compared with non-digital control (-) (d=0.06) Statistically significantly more social responses in control condition, compared with MR intervention (-) (d=0.14)
Mora-Guiard et al. (2017)	0.39	0.33	Observed increases in initiations, acts and responses while using the MR system, between sessions 1 and 3 (+)
Sayis et al. (2022)	0.41	0.38	No significant difference in social initiations using MR, compared with non-digital control (-) (d=0.20) No significant difference in social responses using MR, compared with non-digital control (-) (d=0.32) No significant difference in externalisations using MR, compared with non-digital control (-) (d=0.13)
Tsai et al (2020)	0.5	0.6	Observed improvements in understanding of social initiations between baseline and intervention for all participants. Sustained at maintenance for 2 of the 3 participants (+/-) Statistically significant increase in therapist ratings of role play performance at intervention and maintenance (+)
Pérez-Fuster et al. (2022)	0.65	0.71	Observed increases in number of eye-gaze responses to joint attention attempts measured by the ESCS and idiosyncratic measures. Half of the participants also improved on the responses to joint attention item from the ADOS-2 (+)
Chen et al. (2015)	0.5	0.43	Statistically significant mean improvement in emotion recognition at follow-up in comparison to baseline (+)
Chen et al. (2016)	0.5	0.43	Statistically significant mean improvement in emotion recognition at follow-up in comparison to baseline (+)
Frolli et al. (2022)	0.48	0.54	Statistically significant mean improvement in emotion recognition at follow-up in comparison to baseline on four of four domains used (+) Statistically significant effect of group on time taken to acquire skills on three of four domains used (+)
Wedyan et al. (2021)	0.38	0.38	Observed increases in correctly identified facial expressions after using the intervention (+)

Developing, Maintaining and Understanding Relationships

Three AR studies (Chen et al., 2015, 2016; Wedyan et al., 2021) and one VR study (Frolli et al., 2022) primarily targeted the development, maintenance and understanding of relationships and one VR study included some relational outcome measures (Ip et al., 2018). Chen et al. (2015 & 2016) and Wedyan et al. (2021) targeted emotion recognition which was measured according to participants' ability to correctly identify emotions using unvalidated assessment measures. Chen et al. (2015 & 2016) showed that participants improved between baseline and follow up and informal parent reports suggested that improvements corresponded with real world change in ability to identify emotions however, this was not formally measured. Wedyan et al. (2021) also showed improvements after children completed the intervention however, this was not statistically assessed. Caregiver reports indicated that they found the AR to be engaging, enjoyable and easy to use. Frolli et al. (2022) also targeted emotion recognition and compared the time taken to acquire skills in this area after use of the VR to a control group who partook in a similar but non-immersive training programme. Both groups improved in their emotion recognition ability, as measured using an idiosyncratic assessment method, and those in the VR group acquired 100% accuracy significantly faster than those in the control condition. No indication of maintenance of skills, generalisability or relevance to the young people themselves was specified. In contrast, Ip et al. (2018) found no evidence of improvements on this domain after use of VR, when compared with a waitlist control. They targeted the application of social skills to real life (e.g. ability to maintain relationships), measured using a domain of the Adaptive Behaviour Assessment System (ABAS-II; Harrison & Oakland, 2003), and emotion recognition, which was measured using the Eyes and Faces Tests (Baron-Cohen et al., 1997, 2001). The outcomes of these studies give some initial evidence that

XR may be effective for improving relational social communication, although small sample sizes and lack of statistical analysis in some studies make it difficult to draw firm conclusions. Furthermore, studies have primarily focused on emotion recognition, with other aspects of understanding relationships relatively neglected.

Related Skills

Two studies included outcomes which would theoretically benefit social skills but do not fall under the DSM-5 social communication criteria (APA, 2013), such as emotion regulation (Ip et al., 2018) and flexibility to changes during social situations (Herrero & Lorenzo, 2020). When compared with a waitlist control, the former showed an increase in emotion regulation ratings after use of VR, as measured by the PEP-3 (Schopler et al., 2005), and the latter showed improvements in flexibility to change across all participants in the intervention group, according to idiosyncratic measures.

Quality of Included Studies

Quality ratings ranged from 0.31 to 0.67 (where possible scores are between 0 and 1) with an average quality rating of 0.50. Risk of bias scores ranged from 0.17 to 0.83 with an average of 0.47. Although the adapted method of scoring makes it difficult to give a precise assessment of quality, comparisons with previous applications of this checklist suggests that the studies were generally of poor to fair quality (Hooper et al., 2008). Higher scores were typically given for clarity of reporting, such as describing the main aims, interventions and findings. The majority of studies took place in representative locations, and there was little evidence of non-compliance. Lower quality studies tended to have non-representative samples, unclear recruitment strategies and lack of reporting on attrition. Furthermore, several studies did not attempt to blind those

involved in rating outcomes. It was particularly notable that no studies gave justification for their selected sample size.

Further quality appraisal using the SCED Scale, indicated that the majority of small-N studies were truly experimental in design (8 of 11) and conducted sufficient sampling at baseline and treatment phases; some of these studies also performed statistical analysis. However, a lack of precise and repeatable outcome measures was a significant weakness across small-N studies. In particular, outcomes tended to be rated by one, non-independent assessor and where there were multiple assessors, interrater reliability was not calculated. No attempt was made to show generalisation across settings or therapists.

Discussion

This systematic review identified 21 studies that investigated the potential of XR interventions to enhance the social skills of autistic children. To date, the majority of interventions have targeted social-emotional reciprocity, with relatively little attention given to the non-verbal and relationship aspects of social-communication. The overall quality of the research is relatively low, perhaps reflecting the infancy of the research area. The majority of studies have small sample sizes and a number are not truly experimental, making it difficult to draw firm conclusions about the efficacy of XR, particularly in comparison to current, less costly social skills interventions. Significant heterogeneity means it is not possible to determine the ‘active ingredient’ of XR interventions. Furthermore, only limited attempts have been made as yet to determine generalisability and there is limited exploration of whether the statistically significant changes observed by many of the studies were also clinically significant i.e. led to

improvements in everyday social or occupational skills, or other important areas of functioning.

Limitations of the Current Literature

Given the relative novelty of this research, it is important to consider the limitations of the current evidence base, to be improved upon in future research. In particular, current evidence is limited in its ethical considerations, theoretical grounding, robustness of study design and sample representativeness. It is also unclear as yet, how easily and cost-effectively XR interventions could be scaled to clinical practice.

Ethical Considerations

Social skills interventions are primarily important because of the negative secondary effects of social difficulties and so quality of life has been an important outcome measure across the literature (e.g. Baghdadli et al., 2013; Mitchel et al., 2010). However, none of the studies included in this review measured quality of life, nor did they consider the importance of the intervention target to the participants themselves. Krasny et al. (2003) emphasise that interventions are most efficacious when children understand the relevance of the social skill and work towards individualised goals. Future interventions could capitalise on the customisability of XR to tailor interventions to individual needs. It is perhaps telling that the majority of interventions have targeted social reciprocity which, by definition, impacts people around autistic people. There are few attempts to directly target development and maintenance of relationships, despite this being what some autistic people have expressed as most important (Cresswell et al., 2019). Some studies did use participatory research methods, i.e. the incorporation of the views of autistic people into the design and implementation of the research (Cornwall &

Jewkes, 1995), although the extent of involvement is somewhat limited. Increased meaningful involvement from autistic people within this field of research would help to ensure outputs are relevant and beneficial to the people for whom they are designed (Fletcher-Watson et al., 2019).

Future research should also make a more comprehensive attempt to measure adverse events, particularly given that one study which did comment on negative outcomes, noted problems with restlessness and discomfort, while another reported using positive reinforcement to keep children engaged. Moreover, commercially available VR headsets typically include safety notices regarding their use in children, emphasising the need for robust assessment of potential harm. Future studies could improve this through quantitative measurement of participant experience, such as whether the tool was straightforward to use and physically and sensorially comfortable, and analysis of the relationship to outcomes i.e. whether more positive experiences are associated with greater improvements. As well as the potential adverse effects of XR, future studies should consider the wider impact of teaching children to implement neurotypical social skills. Many autistic people already invest significant time and energy in monitoring and modifying their behaviour in order to align with social norms, known as ‘social camouflaging’. Evidence suggests that this hiding of one’s true self can be mentally and physically draining, and requires excessive concentration, self-control and discomfort (Hull et al., 2017; Mandy, 2019). Further exploration is needed of whether social skills training simply teaches children to mask their true selves, and if so, serious attention should be given to the potential deleterious impact on wellbeing. Perhaps greater research focus could instead be given to how settings, such as schools, could adapt to become more accommodating for autistic people, rather than placing all emphasis on changing autistic children to manage in a neurotypical world.

Theoretical Grounding

Despite the number of theoretical explanations for social differences in autism, the studies reviewed in this research give relatively little theoretical justification to the design of their interventions. As a result, XR has been applied in a number of different ways, often with no clear rationale for why design decisions were made. Greater attention to theory when designing interventions might further improve efficacy. This has proved beneficial for non-XR studies, such as the *Mind Reading* intervention which draws on the Empathising-Systemising Theory by exploiting participants' relative strength in systematising when teaching emotion recognition (Golan & Baron-Cohen, 2006). Limited consideration of the mechanism by which change might occur may also account for lack of specificity of some of the research. In particular, some studies measured a number of outcomes with no clear indication of how these related to the purpose or design of the intervention. Theoretical understandings of the mechanism of change could also inform intervention duration, which varied significantly across the studies. While it seems theoretically unlikely that clinically significant changes could be made in the time of the shortest intervention (15 minutes), strong theoretical justification should be given before subjecting children to high numbers of intervention sessions.

Study Design

Quality appraisal highlighted a number of methodological limitations of the current research. For example, the majority of studies did not attempt to blind the individuals who coded behavioural outcomes and in fact, behaviours were often rated by researchers or parents who knew the purpose of the study, both of whom are likely to have been highly invested in positive outcomes. In particular, parents of autistic

children have been shown to report positive effects when told their child is receiving treatment, even in the absence of an intervention (Jones et al., 2017). It is also possible that improvements occurred as a result of repetition of the outcome measures themselves. For example, a number of studies used questions based on Social Stories™ to measure outcomes, and as a social skills intervention in itself, this may have contributed to improvements. Similarly, when outcomes were measured by *in vivo* performance, improvements may have been due to practice effects, rather than social skill development. Greater use of validated measures of social skills, applied by raters who are blinded to the study purpose and intervention allocation, would improve some of these issues. Future research should also ensure that potential confounders, such as whether participants are receiving any other therapies, are fully considered.

Appropriately for the stage of research, the majority of studies were small-N. However, in larger trials, no details were given about the determination of sample size, and so it may be that those which did not find an effect were underpowered. In one study, a significant proportion of the sample were excluded from analysis based on age, however, the purpose of this is not explained, diminishing the validity of their results. To determine effectiveness, more large-scale trials are required: these should be sufficiently powered and ideally pre-registered, to reduce the likelihood of data dredging and subsequent false positives. More experimental, controlled studies would also make it easier to determine, not only whether XR interventions are effective, but whether they offer any real improvement to social skills, over and above current methods.

Representativeness of Sample

Current research is limited in the extent to which participants are representative of the autistic population as a whole. Around 25% of autistic people are female (Loomes et al., 2017) but only six of the included studies met this threshold. Similarly, up to 50% of the autistic population are estimated to have an Intellectual Disability (ID; Charman et al., 2011; Loomes et al., 2017) yet a number of the included studies excluded participants with an ID or stated a mean IQ within the average range. Limited justification is given for this and it compounds evidence that people with ID are consistently excluded from autism research (G. Russell et al., 2019). Future research should seek to recruit participants who truly reflect the autistic population as a whole, or provide strong justification for exclusion.

Limitations of the Review

The findings of this review should be viewed in the context of its limitations. Firstly, an effect size could only be given for a limited number of studies making it impossible to compare the efficacy of different interventions using meta-analysis. Instead, this study used a systematic but non-analytic approach to synthesis. Vote counting and summary plots were avoided to reduce the potential misrepresentation of efficacy, given the number of low quality studies. This could perhaps have been improved by only including studies which meet a particular quality threshold. However, this was not deemed feasible for this review given the small number of studies. As the research develops and more experimental, controlled studies are carried out, more systematic methods of synthesis will be possible. Furthermore, publication bias will have significantly impacted the number of studies showing positive effects. Grey literature was not included, to ensure included studies were of peer-review quality, but

this may have increased the positive results bias. When determining studies to be included, there were some borderline cases, where insufficient descriptions of study interventions made it difficult to establish whether the XR was immersive. This was resolved through discussion amongst researchers but, could have been improved by contacting study authors for more information.

The conclusions that can be drawn from this review are also constrained by the limited amount of data, both in terms of number of studies and overall sample size. The maximum age of participants across studies was 16 and so it is unclear whether XR interventions could be effective for helping older teenagers and young adults to enhance their social skills. Additionally, only VR studies which altered one's perception of reality were included and so the efficacy of immersive VR could not be compared to that of VLEs, which may be more cheaply and easily implemented in clinical practice.

Conclusion

The potential to digitally alter how one perceives the world offers exciting new possibilities in developing effective and engaging social skills interventions for autistic children. However, enthusiasm for XR interventions should be viewed with caution. While there is some indication that interventions are feasible to implement with autistic children, limited evidence exists for their effectiveness in bringing about meaningful, longstanding improvements in everyday functioning. Furthermore, no assessment has been made of the potential emotional cost of autistic children implementing behaviours which do not come naturally to them. This review demonstrates the need for theoretically grounded interventions, designed with the interests of autistic people at the forefront. Controlled trials and larger sample sizes, as well as other improvements to study design, are required to draw firm conclusions about the efficacy of XR

interventions and their generalisability, before potentially high cost scaling to routine clinical services is considered.

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Service Improvement Project

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Title: Improving Patient Activation in a Community Neurorehabilitation Setting: A Mixed Methods Investigation

Word Count: 5205

Date: October 2022

Internal Supervisor: Dr Nigel King (nigel.king@hmc.ox.ac.uk)

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Proposed Journal and Rationale: The British Journal of Health Psychology. This was chosen due to its focus on aspects of health related behaviour change and managing chronic illness (submission guidelines included in Appendix E).

Abstract

‘Patient activation’, defined as a patient’s knowledge, skills and confidence in managing their health condition, is essential for collaborative neurorehabilitation. It is therefore in the interest of neurorehabilitation services to monitor and seek to improve this. The process of increasing patient activation at a Community Neurorehabilitation Service in Buckinghamshire (CNRS) was hereby explored. This research used a mixed methods design to investigate whether patient activation increases throughout intervention with CNRS and if so, what factors contribute to this. Quantitative analysis showed that activation did increase significantly and this was not impacted by demographic factors. Qualitative interviews with eight former patients and four therapists suggested that the nature of the therapeutic relationship and wider social support may have been important in contributing to the change. Negative past experiences of services and cognitive difficulties seemed to be associated with lower activation. As a result, CNRS embedded a number of service improvements including increased opportunities for peer support and greater consideration of the patient’s wider health journey and emotional difficulties. Future research should seek to measure the impact of these improvements, as well as investigating the potential causal role of the factors identified in this qualitative research.

Key Words: Patient Activation, Neurological Conditions**,** Neurorehabilitation, Self-Management, Behaviour Change

Improving Patient Activation in a Community Neurorehabilitation Setting: a Mixed Methods Investigation

People living with neurological conditions (disorders of the brain, spinal cord or nerves), such as Multiple Sclerosis (MS), Parkinson's Disease (PD), and Stroke, can experience a range of biopsychosocial problems for which they might seek support from a Community Neurorehabilitation Service. Ideally, this rehabilitation is '*an active, time-limited collaboration… to produce sustained reductions in the impact of disease and disability on daily life'* (Royal College Of Physicians, 2010). The need for collaboration necessitates that the patient's role within neurorehabilitation is paramount; it requires a willingness to engage, as well as individual action. Community neurorehabilitation services which understand how best to support patients with this are likely to achieve the best outcomes.

A person's willingness to actively engage with their clinical care can be understood in terms of 'patient activation', whereby the more 'activated' a patient is, the more informed, skilled and confident they are in managing their healthcare (Hibbard et al., 2005). Increased activation is associated with more positive health behaviours and effective long-term condition management (Hibbard & Gilburt, 2014). Levels of activation are typically assessed using the Patient Activation Measure (PAM; Hibbard et al., 2004) which classifies individuals as one of four levels of activation (Table 1). This is predictive of patient outcomes across a range of long-term conditions, including those which are neurological (e.g. Skolasky et al., 2008). Patient activation encompasses the broad range of psychological processes that might impact a person's capacity to enact positive health behaviours, including internal locus of control (Wallston et al., 1994); high self-efficacy (an individual's belief in their capacity to execute behaviours to reach

a particular goal; Bandura, 1977) and readiness for change (Prochaska & DiClemente, 1983).

Table 1

The Four Levels of 'Patient Activation' and Corresponding Scores on the Patient Activation Measure (PAM; Hibbard et al., 2005).

Activation Level	Activation Score	Description
Level 1	47.0 or lower	Individuals tend to be passive and feel overwhelmed by managing their own health. They may not understand their role in the care process.
Level 2	47.1 to 55.1	Individuals may lack the knowledge and confidence to manage their health.
Level 3	55.2 to 67.0	Individuals appear to be taking action but may still lack the confidence and skill to support their behaviours.
Level 4	67.1 or above	Individuals have adopted many of the behaviours needed to support their health but may not be able to maintain them in the face of life stressors.

Patient activation is currently used at the Community Neurorehabilitation Service in Buckinghamshire (CNRS) to understand how patients engage with intervention. The service offers neurorehabilitation to adults with a range of neurological conditions through support from Physiotherapists, Occupational Therapists, Speech and Language Therapists and Clinical Psychologists. The aim of the service is to support individuals to regain independence, maintain participation and manage their condition, and so 'activation' is an important outcome target. This is routinely measured before and after service input using the PAM.

Clinical observations suggest that those patients who remain at low levels of activation throughout treatment, tend to be offered longer, less effective interventions, or are later re-referred. Others become considerably more activated during their intervention, in line with service goals. It is unclear what differentiates these two groups

and what more could be done to support those who remain at a low levels. Previous research has tended to focus on factors leading to initial low activation, many of which are fixed such as degree of disability (Fritz et al., 2019), social support (Blakemore et al., 2016) and education levels (McCabe et al., 2018). Others have focused on improving one specific component of patient activation, such as self-efficacy (Kennedy et al., 2007). Little research has investigated the impact of factors specific to neurological conditions, such as cognition and neurodegeneration, although it has been shown that progressive MS is associated with lower activation than relapse-remitting (Goodworth et al., 2016). The evidence-base for how to support people at lower levels of activation is particularly limited. Motivational Interviewing (Miller, 1983) is commonly used at CNRS, but its efficacy in improving activation has not been evaluated. Lack of best practice guidance presents a challenge for NHS services which are advised to support people to manage their own health (NHS England, 2014; The Stroke Association, 2021).

This research sought to better understand the activation process at CNRS in order to identify avenues for intervention for patients at lower levels of activation. It was thought that this would improve outcomes for patients, as well as service efficiency. The following questions were addressed:

1. Does neurorehabilitation at CNRS help to increase patient activation?

   It was hypothesised that patient activation would increase after accessing a programme of neurorehabilitation.
2. Do demographic factors contribute to improvements in activation at CNRS?

   An exploratory analysis was used to understand whether sex, diagnosis, ethnicity, family involvement, age and waiting time affected the extent to which activation increased during neurorehabilitation.

3. What do patients experience as important in helping them to, or preventing them from, becoming activated during neurorehabilitation at CNRS?

   This was explored qualitatively through patient interviews, in line with the current stage of the evidence.

4. What did neurorehabilitation therapists experience as important in helping specific patients to become activated during neurorehabilitation?

   This was explored qualitatively through therapist interviews, in line with the current stage of the evidence.

A number of service improvements based on the findings of this research were then generated and actioned, in collaboration with the CNRS team.

Method

Design

This study used a mixed methods design. The quantitative component used a retrospective longitudinal design and within group analyses, and the qualitative element used semi-structured interviews interpreted using the principles of Thematic Analysis (Braun & Clarke, 2006). The study was preregistered on the Open Science Framework (DOI: 10.17605/OSF.IO/FKTGH)

Participants

Data from 77 patients at CNRS was collected between 2018 and 2021 (Table 2). Patients were over 18, had a GP within Buckinghamshire and a diagnosed neurological condition or previous stroke. To be included for analysis, patients had completed the PAM before and after neurorehabilitation.

From this data, consecutive sampling was used to recruit two groups of patients for interviewing (Table 3), according to most recent discharge date. We aimed to recruit

five patients who increased from a low to high activation level (i.e. from a level one or two to a level three or four) and five patients who remained at a low patient activation level (i.e. a level one or level two). However, it was only possible to recruit three patients from the latter group due to the majority declining participation, most commonly due to anxiety and cognitive decline. One patient was supported by his wife and the other seven were interviewed alone.

Table 2

Demographics of the Patient Data Used for Analysis

	Number (n)	Percentage
Age (years)		
30-39	4	5.19%
40-49	6	7.79%
50-59	12	15.58%
60-69	16	20.78%
70-79	28	36.36%
80-89	11	14.29%
Gender		
Female	36	46.75%
Male	41	53.25%
Diagnosis		
Stroke	22	28.57%
Parkinson's Disease (PD)	26	33.77%
Multiple Sclerosis (MS)	11	14.29%
Other (Spinal cord injury; Epilepsy; Huntington's Disease; Other disorders of movement and/or neurodegeneration)	18	23.38%

Table 3

Interview Participant Demographics (Patients)

Participant	Sex	Age Group	Diagnosis	PAM Level Pre-rehab	PAM Level Post-rehab
P1	M	70-79	Stroke	2	3
P2	F	70-79	PD	2	4
P3	F	80-89	MS	2	3
P4	F	70-79	MSA	2	4
P5	M	70-79	PD	2	4
P6	M	70-79	PD	1	1
P7	M	70-79	Stroke	2	2
P8	F	50-59	Stroke	2	2

Four therapists (Table 4) were also recruited for interviewing using convenience sampling, based on an expressed interest in participating. The only inclusion criteria was that they were able to reflect on a patient who they perceived to have increased in activation during rehabilitation. All participants recruited for this research provided informed consent under NHS ethics (Ethics Ref: 21/WS/0054, Appendix F).

Table 4

Interview Participant Demographics (Therapists)

Participant	**Profession**	**Intervention Discussed**
T1	Occupational Therapist	Regaining functional arm use after stroke through rehabilitation and adjustments
T2	Psychologist	Overcoming shame of hereditary motor condition through 1:1 therapy
T3	Physiotherapist	Improving strength and balance after stroke through rehabilitation
T4	Occupational Therapist	Supporting emotional processing and decision making within a triage assessment

Measures

Demographic Information

A range of demographic data from patient records was used for analysis: age, sex, ethnicity and primary diagnosis. Whether a family member or friend was present at

the triage appointment was used as an indictor of social support and length of time between the triage assessment and first appointment was used to approximate wait for therapy.

Patient Activation Measure (PAM)

The primary measure used in this research was the Patient Activation Measure (PAM) Short Form (Hibbard et al., 2005), a self-report questionnaire on which patients rate their agreement with 13 statements about their beliefs, confidence in management of health related tasks and self-assessed knowledge. Agreement is scored on a four-point Likert scale, with an additional option to note if the question is not applicable. Total scores are transformed to a 0-100 scale, whereby higher scores indicate increased activation. Scores are then used to determine level of activation, as detailed in Table 1. The PAM is valid and reliable across a range of health problems, including neurological conditions (Packer et al., 2015). The measure was shown to have strong internal consistency in this population (Cronbach's Alpha of 0.87). An increase of four points on the PAM is considered a meaningful increase, corresponding with increased self-management behaviours (Turner et al., 2015).

Quantitative Analysis

A paired samples t-test was used to compare PAM scores before and after contact with the service. Power analysis indicated that the available dataset would be sufficiently powered, assuming a moderate effect size ($d = 0.50$). We used a hierarchical multiple linear regression to determine the extent to which a number of factors influenced the degree of increase. Power analysis indicated that this would be sufficiently powered with a moderate to large effect size ($f^2 = 0.25$). Additional exploratory analyses were also conducted.

Qualitative Component

Data Collection

The first author conducted one-to-one semi-structured interviews with patients and therapists over video conferencing or the telephone. The concept of patient activation was explained to patients and they were encouraged to reflect on their rehabilitation with CNRS, with this in mind. Therapists, who already had an understanding of patient activation, were asked to reflect on a specific patient who they perceived to have become activated and what they considered to have helped with this. This methodology was chosen to generate specific ideas about what contributes to activation increases, rather than general therapist techniques or factors. Interview schedules (Appendix G) were used as a guide but the interviewer steered the conversation according to responses. Interviews were recorded and transcribed by the first author and a member of the clinical team. Collection and analysis was a parallel process: data was coded after each interview and patterns of interest were used to shape subsequent data collection. The interviewer aimed to conduct interviews empathetically, showing interest and non-judgement, while observant of disinterest, misunderstanding or distress (Braun & Clarke, 2013).

Thematic Analysis

The first author familiarised themselves with the data and developed a range of codes based on the theory of patient activation, as well as data-driven codes which became more focused over time. Emergent themes and subthemes were identified through the sorting of codes into categories and identifying the relationships between them. As latent themes were refined, a thematic map was produced to show the relationships between them. These were defined, named and given an analytic narrative

supplemented by specific quotations from the data. The second author quality checked this process through review of six interviews, ensuring codes and themes were consistent with the data. Braun & Clarke's (2006) 15-point checklist was also used as a quality control measure.

Results

Quantitative Analysis

Demographics

Following removal of two significant outliers, the data of 75 former patients aged 34 to 86 (M = 66.48, sd = 12.75) were included for analysis. The sample was 53.3% male, 90.7% White British, and most commonly diagnosed with Stroke (29.3%) or Parkinson's Disease (34.7%). Just under half attended triage with a family member or friend (44%) and waiting time varied significantly (M = 60.52 days, sd = 55.90). On average, PAM scores of participants increased by 5.09 points (sd = 10.22), with initial scores averaging 56.68 (sd = 11.98) and post-intervention scores averaging 61.77 (sd = 13.34).

Analysis

A paired samples t-test[2] indicated that the increase in PAM score was significant at the 5% level, $t(74) = 4.31$, $p < 0.01$, with a medium effect ($d = 0.50$). After neurorehabilitation, 47 patients (62.7%) were at a high level of activation, compared with 32 pre-treatment (42.7%), and 33 patients (44%) made a meaningful increase in activation (i.e. an improvement of 4 points or more).

[2] Assumptions of normality met.

The extent to which demographic factors may have contributed to this change was investigated using a hierarchical multiple linear regression[3] to predict post-intervention activation scores from sex, diagnosis, ethnicity, social support, age and wait for therapy, with initial PAM score as a covariate. Diagnosis and ethnicity were coded as dummy variables where stroke (as the non-degenerative condition) and White-British (as the majority) were used as baselines. A significant regression equation was found for the final model ($F(9, 65) = 8.52$, $p < 0.01$, $f^2 = 1.18$) with an R^2 of 0.54 (Table 5), however, the change statistic showed that adding demographic factors did not significantly improve the model ($F(8, 65) = 1.40$, $p = 0.21$).

Table 5.

Regression Analysis Showing the Relationship Between Pre- and Post-Intervention Scores (Model 1) and the Impact of Demographic Variable (Model 2).

	Model 1			**Model 2**		
	b	**SE (*b*)**	***t***	***b***	**SE (*b*)**	***t***
Constant	18.79	5.55	3.39**	27.96	10.89	2.57**
Pre-Intervention Score	0.76	0.10	7.92**	0.65	0.11	5.98**
Age	-	-	-	-0.03	0.10	-0.26
Sex	-	-	-	4.55	2.51	1.82
Parkinson's Diagnosis	-	-	-	1.75	2.88	0.61
MS Diagnosis	-	-	-	0.88	3.87	0.23
Other Diagnosis	-	-	-	2.75	2.34	0.82
Waiting Time for Therapy	-	-	-	-0.04	0.02	-1.83
Social Support	-	-	-	-4.63	2.40	-1.93
Ethnicity	-	-	-	-1.18	4.10	-0.29

*p<.05, **p<.01.

[3] Assumptions of normality, linearity, homoscedasticity, absence of multicollinearity and non-autocorrelation met.

Explorative Analysis

To investigate how demographic factors might impact in other ways, further exploratory analysis was undertaken. A multiple linear regression[4] was used to predict *initial* activation scores based on sex, diagnosis, ethnicity, social support, age and wait for therapy. A significant regression equation was found ($F(6, 68) = 3.19$, $p = 0.01$, $f^2 = 0.28$), with an R^2 of 0.22. Of these factors, only age significantly contributed to the model ($t(68) = 2.4$, $p = 0.02$), with younger age predicting higher initial score.

Qualitative Analysis

Twelve interviews with therapists and patients were conducted for a mean duration of 44 minutes, over a three month period.

Themes

Five patient themes (Setbacks and Frustrations; Barriers to Change; Positive View of Support; Wider Social Support; Signs of Activation) and three therapist themes (Approaching Difficulties; Patient-led Collaboration; Finding Direction and Confidence)[5] were identified and are described in full in Appendix H. These themes were organised in terms of their relation to activation levels in a thematic map (Figure 1) and analytic narrative.

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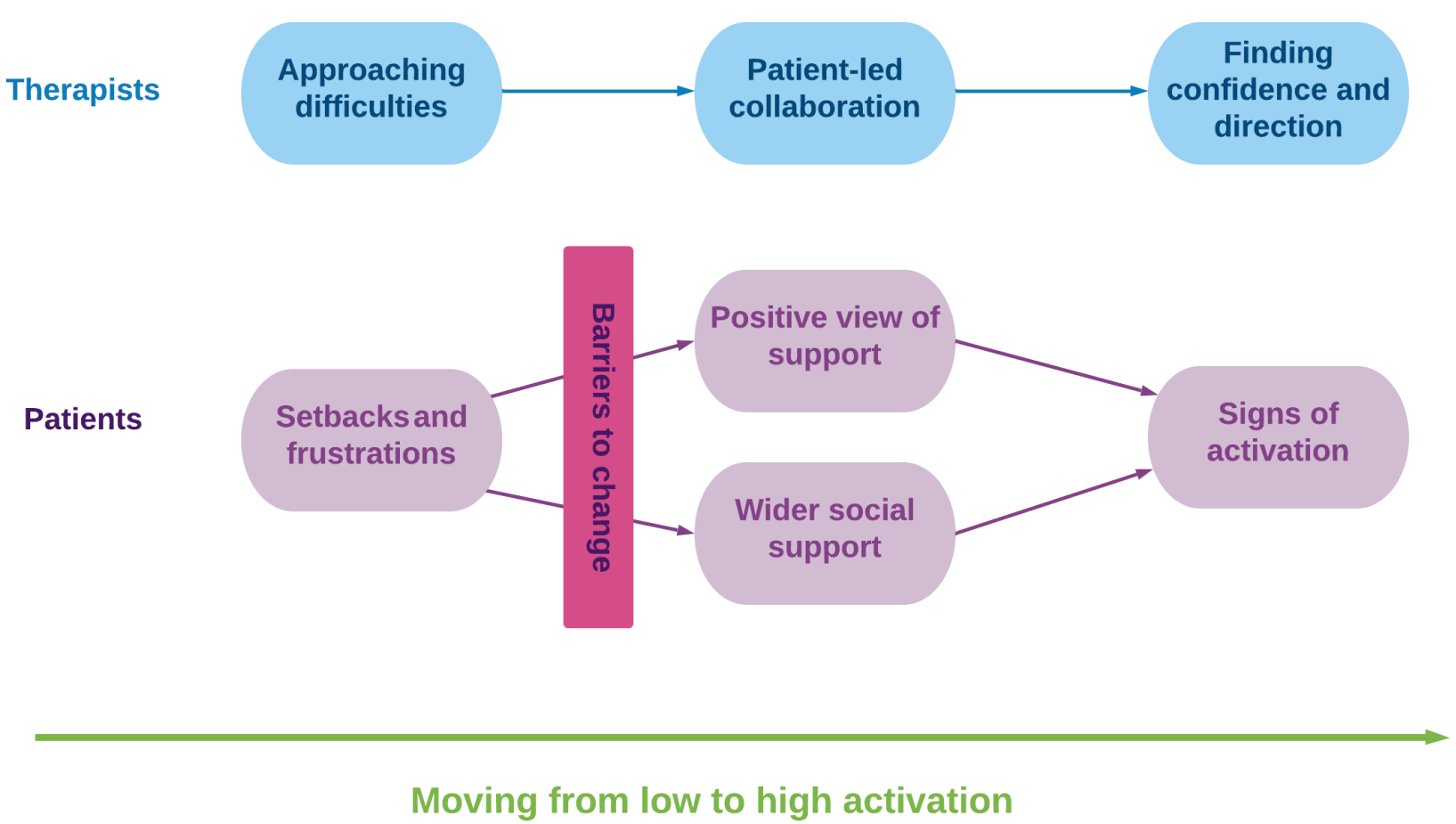
[4] Assumptions of normality, linearity, homoscedasticity, absence of multicollinearity and non-autocorrelation met.

[5] Given the limited number of participants, these are likely to represent some of the themes, rather than an exhaustive finding.

Figure 1.

A Thematic Map Showing Themes Which Were Identified and Then Organised in Relation to Activation Levels



Low Activation. Two themes reflected therapist and patient perceptions of low activation. All patients described a number of 'Setbacks and Frustrations' before, during and after intervention with CNRS. Many had negative experiences of services, including difficulties with diagnosis (P4 – 'we knew… but couldn't get a diagnosis') and post-diagnostic support (P1 – 'nobody checks on you'). For some, this negatively impacted their expectations of therapy (P8 – 'I was of the view that I'd been down that road before with physio') and for others, it prevented them accessing therapy at all (P7 – 'that's something I've refused myself because it's been a waste of time… occupational therapy'). Many reported emotional difficulties including guilt, sadness, embarrassment, and anxiety, which may have contributed to initial low activation. Therapists described how they supported patients to address and move forward with emotional difficulties and hidden symptoms such as fatigue ('Approaching Difficulties'). One therapist spoke about how this required going beyond the role of their specific profession (T4 – 'What I allowed them to do was just process all of their thoughts'). Another emphasised the importance of the time spent listening, as otherwise it 'makes it really really difficult to engage in the exercises' (T2).

Barriers to Change. A number of subthemes identified in the low activation group related to 'Barriers to Change'. Patients seemed to relate to their condition in black and white terms, seeing things as entirely hopeless (P8 – 'It's basically too late') or ignoring the health difficulty altogether (P6 – 'I try to ignore the fact I've got it'). There was also a sense of isolation in this group. Participants spoke of depending on themselves (P8 – 'That's what's got me through… nobody else… me') and expressed frustration that services didn't seem interested in them (P7- 'they ought to be in touch with you a bit more'). Furthermore, all of the low activation group reported cognitive changes which may have inhibited activation.

Increasing Activation. Three themes reflected therapist and patient perceptions of increasing activation. Therapists stressed the importance of 'Patient-led Collaboration', whereby the patient took a lead in the therapy (T2 - 'it was very very guided by her') and the therapist needed to be flexible and realistic to allow for this (T1 - 'If that works for you… then continue'). Patients whose activation increased generally had a 'Positive View of Support' received from CNRS. They descried their therapists as 'confidence building', 'encouraging' and 'understanding', and found it helpful to see evidence of their progress. Again, support was viewed as collaborative, with patients aware of their own role (P2 - 'I try to prepare'), the role of the therapist (P1 - 'doing it together'), family members (P4 - 'a sister-in-law who came with me') and the rest of the team (P3 - 'everyone there was wonderful'). 'Wider Social Support' was a theme across all patients. One spoke of how his friends helped him to make necessary adaptations, while another emphasised the support of her walking group. For many, the most valuable support came from individuals with the same condition (P2 - 'We've got a group that meet together; P3 - 'We share our problems and things') and indicated that it was easier to think about acceptance and adjustment with those suffering from the same difficulties.

High Activation. Two themes reflected activation increases. 'Signs of Activation' in the group who increased included self-efficacy, using language of acceptance and being comfortable to make adjustments and adaptations. One indicator of activation which particularly related to CNRS was the ability to see progress and purpose (P1 – 'I still do the exercises… because it… keeps my body supple') and patients were able to speak matter-of-factly, if not positively, about the future (P3 – 'things won't be as bad as I think they will'). Likewise, therapists spoke about patients 'Finding Direction and Confidence', moving from a place of initial stuckness to

independence. One therapist described how she supported this by giving the patient responsibility for the therapy ending (T2 – 'It gave her a month to see that she could manage this by herself'). Others spoke about how they connected the therapy to its wider purpose (T3 – 'I would explain why') and the individual's values (T4 – 'Are you moving closer to your values or do you need to make a choice?').

Service Improvements

The findings of this research were presented to the CNRS team and potential changes to better support patients in becoming activated were discussed. The team were reassured that much of what they do aligns with what patients reported as helpful. For example, ensuring time is spent at initial assessment hearing someone's journey and the impact of living with their condition. The physiotherapy team were pleased to hear that their creative use of outcome measures was reported as beneficial by some patients. It was useful to the team to know that activation generally increases at CNRS and they put this down, at least in part, to the team's confidence in using motivational interviewing, as well as their focus on listening and working amongst professionals. The team expressed surprise that there was not a specific group of patients who particularly struggled to become activated, and that wait for therapy did not seem to impact patients' capacity for improvement.

A number of potential changes were explored. The team wondered whether more could be done to ask about patients' prior experiences of therapy and consider how this might impact neurorehabilitation. They also wondered how people with lived experience could contribute to supporting with activation at CNRS. Some members of the team thought that this would involve increased signposting or joint working with charitable organisations, while others wondered whether 'expert patients' could

contribute to a group targeted at low activated patients to help them to prepare for therapy. The team also reflected on the importance of deciding when someone is not ready for therapy at the point of triage. Confidence in making this decision and supporting someone psychologically at this point differed across the team. These ideas and reflections were then taken back to the senior team who agreed upon the following plans for service improvement:

1. **Changes to the triage process.** Given the importance of the initial assessment in hearing the patient's journey and making decisions about the timing of therapy, CNRS decided to review this at their away day later in the year. This review would involve identifying the skills required for successful triage and the associated training needs of each member of the team.
2. **Asking about previous experiences of therapy.** Many of the team already ask about past experiences of therapy but this is not systematic and the potential impact on therapy with CNRS is not always explored. The team therefore added a prompt to their triage crib sheet to ensure that this is routinely discussed.
3. **Capitalising on the benefits of mutual support groups.** Although the service often recommends charity support groups, this had not previously been considered as a specific waitlist intervention for those at a low level of activation. The service therefore planned to create a signposting document to be given to those identified as 'low activated' at triage.
4. **Developing specific support for low activation patients.** This was identified as a longer term goal for the leadership team to discuss, incorporating the findings of this research. Ideas included a group focused on psychoeducation, with topics such as managing fatigue and cognitive changes, and 'expert patient' involvement.

Discussion

This research aimed to understand the factors contributing to increases in patient activation at CNRS, to inform support for those at low levels of activation. In line with initial hypotheses, the current neurorehabilitation process at CNRS was shown to be associated with significant increases in patient activation, with almost half of patients having made meaningful improvements. This may be a consequence of rehabilitation in and of itself, or the strategies the team implement to bring about change, such as Motivational Interviewing (Miller, 1983) and Acceptance and Commitment based approaches (Hayes, 2004). From the demographic data recorded by the service, it was not possible to identify any static factors which influenced the extent to which activation increased throughout therapy, and somewhat surprisingly, time spent waiting for treatment neither affected initial activation nor activation changes. Social support did not influence outcomes, in contrast to previous research showing its role in behaviour change (Levy et al., 2009). However, this may be due to lack of a robust measure, as social support had to be estimated based on the presence of a family member or friend at assessment. Likewise, a person's diagnosis did not impact activation changes, despite some neurological conditions being significantly more pervasive and quickly degenerating than others. In line with previous findings (Blakemore et al., 2016), older people were less likely to be activated prior to intervention, however, age did not hinder capacity for improvements in activation. Although not all possible demographic factors were explored, the limited impact of these variables suggests that intrapersonal and relational factors may be more important in increasing activation.

Themes identified in patient and therapist interviews provide further evidence of the importance of these changeable factors. The support of CNRS therapists was suggested to be important in helping patients to approach the difficulties associated with their condition, in order for them to take an active role in self-management. Addressing emotional difficulties may have been particularly important, given the strong association between mood and activation (Blakemore et al., 2016; Magnezi et al., 2014). To do this, the CNRS team take a holistic approach to neurorehabilitation with embedded psychological approaches (e.g. use of values, Hayes, 2004) and team working across professions. The majority of this work takes place at the point of triage and so the team decided to review this process to ensure all clinicians feel equipped to provide the emotional support that may be necessary at the earlier stages of rehabilitation.

The importance of having a sense of purpose was apparent across interviews. Therapists reported helping their patients to find direction and confidence by demonstrating progress and patients expressed that this was an important part of them embedding exercises and adaptations in their daily lives. This involved effective use of outcome measures, explaining the purpose of specific interventions and ensuring the intervention level was likely to achieve success. This is in line with theories of behaviour change which note the importance of a person seeing their own role in the change process and a belief in their capacity to enact improvements (Bandura, 1977; Wallston et al., 1994).

Although quantitative analysis did not demonstrate a role of social support at triage in activation, social support more broadly was a common theme across interviews. Previous research has highlighted the role of helping relationships in initial

activation (Blakemore et al., 2016) but the potential for new social support to increase activation, particularly that of others with the same condition, has not been explored in the literature. Advice from others was invaluable to some patients in moving from being overwhelmed, to finding acceptance and a willingness to make adaptations. This advice tended to be accessed via charitable organisations such as Parkinson's UK but raises the question of what NHS services could do more of to embed mutual support. Many effective self-management programmes are peer-led (Foster et al., 2007; Kennedy et al., 2007) but this is not routinely embedded within service provision at CNRS, as may be the case in other neurorehabilitation services. Based on this finding, the service will now ensure that all patients identified as being low activated are encouraged to access local support groups while waiting for neurorehabilitation. The service also plans to consider how 'expert patients' might contribute to a specific intervention for low activated patients.

Patient interviews highlighted the setbacks and frustrations that may have contributed to initial low activation, as well as the barriers to change. In line with previous research, emotional difficulties seemed to be related to low activation (Blakemore et al., 2016; Magnezi et al., 2014) and negative past experiences sometimes lowered expectations for rehabilitation. This is likely have negatively impacted activation, as believing in the possibility of improvement is an important part of behaviour change (Fishbein & Ajzen, 1975). Reported barriers to change included black and white thinking and a sense of isolation. Specifically, some patients felt CNRS could have done more to listen to them and provide longer-term support. While the service is only commissioned to provide time-limited support, it was hoped that a review of the assessment process could improve the service's ability to help patients feel heard and explore relevant past experiences in detail. It was also notable that all patients in the low

activation group reported cognitive impairment and this also contributed to recruitment difficulties in this population. To our knowledge, the relationship between cognitive difficulties and patient activation is not yet understood and so may be an important avenue for future research.

Limitations

The findings of this research should be considered in the context of its limitations. Retrospective analysis limited the demographic factors which could be investigated and so the potential impact of socio-economic status, education level, and symptom severity could not be identified. There was also not a direct measure of social support available and so this had to be estimated by the presence of a family or friend at assessment. Lack of diversity in ethnicity may also mean the analysis was not sensitive to its potential impact on rehabilitation at CNRS. Given the heterogeneity in neurological conditions, it may be that nuances of specific groups were lost when considered as a collective in this research. Finally, consecutive sampling meant that the interview groups may not have fairly represented the wider population (for example, interview participants were mostly older than the dataset average), limiting the scope of the themes that could be generated.

Conclusion

This research demonstrated that neurorehabilitation can be effective in increasing patient activation in those with neurological conditions, and the holistic, psychologically informed approach of clinicians at CNRS may have contributed to this. A number of fixed factors were shown to have little to no impact on the change process, and so the most important findings of this research were the themes identified from qualitative interviews. Avenues for service improvements identified by the CNRS team

in light of these themes included: embedding peer-support, increasing opportunities for support with emotional difficulties and considering the patient's wider health journey. Future research should seek to understand the potential causal role of these factors and the service should aim to measure the impact these changes.

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Theoretically Driven Research Project

Candidate: Rebecca Roberts (rebecca.roberts@hmc.ox.ac.uk)

Title: Understanding Post-Stroke Cognitive Impairment and its Sequelae Using the OCS-Plus

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Proposed Journal and Rationale: Clinical Rehabilitation. This was chosen because of its clinical and multi-professional focus, covering a range of disabilities including Stroke (submission guidelines included in Appendix J).

Abstract

Understanding cognitive impairment and emotional difficulties after stroke are key priorities for research and clinical practice. However, cognitive screening tools are not always sensitive to impairments after stroke, and psychological distress in this population remains poorly understood, despite high prevalence and secondary implications. Part one of this research aimed to determine the sensitivity and clinical validity of a new stroke-specific, cognitive screening tool, the OCS-Plus, compared with the MoCA, a more established screening tool. Part two sought to better understand why cognitive impairment is associated with greater psychological distress after stroke, by exploring the role of illness appraisals. Eighty participants were consecutively recruited from Oxfordshire stroke wards over a 15-month period and completed a battery of assessments of mood, cognition and illness appraisals which were analysed cross-sectionally. The OCS-Plus was found to have good convergent validity and sensitivity when compared with the MoCA. Cognitive impairment significantly predicted depressive symptoms but was not predictive of anxiety when controlling for depression. The relationship with low mood was shown to be mediated by perceptions of control in this sample. Specifically, this research showed that higher levels of cognitive impairment were significantly associated with lower perceived control and accounted for the relationship between cognitive impairment and mood. Perceptions of threat and illness coherence were also predictive of low mood but were not predicted by degree of cognitive impairment. Illness appraisals may therefore be a target for intervention in the early stages after stroke, although prospective research is required to establish causality.

Key Words: Stroke, Cognitive Impairment, Cognitive Screening, Post-Stroke Depression, Illness Perceptions

Understanding Post-Stroke Cognitive Impairment and its Sequelae Using the OCS-Plus

Stroke is a prevalent cause of disability, with around 130,000 cases occurring in the United Kingdom each year (1). Post-Stroke Cognitive Impairment (PSCI) is a common symptom; as many as 91% of acute stroke patients are estimated to have difficulties in at least one cognitive domain (2,3). Specific impairments vary depending on the location and nature of the infarct or bleed, but deficits in language, attention, memory, praxis and executive function are common (4–7). These difficulties tend to improve over time, most significantly during the acute stage (8), although cognitive impairment can persist or deteriorate to dementia diagnoses (9,10). Difficulties with executive functioning, attention and processing speed are particularly likely to continue over the longer-term (11,12). In milder strokes and for individuals with greater cognitive reserve, impairment can be less severe and span fewer cognitive domains (13).

PSCI can negatively impact engagement with neurorehabilitation (14) and is associated with increased levels of disability, reduced social participation and poorer quality of life (15,16). Individuals with PSCI are also more likely to experience anxiety and depression (17–19). Post-stroke depression, which affects around one third of stroke survivors (20), can impede functional recovery (21) and, post-stroke anxiety, occurring in 10% of stroke patients (22), is predictive of reduced independence (23). Understanding these changes after stroke is therefore a key research priority (24).

Assessing Post-Stroke Cognitive Impairment

PSCI is typically identified using screening tools, such as the Montreal Cognitive Assessment (MOCA (25)), and the Mini-Mental State Examination (MMSE; 26). These tools were not initially designed for use in stroke and so are not always

sensitive to stroke-related cognitive deficits (27). More recently, stroke-specific tools have been developed which accommodate for frequent symptoms of stroke that can impact assessment, such as aphasia and neglect (28,29). For example, the Oxford Cognitive Screen (OCS (2)) is a tool designed specifically to assess domain-specific cognitive impairments in the acute post-stroke phase. However, this acute focus may mean it is not always sensitive to the detection of more subtle PSCI. The OCS-Plus has therefore been designed as a stroke-specific domain-general screening measure, with particular sensitivity to deficits in memory and executive function (30). This measure has been normed and validated in a healthy aging cohort, including a task-by-task validation, and recently has been validated for use in the stroke population (31). However, no thorough comparison has been made to alternatives commonly used in stroke rehabilitation. This is important given the lack of a gold-standard cognitive screening tool in this population. Indeed, assessing the comparative utility of screening measures is a current priority for European stroke research (32).

Cognition, Mood and Illness Appraisals

It is well established that PSCI impacts mood over and above any bidirectional effect of mood on cognition (33) and associations between cognitive impairment and depressive symptoms are consistently identified across the literature (11,19,34). However, there is a lack of consensus as to the mechanism underlying this relationship. Historically, research has focused on understanding neurobiological predictors of post-stroke depression, such as neurotransmitter disturbances (35), hormonal dysfunction (36) and neuroanatomical changes to frontal-subcortical circuitry (37,38). Fewer studies have considered the psychological process, or social and emotional predictors of depression after stroke, such as pre-existing psychological difficulties (22).

A relationship between cognition and anxiety has also been postulated (17), but is less well researched. There is some support for a lesion location hypothesis (39) however, associations may only exist due to co-morbid post-stroke depression (19). Understanding the significance of this potential relationship may be important for determining clinical priorities after stroke.

It has been suggested that the relationship between PSCI and depression could be attributed to perceptions of loss, based on findings that the cognitive deficits most associated with depression are those that are likely to be immediately apparent and disabling, such as aphasia, amnesia and perceptual distortions (40). This is supported by wider research demonstrating a relationship between depression and aphasia (41) and is in line with the Common Sense Model (CSM) which emphasises the role of beliefs in coping with health difficulties (42). Cognitive and emotional representations are proposed to guide the selection of coping strategies, which influence important outcomes, such as engagement in rehabilitation and psychological adjustment. Those with greater perceptions of threat, who consider their illness to be highly symptomatic, chronic and having serious consequences, are more likely to demonstrate avoidance and high emotional expression, whereas those with a strong perception of controllability, who see their illness as coherent and manageable, are less likely to experience psychological distress (43,44). The role of illness appraisals in post-stroke coping has recently been established, with several studies demonstrating a relationship between appraisal style and psychological outcomes (e.g. (45,46)). However, the possible impact of cognition on stroke-related appraisals is yet to be explored. It may be that cognitive impairment is feared after stroke, as is the case with dementia (47), and indeed, clinicians can avoid discussions about risk of dementia after stroke (48). Likewise, cognitive rehabilitation is often a lower priority in stroke care (49), which could

contribute to negative perceptions about recovery. In line with the CSM, those with more significant impairment may therefore consider their stroke to be more threatening and less easily controlled, in turn mediating the relationship between cognition and mood. Identifying a role of illness appraisals would present an avenue for intervention, and perhaps prevention, of psychological distress in those with PSCI.

Aims of this Research

The objectives of this research were twofold. Part one compared the OCS-Plus to a commonly used alternative, the MoCA, in the early stages after stroke. Part two investigated the relationships between cognitive impairments, mood and illness appraisals.

To this end, this study aimed to answer the following questions:

1. Is the OCS-Plus a valid and sensitive tool for detecting cognitive impairment in the stroke population in relation to a current clinical standard?

   It was hypothesised that the OCS-Plus would be at least as successful in detecting cognitive difficulties as the MoCA and have good discriminative validity.

2. Can the relationship between cognition and depressive symptoms after stroke be identified using the OCS-Plus?

   It was hypothesised that there would be a relationship between cognition (MoCA or OCS-Plus score) and depressive symptoms.

3. Is there a relationship between cognition and anxious symptoms after stroke, which can be identified using the OCS-Plus?

It was predicted that any identified relationship between cognition and anxious symptoms would be accounted for by comorbid symptoms of depression.

4. What is the impact of stroke-related illness appraisals on the relationship between cognition and mood?

   It was hypothesised that illness appraisals (perceived threat and control) would at least partially mediate the relationship between PSCI and depressive symptoms after stroke.

Method

Design

This study used a cross-sectional design and reporting is in line with the STROBE checklist (50).

Participants

Participants were consecutively recruited from the John Radcliffe Acute Stroke Unit and the Oxfordshire Stroke Rehabilitation Unit over a 15-month period (June 2021 to August 2022). The majority were assessed while in the subacute hospital, typically at bedside, but a proportion were assessed at home. Patients were invited to participate if they i) Were over 18 years old and had suffered a stroke in the last six months; ii) Were willing and able to give informed consent to participate; iii) Were well enough to concentrate for a 30-60-minute assessment. Patients were excluded if they i) Had a prior intellectual disability; ii) Had insufficient English language skills to comprehend the initial orienting questions on the OCS-Plus; iii) Were judged by the care team to be too unwell to participate (e.g. delirium, fatigue or medical complications).

All participants were recruited within a wider study (OCS-Recovery) and gave informed consent under NHS ethics (Ethics Ref: 18/SC/ 05501, Appendix K[6]). In some cases, assessments were completed in one testing session but typically, assessment batteries were staggered with breaks. Recruitment was terminated when 77 participants had completed the OCS-Plus and questionnaire measures, based on a priori power analyses, ensuring at least 80% power per analysis (detailed in Appendix L and conducted using G*Power (51)).

Measures

Cognition

Cognition was assessed using the OCS-Plus, a tablet-based stroke-specific screening tool comprised of 10 subtests (30). Impairment is scored over 18 criteria in comparison to typically aging adults of a similar age. The OCS-Plus was administered on a Window Surface Tablet Pro via MatLab (52) and takes an average of 21 minutes to complete in the sub-acute population (31). Participants also completed the MoCA Version 8 (25), a pen and paper screening tool, designed to assess mild cognitive impairment, which takes approximately 10 minutes to complete. It is comprised of 11 subtests and a total score below 26/30 is considered indicative of impairment (25). Researchers who administered this tool were certified in administration.

Mood

Mood was assessed using the Patient Health Questionnaire (PHQ-9), a 9 item valid and reliable measure of depression (53), and the Generalised Anxiety Disorder

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[6] A substantial amendment was made for the present study, details of which are given in Part 2 of Appendix K.

Assessment (GAD-7), a seven item valid and reliable measure of anxiety (54). These measures were chosen due to their high sensitivity and specificity, short administration time and use in Stroke Rehabilitation services (55). Both measures have a cut off score of 10 for identifying cases of depression or anxiety.

Illness Appraisals

Illness appraisals were measured using a stroke-specific version of the Revised Illness Perception Questionnaire (Stroke IPQ-R; Appendix M). The IPQ-R (56) is a validated and reliable measure of nine domains of illness appraisals which has been adapted for use in stroke survivors with good internal consistency (57). Scores on the identity, emotional, timeline, consequences and cyclical subscales can be summed to give an overall measure of perceived threat. Likewise, scores on the personal control, treatment control and coherence subscales can be summed to given an overall measure of perceived control (43,44). Questions assessing causality were not administered as this domain can only be analysed using factor analysis.

Stroke Severity and Impact

The Barthel Index for Activities of Daily Living (58) was used as an approximation of physical disability after stroke. Scores across 10 items range from 0-20, with lower scores reliably indicating increased disability. This was either completed by a clinician or the researcher. The NIH Stroke Scale (NIHSS) was also collected from patient records. This is an 11-item scale used to assess the severity of a stroke with high reliability and validity (59).

Data Analysis

Part 1 – Validity of the OCS-Plus

To determine convergent validity, the OCS-Plus was compared to the MoCA using correlational analysis. It was predicted that there would be at least a moderate negative correlation ($r > -0.4$) between the number of impairments on the OCS-Plus and overall score on the MoCA. To determine divergent validity, scores on the OCS-Plus were compared to the Barthel using correlational analysis. It was anticipated that scores would not be significantly correlated. A sample size of 77 was calculated as being sufficiently powered to detect a correlation of at least 0.3.

To assess sensitivity and specificity, the presence of impairments on the OCS-Plus was compared to overall categorisation of impairment on the MoCA (i.e. a score <26). True positives and negatives (i.e. impaired or not impaired participants) were calculated using the MoCA as an approximated 'truth'. It was anticipated that Receiver Operating Characteristic (ROC) analysis would generate an area under the curve (AUC) of at least 0.7, as is considered acceptable in diagnostic test assessment (60).

Part 2 – Cognition, Mood and Appraisals

Number of impairments on the OCS-Plus and total score on the MoCA were compared to scores on the PHQ-9 and GAD-7 using multivariate linear regression, controlling for physical impairment and self-reported history of mental health difficulties. Given past literature, we anticipated that PSCI would significantly predict degree of symptoms of depression and anxiety, with at least a medium effect ($f^2 = 0.15$), irrespective of the screening tool used. PHQ-9 score was controlled for when assessing the relationship between PSCI and anxious symptoms to determine if the relationship

existed aside from the impact of depression. A large effect was predicted for this analysis ($f^2 = 0.35$). If cognition was found to be predictive of depressive symptoms as hypothesised, we planned to conduct mediation analyses to determine the impact of appraisals on this relationship. It was predicted that illness appraisals (perceived threat and perceived control) would significantly account for the variability in depressive symptoms, with a large effect ($f^2 = 0.35$), and that the effect of cognitive impairment on depressive symptoms would decrease substantially when perceived threat and perceived control were entered into the model (61). A sample of 77 participants was considered sufficient to ensure each of these analyses had at least 80% power.

Results

Analyses were carried out in R Studio (R version 4.2.1) and are reported below. Supplementary analyses are included in Appendix L.

Demographics

Eighty participants took part in this study in total. However, due to the feasibility constraints of completing staggered research visits on a busy rehabilitation ward, not all measures were completed by all participants (see Table 1). Questionnaire measures were prioritised as the primary outcome of this research. Similarly, 12 participants did not complete all subtasks of the OCS-Plus due to ward-based interruptions (e.g. therapy and family visits) or severity of pre-existing visual impairment (e.g. macular degeneration), and so impairments were scored as a proportion of the tasks completed. The demographics of the sample are presented in Table 2 and stroke-specific information in Table 3. No participants had a prior diagnosis

of dementia, although three were reported to have Mild Cognitive Impairment (MCI)[7] and 30 had at least one previous stroke.

Table 1.

A Breakdown of the Tasks Completed by Participants in Addition to the OCS-Plus, Which was Completed by all 80 Participants

Task	n
Questionnaires and MoCA	58
Questionnaires only	19
MoCA only	3

Table 2.

Demographic Information

Characteristic	Mean (SD)	Percentage
Age (years)	73.55 (12.61)	<60: 12.5%; 60-70: 27.5%: >70: 60%
Education (years)	13.59 (3.43)	≤12 years: 42.5%, >12 years: 57.5%
Sex	-	F: 46.3%; M: 53.8%
Handedness	-	L: 11.3%; R: 87.5%; A: 1.3%
Ethnicity	-	White British: 95%; Asian British: 2.6%; White Other: 1.3% Black British: 1.3%

[7] MCI is defined as cognitive decline greater than expected for a person's age and education that does not significantly impact activities of daily living (62).

Table 3.

Stroke Specific Information

	n	Mean (SD)	Percentage
Time Since Stroke (days)	80	35.77 (23.05)	-
Stroke Type	80	-	Ischemic: 88.8%; Haemorrhagic: 10.0%; Other/ Unspecified: 1.3%
Stroke Side	80	-	L: 38.8%, R: 51.3%, B:10.0%
Stroke Severity (NIHSS Score)	71	8.20 (5.58)	-
Degree of Physical Difficulty (Barthel)	80	10.45 (6.54)	-
Past History of Stroke	80	-	None: 68.9% One: 21.3%, Multiple: 5.0%, Unknown: 5.0%
Reported History of Mood Difficulties	80	-	25.0% (Depression: 17.5%; Anxiety: 7.5% Other: 5%)
Current Depression (PHQ-9)	77	7.40 (6.4)	Scored above clinical threshold: 27.5%
Current Anxiety (GAD-7)	77	4.75 (5.69)	Scored above clinical threshold: 21.3%

Note: n is lower for NIHSS as this was not always available from patient records. n is lower for PHQ-9 and GAD-7 due to some participants not completing all measures.

Analysis

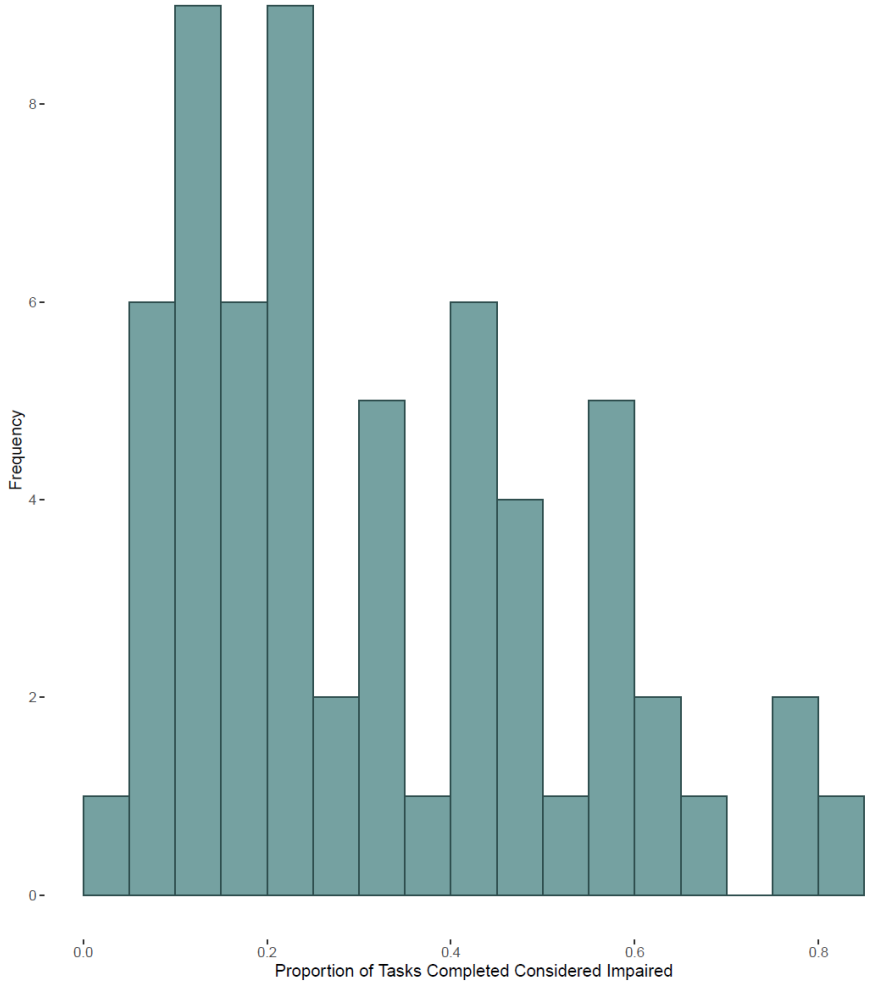
Part 1 – Validity of the OCS-Plus

Convergent Validity. When using the OCS-Plus, 99% of the sample were considered to have an impairment in at least one domain (see Figure 1 for frequencies of impairments). In contrast, only 65% were identified as having an impairment using the 26 point cut-off from the MoCA. As predicted, there was a strong negative correlation between proportion of impairments on the OCS-Plus and overall score on the MoCA; $r(59) = -.77$, $p < .01$)[8].

[8] Assumptions of normality assumed due to large sample size.

Figure 1.

The Proportion of Impaired Tasks on the OCS-Plus, as Frequencies Within the Sample Population



Divergent Validity. In contrast to our prediction, there was a moderate correlation between scores on Barthel and proportion of impairments on the OCS-Plus; $r(78) = -.35$, $p = <.01$)[9].

[9] Assumptions of normality assumed due to large sample size.

Sensitivity and Specificity. Sensitivity and specificity, reported in Table 4, were calculated using the MoCA as a reference standard for cognitive impairment. As shown in Figure 2, a ROC curve analysis was suggestive of excellent discrimination between those with and without cognitive impairment; AUC = 0.93 (63).

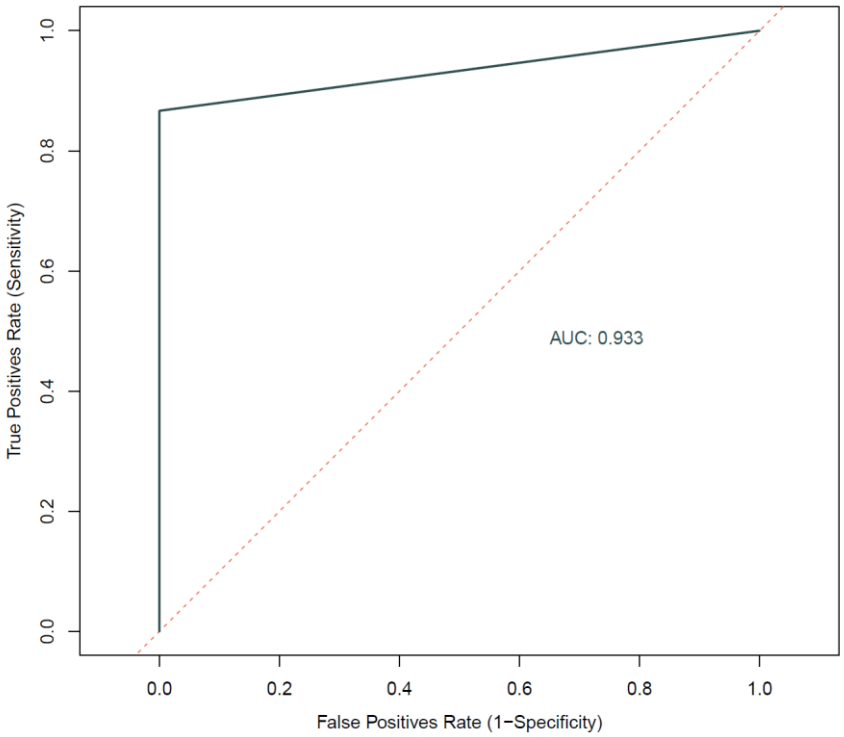
Table 4.

Sensitivity and Specificity of the OCS-Plus Compared to the MoCA (n=61)

		Cognitive Impairment	
		+	-
OCS-Plus	+	True Positives – 52	False Positives – 8
	-	False Negatives - 0	True Negatives – 1
		Sensitivity – 100	**Specificity** – 11.1

Figure 2.

A ROC Curve Visualisation of the Relationship Between the Sensitivity and Specificity of the OCS-Plus



Part 2 – Cognition, Mood and Appraisals

Cognition and Mood. Multivariate linear regression was used to investigate the impact of cognitive impairment on anxious and depressive symptoms, controlling for physical impairment and history of mental health difficulties[10]. A significant regression model was found for the relationship between PSCI and depressive symptoms using both the MoCA and OCS-Plus (Table 5), with cognitive impairment significantly predicting depressive symptoms. A significant regression model was also found for the

[10] Assumptions of linearity, absence of multicollinearity and non-autocorrelation met. Normality assumed based on sample size and regression analysis considered robust to slight heteroskedasticity in the data.

relationship between PSCI and anxious symptoms using the MoCA and OCS-Plus, with cognitive impairment significantly predicting anxious symptoms (Table 6 – Model 1). However, when depressive symptoms were included in the model, cognitive impairment was no longer a significant predictor of anxiety (Table 6 – Model 2). The relationships between OCS-Plus impairments and psychological distress are shown graphically in Figure 3.

Table 5.

Regression Analyses Showing the Relationship Between Cognitive Impairment and Depressive Symptoms, Using the OCS-Plus (Regression 1) and MoCA (Regression 2)

	b	**SE (*b*)**	***t***
Regression 1 (Impact of OCS-Plus Impairments on PHQ-9 Score)			
Constant	1.83	2.05	0.90
OCS-Plus Impairments	10.20	3.35	3.05**
Mental Health History	4.33	1.62	2.66**
Barthel Score	0.11	0.11	1.03
Regression 2 (Impact of MoCA Score on PHQ-9 Score)			
Constant	13.35	3.21	4.16**
MoCA Score	-0.40	0.14	-2.85**
Mental Health History	3.49	1.74	2.00
Barthel Score	0.04	0.13	0.29

Notes: R^2 = .19 for Regression 1 (n=77). R^2 = .21 for Regression 2 (n=57).

*p<.05, **p<.01.

Table 6.

Regression Analyses Showing the Relationship Between Cognitive Impairment and Anxious Symptoms, Using the OCS-Plus (Regression 3) and MoCA (Regression 4)

	Model 1			Model 2		
	b	**SE (*b*)**	***t***	***b***	**SE (*b*)**	***t***
Regression 3 (Impact of OCS-Plus Impairments on GAD-7 Score)						
Constant	-0.76	1.73	-0.44	-1.72	1.36	-1.27
OCS-Plus Impairments	9.94	2.83	3.51**	4.53	2.35	1.93
Mental Health History	3.63	1.37	2.65**	1.34	1.12	1.19
Barthel Score	0.14	0.10	1.40	0.07	0.08	0.96
PHQ-9 Score	-	-	-	0.53	0.08	6.85**
Regression 4 (Impact of MoCA Score on GAD-7 Score)						
Constant	9.20	3.07	2.30**	-0.05	2.46	-0.02
MoCA	-0.31	0.13	-2.31*	-0.03	0.10	-0.33
Mental Health History	2.97	1.67	1.78	0.55	1.21	0.46
Barthel Score	0.07	0.13	0.51	0.04	0.09	0.44
PHQ-9 Score	-	-	-	0.69	0.09	7.62**

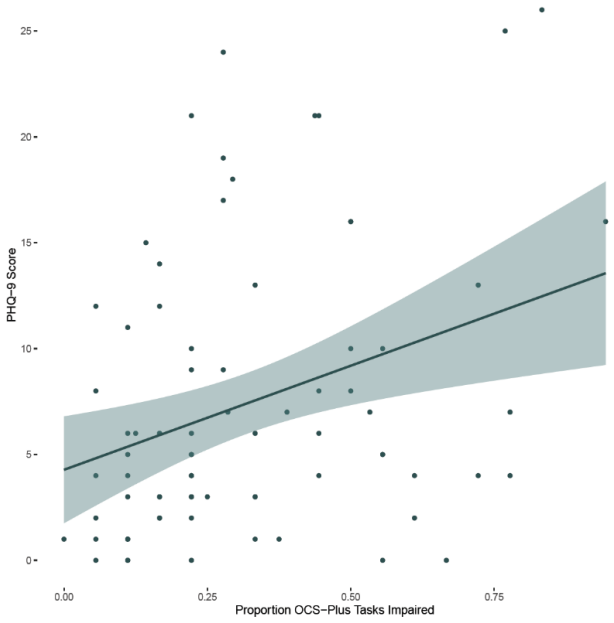
Notes: R^2 = .22 for Regression 2 (n=77). R^2 = .16 for Regression 4 (n= 57)

*p<.05, **p<.01.

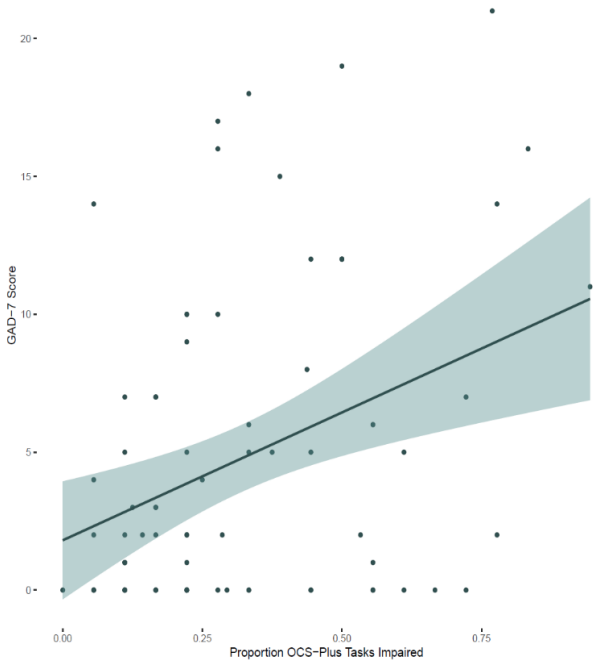
Figure 3.

Graphs to Show the Relationship Between Proportion of Impairments on the OCS-Plus and PHQ-9 Score (a) and GAD-7 Score (b), With Regression Line and Confidence Interval (Shaded Area)

a)



b)



Impact of Illness Appraisals. Causal mediation analysis was carried out to consider the possible impact of appraisals (perceived threat and control) on the relationship between cognition and depressive symptoms. When calculating overall scores for perceived threat and perceived control using the Stroke IPQ-R, only the former had acceptable internal consistency (α = .82). A perceived control score was therefore generated by summing the personal and treatment control subscales (α = .74) only, with illness coherence considered separately. A significant regression model was found for the relationship between degree of PSCI and perceived threat (controlling for physical health and mental health history), but PSCI was not a significant predictor. A significant regression model was found for the relationship between PSCI and depressive symptoms (65% of the variance explained), with greater perceived threat significantly predicting increased depressive symptoms (*b = 0.18).*

A significant regression model was found for the relationship between PSCI and perceived control (9% of the variance explained), with higher levels of PSCI predicting lower perceived control (*b = -9.28*). A significant regression model was also found for the relationship between PSCI and illness coherence, accounting for 13% of the variance, with higher levels PSCI predicting reduced illness coherence (*b = -7.98*). These variables were also significantly predictive of depressive symptoms: greater perceived control predicted lower PHQ-9 score (*b = -0.32,* accounting for 31% of the variance), as did greater illness coherence (*b = -0.30,* accounting for 24% of the variance). The association between cognitive impairment and depressive symptoms decreased but was still significant when perceived controllability was included in the model ($F(4, 72) = 7.95$, $p < 0.01$, $f^2 = 0.44$), with an R^2 of 0.31 (Table 7). A bootstrapping procedure with 1000 resamples indicated a significant indirect effect of cognition on depressive symptoms through perceived controllability ($p < .05$), as shown

in Figure 4. The association between cognitive impairment and depressive symptoms decreased when illness coherence was included in the model (*b = 7.79*), but the indirect effect was not significant (p = .80).

Table 7.

Regression Analysis Showing the Impact of Perceived Control on the Relationship Between Cognitive Impairment and Depressive Symptoms.

	b	**SE (*b*)**	***t***
Constant	19.76	5.54	3.57**
OCS-Plus Impairments	7.27	3.24	2.25*
Perceived Control Score	-0.32	0.09	-3.45**
Mental Health History	3.64	1.53	2.38*
Barthel Score	0.10	0.11	0.92

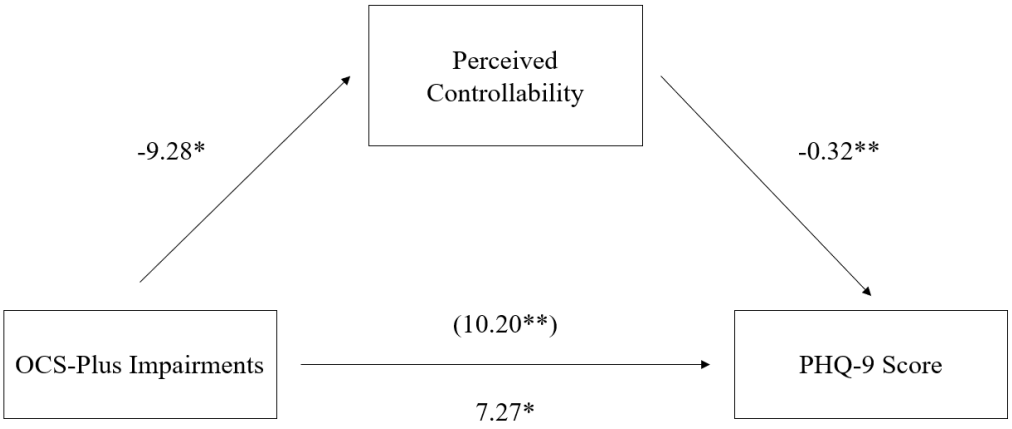
Notes: R^2 = .31, n=77.

*p<.05, **p<.01.

Figure 4.

Graphical Representation of the Mediation Analysis between OCS-Plus Impairments (Independent Variable), PHQ-9 Score (Dependent Variable) and Perceived Controllability (Mediator). Co-efficients for Direct and Indirect Effects are Stated, With Brackets Indicating an Indirect Effect. n=77.

**p<.05, **p<.01.*



Discussion

This research demonstrated that the OCS-Plus is a valid and sensitive tool for assessing cognition in the stroke population and was successfully applied as a research tool in understanding the relationship between PSCI and mood after stroke, with perceptions of control found to have a potentially mediating role.

Validity of the OCS-Plus

Part one of this study demonstrated that the OCS-Plus can be used to detect cognitive difficulties at least as successfully as the MoCA, but is more sensitive to subtle cognitive changes. The majority of participants were reported as having at least

some degree of PSCI as measured by the OCS-Plus. This is in line with prevalence estimates in the early stages post-stroke (2,3) and higher than identified using the MoCA. This sensitivity came at the cost of specificity. However, this may well be an artefact of very few people in the study having no cognitive impairment and, so may be better measured in a mixed sample including healthy aging participants. Moderate correlation between the OCS-Plus and physical impairment was unexpected and suggestive of weaker discriminative validity. However, cognitive impairment may have impacted some items on the Barthel e.g. independence in self-care tasks, which would explain the correlation found. Indeed, overall we might still expect that factors like stroke severity would have a weak relationship with cognitive impairment.

Given the discrepancy in rate of impairment identified by the MoCA and OCS-Plus, subtle cognitive changes may be underappreciated in clinical settings, where non-stroke-specific screening tools are used. This may mean that impact on engagement with neurorehabilitation (14) is overlooked. Use of a more sensitive tool, such as the OCS-Plus, could therefore be helpful in developing robust formulations of post-stroke difficulties and managing patient expectations as they return to tasks of daily living which could be impacted by subtle changes.

Cognition, Mood and Illness Appraisals

In the second part of this study, the OCS-Plus was applied as a research tool to investigate the relationship between cognitive impairment and mood. In line with initial hypotheses and previous research (17,19,33,40), degree of PSCI was shown to predict depressive symptoms, even when controlling for physical impairment and previous psychological difficulties. A similar relationship with anxiety did not exist when considering the impact of comorbid depression. A mediation analysis was then

undertaken to better understand why this relationship between cognitive impairment and depressive symptoms exists. Consistent with cognitive theory and previous research (42,45,46), we demonstrated that stroke-related illness appraisals (namely, total threat, controllability and illness coherence) are related to levels of depressive symptoms. Of these, threat perceptions explained more of the variance in mood, consistent with wider health research (43). Furthermore, this study presents new evidence for a relationship between cognitive impairment and perceived controllability, which mediated the relationship between cognition and mood in this sample. This sheds light on the suggestion that this relationship can be understood with respect to the immediate sense of disability caused by post-stroke changes in cognition (40). Rather than an increased sense of threat, it may be that these difficulties feel less easy to personally manage and more resistant to intervention, which in turn, impacts mood. This is similar to recent findings in the dementia population, whereby locus of control was shown to be a key predictor of depressive symptoms (64), although a relationship between cognitive impairment and perceptions of control is a relatively novel finding in the stroke population. This is consistent however, with lay perceptions of cognitive impairment, such as negative beliefs about control in carers of those with dementia (65), and with reports by stroke-survivors of receiving limited support with memory problems after stroke (66).

PSCI explained only 9% of the variance in perceived control, which is in line with previous suggestions that illness characteristics are not always the most important predictor of how a person perceives their illness (67,68). It is likely that perceptions of controllability form just one part of the relationship between PSCI and mood, with other contributing factors. For example, impairments may increase maladaptive coping strategies such as wishful thinking, catastrophising or helplessness (67), due to rigidity

of thinking. PSCI may also impact sense of self and personhood, which would in turn impact adjustment (69). Future research could seek to determine whether the relationship between cognition and mood breaks down at higher and lower levels of impairment. For example, those with greater PSCI may have lower levels of insight which could bring about more positive coping (70,71).

Although exploratory in nature, the findings of this research suggest that even in the very early stages after stroke, individuals have beliefs about their stroke which impact upon coping and adjustment. In particular, perceptions of controllability and illness coherence seem to be influenced by severity of cognitive impairment. Interventions which support people to reappraise assumptions of uncontrollability may be effective in reducing initial psychological distress, which may prevent later mood-related difficulties. Likewise, psycho-education for those struggling to understand their stroke and its impact may also be important. Investigating this relationship, with more specific initial hypotheses about controllability and coherence, may be an important avenue for future research, as well as investigating whether these factors also contribute to anxiety after stroke. Future research could also investigate how deficits in specific domains impact mood and illness appraisals.

Strengths and Limitations

The recruited sample is largely representative of the clinical population due to consecutive sampling from clinical settings, and so the findings of this research are likely to be directly relevant to the stroke population, despite the inevitable heterogeneity in this group (e.g. lesion location, severity etc), and difficulties with not all participants completing all measures resulting from the logistical constraints of ward-based research. This inclusive recruitment approach is also likely to have reduced

sources of bias. However, sampling in Oxfordshire meant that the recruited sample was not representative of the wider UK in terms of ethnicity and education. Similarly, the impact of demographic and social factors, which may well have impacted illness beliefs and coping style (72), were not explored in this study. Limits to cross-sectional methodology mean that further research is needed to truly understand the causal role of cognitive impairment and illness appraisals in symptoms of depression and anxiety. By considering impairment at only one time point, it was not possible to distinguish cognitive impairment resulting from the current stroke from pre-existing impairment associated with a previous stroke or MCI. Furthermore, we did not explore the reciprocal relations between mood, illness appraisals and cognition but note the possibility that those with lower mood are in turn likely to have more negative illness appraisals and perhaps increased sensitivity to subjective cognitive impairment (73).

Conclusions

This research demonstrated that the OCS-Plus is a valid screen for identifying more subtle cognitive impairment after stroke and can be applied successfully as a tool for psychological research. It may therefore be useful to clinicians seeking a more sensitive screen of PSCI. When exploring relationships between PSCI, illness appraisals and mood, there was a clear impact of perceptions of threat, controllability and illness coherence on depressive symptoms. Perceptions of controllability in particular, may be important for better understanding the relationship between cognitive impairment and low mood after stroke, although further investigation of this mediating relationship will be important for establishing causation.

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Executive Summary of Theoretically Driven Research Project

Background and Aims

Many people experience difficulties with their thinking skills and emotions after having a stroke but much is still unknown about how best to assess, predict and manage these difficulties. Therefore, people who have experienced stroke, and professionals working clinically with stroke-survivors, have highlighted this as a key priority for research. This study was conducted in two parts and the background to each is summarised below:

1) Measures known as 'cognitive screening tools' are used to assess changes in thinking skills after stroke. However, it is not always easy to know which measure is best to use. This research sought to determine whether a relatively new screening tool, the Oxford Cognitive Screen Plus (OCS-Plus), is as good as, or even better than, a commonly used alternative, the Montreal Cognitive Assessment (MoCA), at detecting changes to thinking skills (i.e. 'cognitive impairment') after stroke.
2) Cognitive impairment has been shown to predict severity of emotional difficulties after stroke, however it is unclear why this is the case. Some have suggested that changes in mood occur as a result of disruptions in the brain caused by the stroke. However, this research proposed that the psychological response to having a stroke may also be important, and therefore sought to investigate whether the thoughts that people have about their stroke might explain the relationship between cognitive impairment and mood. In particular, it was predicted that those with greater changes to their thinking skills might see their stroke as more threatening and less easy to control, which could lead to them having more emotional difficulties.

How this Research was Conducted

Eighty people who had a stroke within the last six months were recruited from two stroke wards in Oxfordshire, over a period of 15 months. Participants completed a number of assessments of their thinking skills, mood and thoughts about their stroke. In part one, participants' scores on the OCS-Plus were compared to scores on the MoCA, to determine whether the measures were similarly able to detect cognitive impairment. The OCS-Plus was also compared to the MoCA to assess its sensitivity (i.e. its ability to correctly detect those with changes to their thinking skills) and its specificity (i.e. its ability to detect those who do not have changes to their thinking skills). In part two, this research tested whether the degree of cognitive impairment, predicted symptoms of low mood and anxiety in this sample. Statistical analysis was then used to determine whether people's thoughts about their stroke (i.e. how threatening and controllable they perceived it to be), could explain the relationship between cognitive impairment and mood.

Findings

Part one of this study showed that the OCS-Plus can be used to detect changes in thinking skills at least as successfully as the MoCA, and is better able to detect subtle changes. It was less successful in identifying those who do not have changes to their thinking skills after stroke. However, this could be explained by there being very few people in this sample with no cognitive impairment. Part two of this study showed that degree of cognitive impairment predicted low mood but not anxiety. Beliefs about the controllability of one's stroke seemed to explain this relationship, i.e. those with more severe changes to their thinking skills saw their stroke as more difficult to control and therefore experienced worse symptoms of low mood. Beliefs about the stroke being

more threatening (including how long the effects of the stroke might last and how severe the consequences might be) and having a reduced understanding of the stroke, were also important in predicting greater mood difficulties after stroke but were less strongly related to cognitive impairment.

Conclusions and Implications

This research showed that the OCS-Plus is a good measure of cognitive impairment after stroke and may be useful for clinicians seeking a sensitive screening tool. It could be used to help stroke-survivors and their families to better understand changes in their thinking skills after stroke and make appropriate adaptations. This research also showed that beliefs about controllability are important for understanding the relationship between cognitive impairment and low mood. Interventions which support individuals with understanding and managing their cognitive impairment after stroke may be important in preventing mood-related difficulties. However, the limitations of this study mean that further research is needed to confirm that this is a universal phenomenon.

Connecting Narrative

The elements of this thesis demonstrate several distinct contributions of Clinical Psychologists to an NHS workforce that is rapidly expanding to meet increasing demands for psychological therapy and interventions. There are now twelve registered psychological professions in the UK and Higher Education England suggest that by 2024, a 60% increase in the workforce is needed on 2020 levels to deliver the requirements of the NHS Long Term Plan (Health Education England, 2021). This means it is an exciting time to be beginning a psychological profession. However, it is also a time for Clinical Psychologists to be mindful of their highly qualified position within NHS teams and ensure that this can be justified. This thesis, with its focus on neurodevelopment, health psychology, adjustment and neuropsychology, exemplifies some of the skills of a Clinical Psychologist beyond therapeutic intervention. By drawing on a range of neuropsychological, cognitive and developmental theories, the broad contribution of a Clinical Psychologist is hereby illustrated.

The critical review of the literature highlights the ways in which Clinical Psychologists can contribute to advances in neurodevelopmental research, not just in terms of traditional therapies, but also in supporting with the development of technological interventions, as these approaches rapidly advance. The reviewed papers focus heavily on details relating to the technological aspect of extended reality interventions which, while exciting, means that the specific aims of the interventions, and how these relate to service user views and psychological theory, are somewhat neglected. When approaching the analysis of these papers, it felt important to me as a Clinical Psychology Trainee to consider this gap in the research and how it might be addressed. My position also helped me to carefully reflect on the language I used, as

well as the terminology of the studies I reviewed. I was taken aback by the deficit-based approach to discussing autism in the research, which starkly contrasted my own clinical training and work with autistic people. Clinical Psychologists may therefore be important contributors to future research in considering how autistic people can best be supported to enhance skills that are important to them, without a deficit-based framework.

The service improvement project demonstrates the leadership competencies of a Clinical Psychologist, not just in terms of the capacity to conduct clinical research but also their leading role in supporting patients who do not seem to benefit from mainstream clinical interventions. I saw how psychological theory can be vital when considering possible barriers to change and bringing a different approach to the formulation of clients who might otherwise be labelled as 'difficult'. When interviewing clinicians from the service, I was inspired by their passion to improve the service for this group and saw their appreciation of psychological leadership. This made me think more about the unique contribution that Clinical Psychologists can offer to physical health teams.

The theory-driven project in many ways exemplified to me how psychologists can truly act as scientist-practitioners. I found that my knowledge of real NHS contexts was invaluable to designing and conducting this research and likewise, my clinical skills were certainly enhanced throughout the research process. This for me, brought to life the idea that research within clinical settings can improve patient outcomes and satisfaction (NIHR, 2022). This project also helped me to develop my neuropsychological understanding and I saw how this knowledge of brain-behaviour relationships is essential to informing psychological intervention within the brain injury

population. In stroke, for example, it seemed impossible to consider the psychological response to the injury without also considering the structural impact on the brain and vice versa.

The reflective, research and leadership skills highlighted so far in this narrative, mean that Clinical Psychologists are potentially well placed to contribute to efforts to address issues of diversity, which have been particularly pertinent to my training journey in the wake of social justice movements and national conversations about race inequality. This subject matter is notably missing from this thesis and I am aware of the lack of diversity of the research samples recruited to these studies. While it may be easy to attribute this entirely to the demographics of Oxfordshire and Buckinghamshire, it is also important to consider where I might have improved this, for example, by considering the use of translators in my research or seeking to recruit from a wider range of services. I hope that this is something I continue not just to think about but act on as I begin my professional career.

Conducting this thesis has also developed my skills as a researcher, in and of itself. I have learned how to submit for ethical approval, perform statistical analysis using R, conduct a systematic review and use qualitative methodologies. I also learned the value of involving service users in the design of research and this helped me to reflect on who research is for and what it might meant to truly ‘do with’ rather than ‘do to’. I see the limits to my own research in this respect and have learned ways in which I might act differently in future. As my doctoral research comes to an end, I am thinking carefully about dissemination and knowledge translation, which is an intrinsic but often underestimated aspect of the research process. I hope to do service to those who

contributed to the design of my research and participated, by ensuring that this is not simply an after thought.

Overall, this thesis has supported my understanding of what it means to be a Clinical Psychologist and Researcher in today's NHS. I look forward to bringing this scientist-practitioner approach into my career as a Clinical Psychologist and hope that ultimately, I will be able to use these skills to improve patient care.

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Appendix A: Relevant Sections of Submission Guidelines for the Review Journal of Autism and Developmental Disorders

Types of papers

- ***Review Articles:*** *The preferred article length is 20-23 manuscript pages long (not including title page, abstract, tables, figures, addendums, etc.) Manuscripts of 40 pages (references, tables and figures counted as pages) have been published. The reviewers or the editor for your review will advise you if a longer submission must be shortened.*
- ***Brief Report:*** *The purpose of the concise review is to summarize literature on a specific topic and provide directions for future research or for improvement in clinical practice. Authors interested in writing a brief review should send an abstract and reference list to the Editor (Jill Fodstad, PhD) for initial consideration prior to standard peer review process. Please contact Dr. Fodstad at jfodstad@iupui.edu. Articles should be no more than 2,500 total words, not including the Title Page, Author Note, References, and Tables/Figures. Max of 2 Tables/Figures. No Supplementary Material. May fall under one of two areas:*

Abstract

Please provide an abstract of 120 words or less. The abstract should not contain any undefined abbreviations or unspecified references.

Keywords

Please provide 4 to 6 keywords which can be used for indexing purposes.

Text Formatting

Manuscripts should be submitted in Word.

- *Use a normal, plain font (e.g., 10-point Times Roman) for text.*
- *Use italics for emphasis.*
- *Use the automatic page numbering function to number the pages.*
- *Do not use field functions.*
- *Use tab stops or other commands for indents, not the space bar.*
- *Use the table function, not spreadsheets, to make tables.*
- *Use the equation editor or MathType for equations.*
- *Save your file in docx format (Word 2007 or higher) or doc format (older Word versions).*

Headings

Please use no more than three levels of displayed headings.

Abbreviations

Abbreviations should be defined at first mention and used consistently thereafter.

Footnotes

Footnotes can be used to give additional information, which may include the citation of a reference included in the reference list. They should not consist solely of a reference citation, and they should never include the bibliographic details of a reference. They should also not contain any figures or tables.

Footnotes to the text are numbered consecutively; those to tables should be indicated by superscript lower-case letters (or asterisks for significance values and other statistical data). Footnotes to the title or the authors of the article are not given reference symbols.

Always use footnotes instead of endnotes.

Terminology

Please always use internationally accepted signs and symbols for units (SI units).

References

Citation

Cite references in the text by name and year in parentheses. Some examples:

- *Negotiation research spans many disciplines (Thompson 1990).*
- *This result was later contradicted by Becker and Seligman (1996).*
- *This effect has been widely studied (Abbott 1991; Barakat et al. 1995; Kelso and Smith 1998; Medvec et al. 1999).*

Reference list

The list of references should only include works that are cited in the text and that have been published or accepted for publication. Personal communications and unpublished works should only be mentioned in the text. Do not use footnotes or endnotes as a substitute for a reference list.

Reference list entries should be alphabetized by the last names of the first author of each work. Journal names and book titles should be italicized

Tables

- *All tables are to be numbered using Arabic numerals.*
- *Tables should always be cited in text in consecutive numerical order.*
- *For each table, please supply a table caption (title) explaining the components of the table.*
- *Identify any previously published material by giving the original source in the form of a reference at the end of the table caption.*

- *Footnotes to tables should be indicated by superscript lower-case letters (or asterisks for significance values and other statistical data) and included beneath the table body.*

Electronic Figure Submission

- *Supply all figures electronically.*
- *Indicate what graphics program was used to create the artwork.*
- *For vector graphics, the preferred format is EPS; for halftones, please use TIFF format. MSOffice files are also acceptable.*
- *Vector graphics containing fonts must have the fonts embedded in the files.*
- *Name your figure files with "Fig" and the figure number, e.g., Fig1.eps.*

Figure Numbering

- *All figures are to be numbered using Arabic numerals.*
- *Figures should always be cited in text in consecutive numerical order.*
- *Figure parts should be denoted by lowercase letters (a, b, c, etc.).*
- *If an appendix appears in your article and it contains one or more figures, continue the consecutive numbering of the main text. Do not number the appendix figures,"A1, A2, A3, etc." Figures in online appendices [Supplementary Information (SI)] should, however, be numbered separately.*

Figure Captions

- *Each figure should have a concise caption describing accurately what the figure depicts. Include the captions in the text file of the manuscript, not in the figure file.*
- *Figure captions begin with the term Fig. in bold type, followed by the figure number, also in bold type.*
- *No punctuation is to be included after the number, nor is any punctuation to be placed at the end of the caption.*
- *Identify all elements found in the figure in the figure caption; and use boxes, circles, etc., as coordinate points in graphs.*
- *Identify previously published material by giving the original source in the form of a reference citation at the end of the figure caption.*

Appendix B: CRL Search Terms

Search Terms*

1. ‘Extended Reality’ OR ‘Virtual Reality’ OR ‘Virtual Environment’ OR ‘Augmented Reality’ OR ‘Augmented Environment’ OR ‘Simulat* Reality’ OR ‘Mixed Reality’ OR ‘VR’ OR ‘AR’ OR ‘XR’ OR ‘MR’
2. ‘Child*’ OR ‘Young’ OR ‘Youth*’ OR ‘Adolescen*’ OR ‘Student*’ OR ‘Teen*’ OR ‘Boy*’ OR ‘Girl*’ OR ‘P?ediatric*’ OR ‘Minor*’
3. “Social Skill*’ OR “Social Interacti*’ OR “Social Competence’ OR “Social Responsiveness’ OR ‘Social Motivation’ OR ‘Social Abilit*’ OR ‘Social Awareness’ OR ‘Conversation Skill*’ OR ‘Conversation Abilit*’ OR ‘Friendship Skill*’ OR ‘Peer interacti*’ OR ‘Emotion* Recognition’ OR ‘Facial Recognition’ OR ‘Perspective Taking’ OR ‘Turn Taking’ OR ‘Turn-Taking’ OR ‘Theory of Mind’ OR ‘Communicat*’ OR ‘Interact*’
4. ‘Autis*’ OR ‘Asperger*’ OR ‘ASD’ OR ‘ASC’ OR 'Pervasive Developmental Disorder' OR ‘PDD’ OR ‘HFA’
5. 1 AND 2 AND 3 AND 4

*Key word and thesaurus searches were also conducted for each of the above terms, where possible within each database.

Terms Considered but Excluded

‘TOM’, ‘Toddler*’, ‘Baby’, ‘Babies’, ‘Newborn*’, ‘Pre-school*’, ‘Preschool*’

Appendix C: The Downs and Black Checklist

The Downs and Black Checklist (below) was used to assess study quality. The scoring of item 27, referring to the power of the study, was modified. Instead of rating according to an available range of study powers, we rated whether or not a power calculation was performed. Items which were not applicable to single case and small-N studies are marked*. Items which were not applicable to studies which did not use statistical analysis are marked †. Questions 14 and 22 were not applicable to studies which did not have a control intervention. All other items were rated as *Yes* (scored as 1) or *No* (scored as 0) or *Unable to Determine* (scored as 0). Items 14 to 26 specifically assess bias and so were used to generate a risk of bias score.

1. *Is the hypothesis/aim/objective of the study clearly described?*
2. *Are the main outcomes to be measured clearly described in the Introduction or Methods section?* If the main outcomes are first mentioned in the Results section, the question should be answered no.
3. *Are the characteristics of the participants included in the study clearly described?* In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case-control studies, a case-definition and the source for controls should be given.
4. *Are the interventions of interest clearly described?* Treatments and placebo (where relevant) that are to be compared should be clearly described.
5. *Are the distributions of principal confounders in each group of subjects to be compared clearly described?* A list of principal confounders is provided.
6. *Are the main findings of the study clearly described?* Simple outcome data (including denominators and numerators) should be reported for all major findings so that the reader can check the major analyses and conclusions.
7. *Does the study provide estimates of the random variability in the data for the main outcomes?* In non-normally distributed data the inter-quartile range of results should be reported. In normally distributed data the standard error, standard deviation or confidence intervals should be reported. If the distribution of the data is not described, it must be assumed that the estimates used were appropriate and the question should be answered Yes.
8. *Have all important adverse events that may be a consequence of the intervention been reported?* This should be answered yes if the study demonstrates that there was a comprehensive attempt to measure adverse events. (A list of possible adverse events is provided).

9. *Have the characteristics of patients lost to follow up been described?* This should be answered yes where there were no losses to follow up or where losses were so small that findings would be unaffected by their inclusion. This should be answered no, where the study does not report the number of patients lost to follow up.*
10. *Have actual probability values been reported (e.g. 0.035 rather than <.05) for the main outcomes except where the probability value is less than 0.001?* †
11. *Were the subjects asked to participate in the study representative of the entire population from which they were recruited?* The study must identify the source population for participants and describe how the participants were selected. Participants would be representative if they comprised the entire source population, an unselected sample of consecutive participants, or a random sample. Random sampling is only feasible where a list of all members of the relevant population exists. Where a study does not report the proportion of the source population from which the patients are derived, the question should be answered as unable to determine.
12. *Were those subjects who were prepared to participate representative of the entire population from which they were recruited?* The proportion of those asked who agreed should be stated. Validation that the sample was representative would include demonstrating that the distribution of the main confounding factors was the same in the study sample and the source population.
13. We*re the staff, places and facilities where the patients were treated, representative of the treatment the majority of patients receive?* For the question to be answered yes the study should demonstrate that the intervention was representative of that in use in the source population. The questions should be answered no if, for example, the intervention was undertaken in a specialist centre unrepresentative of the hospitals most of the source population would attend.
14. *Was an attempt made to blind study subjects to the intervention they have received?* For studies where the patients would have no way of knowing which intervention they received, this should be answered yes.*
15. *Was an attempt made to blind those measuring the outcomes of the intervention?*
16. *If any of the results of the study were based on 'data dredging', was this made clear?* Any analysis that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analyses were reported, then answer yes.
17. *In trials and cohort studies, do the analyses adjust for different lengths of follow up of patients, or in the case-control studies, is the time period between the intervention and outcome the same for cases and controls?* Where follow-up was the same for all study patients, the answer should be yes. If different lengths of follow-up were adjusted for by, example, survival analysis the answer should be yes, studies where differences in follow-up are ignored should be answered no.**
18. *Were the statistical tests used to assess the main outcomes appropriate?* The statistical techniques must be appropriate to the data. For example non-parametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered yes. If the distribution of the data (normal or not) is not described it must be assumed that the estimates used were appropriate and the questions should be answered yes.
19. *Was compliance with the intervention/s reliable?* Where there was non-compliance with the allocated treatment or where there was contamination of one group, the question should be answered no. For studies where the effect of any misclassification was likely to bias any association to the null, the question should be answered yes.

20. *Were the main outcome measures used accurate (valid and reliable)?* For studies where the outcome measures are clearly described, the question should be answered yes. For studies which refer to other work or that demonstrated the outcomes measures are accurate, the question should be answered as yes.
21. *Were the participants in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?* For example, patients for all comparison groups should be selected from the same hospital. The question should be answered unable to determine for cohort and case control studies where there is no information concerning the source of patients included in the study.* †
22. *Were study participants in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?* For a study which does not specify the time period over which patients were recruited, the question should be answered as unable to determine.*
23. *Were study subjects randomised to intervention groups?* Studies which state patients were randomised should be answered yes except where method of randomisation would not ensure random allocation. For example, alternate allocation would score no because it is predictable.* †
24. *Was the randomised intervention assignment concealed from both patients and health care staff until recruitment was complete and irrevocable?* All non-randomised studies should be answered no. If assignment was concealed from patients but not from staff, it should be answered no.* †
25. *Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?* This question should be answered no for trials if: the main conclusion of the study were based on analyses of treatment rather than intention to treat; the distribution of known confounders in the different treatment groups was not described; or the distribution of known confounders differed between the treatment groups but was not taken into account in the analyses. In non-randomised studies, if the effect of the main confounders was not investigated or confounding was demonstrated but no adjustment was made in the final analyses the question should be answered as no.* †
26. *Were losses of patients to follow up taken into account?* If the numbers of patients lost to follow-up are not reported, the question should be answered as unable to determine. If the proportion lost to follow up was too small to affect the main findings, the question should be answered yes.
27. *Is there any evidence that a power calculation or reasonable equivalent was used to determine sample size?*

Appendix D: The SCED Scale

The SCED scale (below) was used to qualitatively assess the quality of the single case and small-N studies included in this review. All items are scored as *Yes* or *No*.

1. Clinical history was specified. Must include Age, Sex, Aetiology and Severity*?*
2. Target behaviours. Precise and repeatable measures that are operationally defined. Specify measure of target behaviour.
3. Design 1: 3 phases. Study must be either A-B-A or multiple baseline.
4. Design 2: Baseline (pre-treatment phase). Sufficient sampling was conducted.
5. Design 3: Treatment phase. Sufficient sampling was conducted.
6. Design 4: Data record. Raw data points were reported.
7. Observer bias: Inter-rater reliability was established for at least one measure of target behaviour.
8. Independence of assessors.
9. Statistical analysis.
10. Replication: either across subjects, therapists or settings.
11. Evidence for generalisation.

Appendix E: Relevant Sections of Submission Guidelines for the British Journal of Health Psychology

The types of paper invited are:

- *papers reporting original empirical investigations, using quantitative, qualitative or mixed methods;*
- *theoretical papers which report analyses of theories in health psychology;*
- *review papers, which should 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);*
- *methodological papers dealing with methodological issues of particular relevance to health psychology;*
- *we particularly welcome papers reporting effectiveness (for example, Randomised Controlled Trials) and process evaluations of interventions in clinical and non-clinical populations.*

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 or mixed methods 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 and clinical trials RCTs must be pre-registered. The pre-registered details should be given in the methods section but anonymised for peer review (i.e., 'the review was preregistered at [ANONYMISED]'); 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.

Submission Guidance:

Before you submit, you will need:

- *Your manuscript: this can be a single file including text, figures, and tables, or separate files – whichever you prefer. All required sections should be contained in your manuscript, including abstract, introduction, methods, results, and conclusions. Figures and tables should have legends. References may be submitted in any style or format, as long as it is consistent throughout the*

manuscript. If the manuscript, figures or tables are difficult for 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.

- *The title page of the manuscript, including a data availability statement and your co-author details with affiliations. (Why is this important? We need to keep all co-authors informed of the outcome of the peer review process.) You may like to use this template for your title page.*

Parts of the Manuscript:

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.

Acknowledgements

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.

Main Text File

As papers are double-anonymous peer reviewed, the main text file should not include any information that might identify the authors.

The main text file should be presented in the following order:

- *Title*
- *Main text*
- *References*
- *Tables and figures (each complete with title and footnotes)*
- *Appendices (if relevant)*

Supporting information should be supplied as separate files. Tables and figures can be included at the end of the main document or attached as separate files but they must be mentioned in the text.

- *The main text file should not include any information that might identify the authors. Please do not mention the authors' names or affiliations and always refer to any previous work in the third person.*
- *The journal uses British spelling; however, authors may submit using either option, as spelling of accepted papers is converted during the production process.*

References

This journal uses APA reference style; as the journal offers Free Format submission, however, this is for information only and you do not need to format the references in your article. This will instead be taken care of by the typesetter.

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.*

Figures

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.

Click here for the basic figure requirements for figures submitted with manuscripts for initial peer review, as well as the more detailed post-acceptance figure requirements.

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.

Supporting Information

Supporting information is information that is not essential to the article, but provides greater depth and background. It is hosted online and appears without editing or typesetting. It may include tables, figures, videos, datasets, etc.

Click here for Wiley's FAQs on supporting information.

Note: if data, scripts, or other artefacts used to generate the analyses presented in the paper are available via a publicly available data repository, authors should include a reference to the location of the material within their paper.

General Style Points

For guidelines on editorial style, please consult the APA Publication Manual published by the American Psychological Association. The following points provide general advice on formatting and style.

- ***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.*
- ***Effect size:*** *In normal circumstances, effect size should be incorporated.*
- ***Numbers:*** *numbers under 10 are spelt out, except for: measurements with a unit (8mmol/l); age (6 weeks old), or lists with other numbers (11 dogs, 9 cats, 4 gerbils).*

Appendix F: Service Improvement Project Ethical Approval

Favourable Ethical Approval

WoSRES West of Scotland Research Ethics Service

Ms Rebecca Roberts **West of Scotland REC 4**

Warneford Hospital
Warneford Lane
Oxford
OX3 7JX

Research Ethics
Ward 11, Dykebar Hospital
Grahamston Road
Paisley
PA2 7DE

Date 22 June 2021
Direct line 0141 314 0213
E-mail WoSREC4@ggc.scot.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

Dear Ms Roberts

Study title:	**Patient Activation in a Community Neurorehabilitation Setting: Understanding Patient and Staff Perspectives**
REC reference:	**21/WS/0054**
Protocol number:	**n/a**
IRAS project ID:	**289260**

Thank you for your letter of 21 June 2021, responding to the Research Ethics Committee's (REC) 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 Alternate Vice-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.

Good practice principles and responsibilities

The UK Policy Framework for Health and Social Care Research sets out principles of good practice in the management and conduct of health and social care research. It also outlines the responsibilities of individuals and organisations, including those related to the four elements of research transparency:

1. registering research studies
2. reporting results
3. informing participants
4. sharing study data and tissue

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

All research should be registered in a publicly accessible database and we expect all researchers, research sponsors and others to meet this fundamental best practice standard.

It is a condition of the REC favourable opinion that **all clinical trials are registered** on a publicly accessible database within six weeks of recruiting the first research participant. For this purpose, 'clinical trials' are defined as the first four project categories in IRAS project filter question 2. Failure to register a clinical trial 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-registratio n-research-project-identifiers/

If you have not already included registration details in your IRAS application form, you should notify the REC of the registration details as soon as possible.

Further guidance on registration is available at:

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

Publication of Your Research Summary

We will publish your research summary for the above study on the research summaries section of our website, together with your contact details, no earlier than three months from the date of this favourable opinion letter.

Should you wish to provide a substitute contact point, make a request to defer, or require further information, please visit: https://www.hra.nhs.uk/planning-and-improving-research/application-summaries/research-sum maries/

N.B. If your study is related to COVID-19 we will aim to publish your research summary within 3 days rather than three months.

During this public health emergency, it is vital that everyone can promptly identify all relevant research related to COVID-19 that is taking place globally. If you haven't already done so, please register your study on a public registry as soon as possible and provide the REC with the registration detail, which will be posted alongside other information relating to your project. We are also asking sponsors not to request deferral of publication of research summary for any projects relating to COVID-19. In addition, to facilitate finding and extracting studies related to COVID-19 from public databases, please enter the WHO official acronym for the coronavirus disease

(COVID-19) in the full title of your study. Approved COVID-19 studies can be found at:

https://www.hra.nhs.uk/covid-19-research/approved-covid-19-research/

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
- Reporting results

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 taking part in the study, 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*
Covering letter on headed paper [Cover letter_Patient Activation in Neurorehab_v1.0]	1.0	27 May 2021
Covering letter on headed paper [Cover letter_Patient Activation in Neurorehab_v1.0]	1.0	21 June 2021
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Schedule of Insurance]		11 August 2020
GP/consultant information sheets or letters [GP Notification letter_Patient Activation in Neurorehab_v1.0]	1.0	19 June 2021
Interview schedules or topic guides for participants [Client Interview Schedule_Patient Activation in Neurorehab_v1.0]	1.0	22 March 2021
Interview schedules or topic guides for participants [Therapist Interview Schedule_Patient Activation in Neurorehab_v1.0]	1.0	22 March 2021
IRAS Application Form [IRAS_Form_09042021]		09 April 2021
Letter from funder [Funding Letter_Patient Activation in Neurorehab]		08 February 2021
Letter from sponsor [Sponsor Letter_Patient Activation in Neurorehab]		26 March 2021
Participant consent form [Therapist Consent Form_Patient Activation in Neurorehab_v1.0]	1.1	27 May 2021
Participant consent form [Client Consent Form_Patient Activation in Neurorehab_v1.0]	1.2	19 June 2021
Participant information sheet (PIS) [Therapist Participant Information Sheet_Patient Activation in Neurorehab_v1.0]	1.1	27 May 2021
Participant information sheet (PIS) [Easy Read Client Participant Information Sheet Supplement_Patient Activation in Neurorehab_v1.0]	1.0	27 May 2021
Participant information sheet (PIS) [Client Participant Information Sheet_Patient Activation in Neurorehab_v1.0]	1.2	19 June 2021
Participant information sheet (PIS) [Easy Read Client Participant Information Sheet Supplement_Patient Activation in Neurorehab_v1.0]	1.1	21 June 2021
Research protocol or project proposal [Protocol_Patient Activation in Neurorehab_v1.0]	1.1	27 May 2021
Summary CV for Chief Investigator (CI) [CI Research CV]		22 March 2021

Summary CV for supervisor (student research) [Supervisor CV_Patient Activation in Neurorehab]		31 March 2021

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/

IRAS project ID: 289260 Please quote this number on all correspondence

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

Yours sincerely

Abibat Ackwumi

On behalf of
Dr Ken James
Chair

Enclosures: "After ethical review – guidance for researchers"

Copy to: CTRG
Lead Nation England: approvals@hra.nhs

Appendix G: Patient and Therapist Interview Schedules

Patient Interview Schedule

	Main Question	Prompts	Clarifying Questions
1.	Can you tell me a little about your experiences of living with [x], from diagnosis to today?	What were things like when first diagnosed? How have things changed over time? How have you personally responded to living with [x]?	Can you tell me more about that? What happened after that? What happened next?
2.	During your time with CNRS, did you feel that your understanding of your condition changed at all?	Was there anything you learned about: How best to manage it? Medications and treatments? The prognosis? How to prevent further problems?	
	2.a. If it increased, what do you think helped you to understand more about your condition?	Where did you get this knowledge from? What helped you to learn more about your condition?	Can you tell me more about that? *How* did that allow you to learn more about your condition?
	2.b. If it did not increase, what do you think got in the way of you understanding more about your condition?	Was there anything in particular you feel you didn't know or should have known more about? Was there anything that you think prevented you from knowing more about your condition?	Can you tell me more about that? *How* did that prevent you from knowing more about your condition?
3.	During your time with CNRS, did you feel able to put what you learned about your condition into action?	Was there anything that enabled you to: Actually carry out the rehab programme/ exercises day to day? Put into practice recommended lifestyle changes? Adhere to a medication routine?	
	3.a. If yes, what do you think helped with this?	Was there anything that enabled this? Was there anything that helped you to put what you knew about your condition into action?	Can you tell me more about that? *How* did that help you to put the knowledge into action?
	3.b. If no, what do you think prevented this?	Was there anything that you think prevented you from being able to do this? Was there anything that you think prevented you from putting changes into action?	Can you tell me more about that? *How* did that prevent you from putting the knowledge into action?

4.	During your time with CNRS, did you feel that you became any more confident and equipped to (live with [x]/ live life after [x])?	Was there anything that enabled you to: Better manage your condition day to day? Feel more confident in working out solutions to problems relating to your condition? Feel more confident in seeking the correct support from health professionals if needed?	
	4.a. If yes, what do you think helped with this?	Was there anything that enabled this? Was there anything that helped you to become more confident in doing this? Was there anything that enabled you to manage things more independently?	Can you tell me more about that? *How* did that help you to become more confident/ equipped?
	4.b. If no, what do you think prevented this?	Was there anything that you think prevented you from being able to do this? Was there anything that you think prevented you from becoming more confident? Was there anything that stopped you from being able to manage things more independently?	Can you tell me more about that? *How* did that prevent you from becoming more confident/ equipped?
5.	Was there anything that the service did, in particular, that was helpful (in terms of becoming more informed, equipped and confident)?	Was there anything that the therapist did that was helpful? Did you have any involvement with anyone else from the service?	Can you remember any specific examples of something the professional did that you found helpful? Was there a link between this and how confident and equipped you felt to (live with [x]/ live life after [x])?
6.	Was there anything that the service did that was less helpful (in terms of becoming more informed, equipped and confident)?	Was there anything you felt should have been done differently? Was there any more that you felt could have been done?	Was there a link between this and how confident and equipped you felt to (live with [x]/ live life after [x])?
7.	Was there anything in your personal or home life that affected how confident and equipped you felt to (live with [x]/ live life after [x])?	How well supported did you feel at home? How were the people around you coping around the time you were with CNRS? Did you have any setbacks during your time with CNRS?	Can you say a bit more about what that was and how it affected you?

8.	Can you tell me about how you generally manage difficulties and face challenges that come up your life, either now or in the past?	How have you approached difficult experiences in the past? How confident have you felt dealing with difficulties in your life in the past? How do you generally go about solving problems that have come up for you?	Can you give an example of this?
9.	Is there any advice that you would give to others who are just starting out on their rehabilitation journey?	Was there anything you did that really worked? Was there anything that you found particularly helpful? Was there anything, in hindsight, that you would've liked to have done differently?	Was there a link between this and how confident and equipped you felt to (live with [x]/ live life after [x])?

N.B. The wording of questions containing square brackets was adapted based on the person's specific condition. This was to account for the differences between stroke recovery and ongoing conditions e.g. 'living with Parkinson's' or 'living life after stroke'.

Therapist Interview Schedule

	Main Question	Prompts	Clarifying Questions
1.	Can you tell me a little about your experience of working with [client name]?	Can you give a general overview? What sort of work did you do together? What was your therapeutic relationship like?	Can you tell me more about that? Can you give an example of some of the work you did together?
2.	What were the signs that indicated to you that this person was low in activation?	Were there any signs that this person lacked knowledge, skill and confidence in managing their condition? What was their attitude towards neurorehabilitation?	Can you give me an example of something you noticed?
3.	What were the signs that indicated to you that this person became more activated during the time you worked with them?	Were there any signs that this person gained knowledge, skill and confidence in managing their condition? Were there any changes in this person's attitude/ approach during treatment? Did you use PAM data or any other type of measure?	Can you give me an example of something your noticed?

4.	What strategies did you use to motivate and encourage this person?	Did you use any specific techniques? Was there anything in particular you did with this client that went 'above and beyond' your usual practice?	Can you give an example of this? Was there any link between this and their level of activation?
5.	Was there anything else that you think you or the wider service did that may have helped them to become more activated?	How did you work with other members of the team? Did you link in with any other services?	Can you tell me a bit more about this? Was there any link between this and their level of activation?
6.	Was there anything you noticed that might have got in the way of them becoming more activated?	Anything you did that worked less well? Anything that caused them to become less engaged?	Can you give a specific example of this?
7.	Was there anything about the client or their wider system that you feel was important in terms of their activation level?	Anything that happened during the course of treatment? Any ways in which the client was supported to become activated outside of CNRS?	Can you be specific about what this was?
8.	Looking back now, is there anything you would have done differently?	For example, if you had more resources or time? Anything you would do more or less of?	Can you say a little more about this?
9.	Is there anything else that we haven't discussed already, that you find generally helps the people you work with to become more equipped and confident to manage their health condition?	Any techniques or strategies you usually use? Anything you do regularly that you find is helpful? Anything you think the service should be encouraging more of?	Can you give a specific example of when you've done this and how it helped?

Appendix H: Themes, Subthemes and Supporting Quotes

Interview Group	Theme	Subtheme	Participant Quotes
Therapists	Approaching difficulties	Denial to acceptance	‘He wanted I suppose… he just wanted… life back… back to normal.’ (T1) ‘I think her relationship with her disease changed quite a lot. Um so she was much more… sort of accepting of it by the end.’ (T2) ‘Guiding them through… uh… kind of got getting back to be themselves, but in a… with a different… different abilities…’ (T3) ‘The conversation was all about moving forward rather than reflecting on the past.’ (T4)
		Acknowledging fatigue	‘But I think it was just an acknowledgement… that there are… hidden symptoms... and and seeing things that other people might perceive you as lazy or unmotivated or… um… depressed... But it it… it can... it can be… that you’re just mentally and physically fatigued…’ (T1) ‘And we were trying to use compassion to be able to kind of be kind to yourself. Ask for help when you need it. Take rests when you need it.’ (T2)
		Addressing emotional difficulties	‘I think there was a lot of grief and loss and… frustration and you know... felt useless.’ (T1) ‘And for someone that's already really, really stuck in this cloud of shame around their disease, that sense that you have to do this to get better or you're going to get worse. It's just intolerable and it makes it <u>really, really</u> difficult to then engage in the exercises.’ (T2) ‘What I allowed them to do was just process all of their thoughts… but put it within a context of how do I move forward.’ (T4)
	Patient-led Collaboration	Patient taking the lead	‘He identified the tasks…. he wrote them down.’ (T1) ‘So it was <u>very, very</u> guided by her.’ (T2) ‘She needed to have that… autonomy over deciding. I'm ready now I can do this by myself.’ (T2) ‘He would say yes or no... you know we would negotiate.’ (T3) ‘And he came up with a plan that I wasn't expecting… It was all about what was right for him because… you know that that that wasn't the plan that <u>I</u> would have generated for him.’ (T4)

Interview Group	Theme	Subtheme	Participant Quotes
		Flexible and realistic therapist	‘But you know it worked for him… and I… and we just reinforced that... if that works for you and that's working for your wife and your two are both happy with it... then continue…’ (T1) ‘We thought about putting post-it notes around the house… or… but he… he didn't want any of that.’ (T1) ‘So you're not turning 180… because that's that's too too much difference… there are reasons why people are going the direction they're heading.’ (P4) ‘From a therapist point of view, using a fix it approach that was you know, no question to me the right pathway. (pause) Actually, that really was not what he wanted to do at all.’ (T4)
		Collaboration (patient, therapist and team)	‘And I did a lot of liaison with the physio at the time.’ (T2) ‘It's a partnership. Yes, I'm I'm suggesting what to do, but actually, who is doing it… it’s not me.’ (T3) ‘I think sometimes with those patients when you need to put a lot of effort at the beginning to buy their trust... I sometimes find that there is a very unique relationship.’ (T3) ‘They said right at the end… thank you so much for being… what did they say… something like an agony aunt or… yeah… which is essentially thank you so much for listening.’ (T4)
	Finding Direction and Confidence	Initial stuckness	‘He just didn't know how to progress forward.’ (T1) ‘So she had deteriorated a lot. But there was this sense of like I can't go out and do anything.’ (T2) ‘He kind of couldn't find his er… way round... how could he do things he enjoyed and almost become more passive.’ (T3) ‘They looked overwhelmed and they talked about feeling exhausted.’ (T4)
		Purpose and values	‘They [psychology] did um beliefs and values… we did a joint [session]… and that really got us going in him being able to identify the three tasks… that he's going to do.’ (T1) ‘But it started with “I'll try anything to make this better so I can get back to work” and then by the end it was very much like…”I'll try anything to give me a better quality of life”.’ (T2) ‘I would explain why.’ (T3) ‘So we said the current route that you’re on… are you moving closer to your values or do you need to make a choice?’ (T4)

Interview Group	Theme	Subtheme	Participant Quotes
		Building confidence	‘But then when he got to the point where he was like… right… I arrived and… he said “I'm showing you” … “Come with me. This is what I've been doing”.’ (T1) ‘It [the therapy break] gave her a month to see that she could manage this by herself and she didn't need to check in with me all the time.’ (T2) ‘You need to choose well to give them feeling of the winning… uh… to actually give err… so they need to be able to do it, and they need to be able to improve…’ (T3)
Patients	Setbacks and frustrations	Difficult emotions	‘I felt frustrated.’ (P1) ‘These days some of the smaller tasks make me anxious.’ (P5) ‘I get depressed with it you know.’ (P7) ‘I just felt guilty that I don’t do more.’ (P8)
		Negative experience of services	‘Nobody… checks on you… to see how you’re doing… don’t even bother to contact you.’ (P1) ‘We knew for a long time but I couldn’t get a diagnosis.’ (P4) ‘No one asks what’s happened… you just lose confidence in everybody.’ (P7) ‘[previous physio] was a complete and utter waste of time.’ (P8)
	Barriers to Change	Black and white thinking	‘I try to ignore the fact I’ve got it if I possibly can.’ (P6) ‘It’s basically too late.’ (P8)
		Cognitive Difficulties	‘She actually did suggest would you like to go to the memory clinic.’ (P6) ‘It has affected my memory.’ (P7) ‘I struggle to find the right word.’ (P8)
		Isolation	‘I think they ought to be in touch with you a bit more… see how you’re getting on and everything.’ (P7) ‘Nobody’s listening… I’ve never been able to talk to anybody about it.’ (P8)
	Wider social support	Mutual support	‘Its very helpful that we've got a group [PD] that meet together.’ (P2) ‘We’ve all got the same thing so we all sort of share our problems and things.’ (P3) ‘More than anything was talking to other people with the condition… I didn’t really know anybody that had it.’ (P6) ‘Its actually sort of knowing that I’m not alone and that that’s just what happens when you have a stroke.’ (P8)

Interview Group	Theme	Subtheme	Participant Quotes
		Family and friends support	‘It is important… as far as I’m concerned… that the rest of the family understands what it means for that person.’ (P1) ‘Well I I think he [husband] knows when I need some support and he’s ready to give me that when I when I need it.’ (P2) ‘I've got my old work colleagues that I go out with and… they know me really well and they’ve seen me through it all.’ (P3) ‘Yes because the exercise group which I went to and the Nordic walking group are extremely supportive… and it was a lovely group.’ (P4) ‘Support from the family is good.’ (P5) ‘Me son comes to see me everyday.’ (P7)
	Positive view of support	Supportive therapist	‘She gave me confidence that I can do it. “Come on, you can do that”.’ (P1) ‘Any time I’d seen her “You are making progress”, “You’re working better” or “You’re doing this better”… which is… encouraging you know and you’d have her you know… “I’ll see you in a week’s time” which is encouraging when someone says… they’re making a… they wanna see you.’ (P1) ‘I went back to be reassessed to see how much progress I'd made and that there was actually progress and er some of the areas… it was so affirming.’ (P2) ‘She [the therapist] boosted my confidence.’ (P3) ‘They were very understanding of if I hadn’t done enough… they cut me some slack.’ (P4)
		Collaboration (patient, therapist, team and family)	‘Something about doing it together… it’s far easier than trying to do things from a book.’ (P1) ‘Being given information and being asked if there's any questions. Erm and also if I’m going for an appointment I try to prepare and think about what I want to talk about.’ (P2) ‘I found that everyone there was wonderful… Even from like the receptionist and everything.’ (P3) ‘A sister in law who came with me on one occasion to a relatives meeting… and a friend who came with me on another.’ (P4)
	Signs of Activation	Self-efficacy	‘Oh well I’m determined because I’m a vegetable guy erm and I was determined I was going to get back and get on my vegetables again.’ (P1) ‘You know… if you don’t do these things you’re just robbing yourself of it…’ (P3) ‘Looking for help and researching what it is.’ (P4) ‘You know I wanna hang on to that because I can see that you could lapse back to watching the telly and doing nothing at all.’ (P5)

Interview Group	Theme	Subtheme	Participant Quotes
		Language of acceptance	‘But I don’t feel sorry for me I just get on.’ (P1) ‘I've got to realise my limitations.’ (P2) ‘You know… I had to be realistic.’ (P3) ‘Not struggling against it… accepting it… there’s no point wasting energy.’ (P4)
		Making adjustments and adaptations	‘I basically say well I can’t do it then I can’t do it so I’ll do something by a different method instead.’ (P1) ‘Each day I have priority tasks that I need to do and then make sure that… either I get those done first or if they've got to be done later on I’ll give myself some rest time. So I plan out my time.’ (P2) ‘But you know I’ve got round that now and I can do most things.’ (P5)
		Seeing progress and purpose	‘I still do some of the exercises that she give me one after the other because it gives me… keeps my body supple… it’s very important for my future I’ve been told.’ (P1) ‘I always try and be upbeat to people and tell them… you know that hopefully things won’t be as bad as they think they will.’ (P3) ‘I was very pleased with the progress.’ (P4)

Appendix I: Lay Summary

Why we did this research

This research wanted to help people with neurological conditions to get the most out of their rehabilitation at the Community Neurorehabilitation Service in Buckinghamshire (CNRS). Previous research has shown that people benefit most from therapeutic interventions when they take an active role in the management of their own health condition (i.e. are 'activated'). This research sought to understand whether current practices at CNRS help people to become activated and, what more could be done to support patients with this.

What we did

We compared the questionnaire data of 77 patients before and after rehabilitation with CNRS. We also spoke to four members of the therapy team and eight former patients to see what they thought helped or hindered activation.

What we found

Questionnaire data showed that most people who have rehab with CNRS become activated and that this was not affected by factors such as age, diagnosis, wait time or social support.

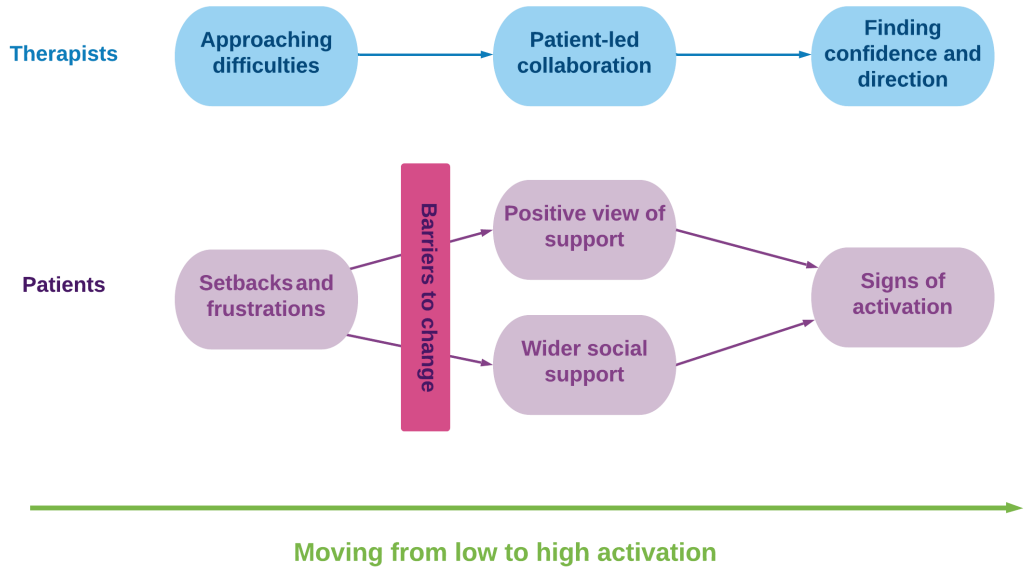
Interviews with therapists showed the importance of helping someone to approach their difficulties (e.g. helping them to acknowledge fatigue or supporting with emotional difficulties). Therapists also emphasised that they needed to be flexible, allow the patient to take a lead, and help the patient to find confidence and direction.

Interviews with patients showed the importance of wider social support including from others with the same condition. They expressed that the therapist needed to be supportive and

collaborative, and that it helped to see the purpose of the rehabilitation i.e. how will this intervention help me with what's important to me.

Interviews with patients also showed that negative past experiences of services and difficult emotions were important to consider. Feelings of hopelessness or ignoring the problem, isolation and cognitive difficulties seemed to make it harder for people to become activated.

The important themes of these discussions are shown in the diagram below:



What changed

CNRS implemented a number of changes based on the findings of this research.

1. Reviewing the initial assessment to check that all therapists take time to hear a patient's journey and support them with the emotional side of living with a neurological condition.
2. Ensuring all patients are asked about their past experiences of rehabilitation so that CNRS can avoid repeating previous frustrations.

3. Making sure to connect people with local support groups e.g. Stroke Association/ Parkinson's UK.
4. Developing a specific intervention for people who struggle with actively managing their health condition.

Conclusions

Neurorehabilitation can help people to become more activated and changeable factors such as emotions, social support and seeing progress and purpose seem to be more important than fixed factors such as age and diagnosis. There is lots that neurorehabilitation services can do to help patients with this, including making sure people are listened to and understood and linking people in with others with the same condition.

Appendix J: Relevant Sections of Submission Guidelines for the Journal of Clinical Rehabilitation

Article Types:

The journal publishes original papers, systematic reviews, Rehabilitation in Practice and Rehabilitation in Theory articles and correspondence relating to published papers. Other article types should be discussed with the editor before submission. Classification is undertaken by the Editor, all articles should be submitted as original articles.

Summary of Manuscript Structure:

- *A title page with names and contact details for all authors;*
- *A **structured** abstract of **no more than 250 words** (the website checks this);*
- *The text (usually Introduction, Methods, Results, Discussion);*
- *Clinical Messages (2-4 bullet points, 50 words or less);*
- *Acknowledgements, author contributions, competing interests and funding support;*
- *References (Vancouver style);*
- *Tables, each starting on a new page;*
- *Figures, each starting on a new page;*
- *Appendix (if any).*

Appendix K: Theory Driven Research Project Ethical Approval and Amendment

Favourable opinion for substantial amendment

Health Research Authority

South Central - Oxford C Research Ethics Committee
Health Research Authority (Bristol)
Ground Floor, Temple Quay House, 2 The Square
BS1 6PN

Tel: 0207 104 8379

Please note: This is the favourable opinion of the REC only and does not allow the amendment to be implemented at NHS sites in England until the outcome of the HRA assessment has been confirmed.

25 May 2021

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**
Amendment number: **Substantial Amendment 1**
Amendment date: **29 March 2021**
IRAS project ID: **248483**

The above amendment was reviewed by the Sub-Committee in correspondence.

Ethical opinion

The members of the Committee taking part in the review gave a favourable ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

Approved documents

The documents reviewed and approved at the meeting were:

Document	*Version*	*Date*
Completed Amendment Tool [248843_Substantial Amendment]	1	29 March 2021
Research protocol or project proposal [Protocol_tracked changes]	0.5	22 March 2021
Research protocol or project proposal [Clean protocol]	0.5	22 March 2021
Validated questionnaire [Illness Perception questionnaire]	*received on 17/05/2021	

Membership of the Committee

The members of the Committee who took part in the review are listed on the attached sheet.

Working with NHS Care Organisations

Sponsors should ensure that they notify the R&D office for the relevant NHS care organisation of this amendment in line with the terms detailed in the categorisation email issued by the lead nation for the study.

Amendments related to COVID-19

We will update your research summary for the above study on the research summaries section of our website. During this public health emergency, it is vital that everyone can promptly identify all relevant research related to COVID-19 that is taking place globally. If you have not already done so, please register your study on a public registry as soon as possible and provide the HRA with the registration detail, which will be posted alongside other information relating to your project.

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.

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-andimproving-research/learning/

IRAS Project ID - 248483:	**Please quote this number on all correspondence**

Yours sincerely



Dr Lee Potiphar Chair

E-mail: oxfordc.rec@hra.nhs.uk

Enclosures: *List of names and professions of members who took part in the review*

Copy to: *Miss Margaret Moore*

South Central - Oxford C Research Ethics Committee

Attendance at Sub-Committee of the REC in correspondence

Committee Members:

Name	*Profession*	*Present*	*Notes*
Dr Linda Cartwright	Retired Consultant Epidemiologist	Yes	Chair of Sub-Committee meeting
Mr Barjinder Sahota	Solicitor Advocate	Yes	

Also in attendance:

Name	*Position (or reason for attending)*
Miss Theodora Chortara	Approvals Administrator

Appendix L: Theory Driven Research Project Supplementary Analysis

Table L1.

A Priori and Post-hoc Power Calculations for Pearson's Correlation Analyses Conducted in the Main Text

Variables	Correlation (*r*)		Statistical Power	
	Predicted	Actual	Predicted	Actual
MoCA score and OCS-Plus impairments	-.3	-.77	85%	100%
OCS-Plus impairments and Barthel score	-.3	-.35	85%	87%

Table L2.

A Priori and Post-hoc Power Calculations for Multivariate Linear Regression Analyses Conducted in the Main Text

Variables	Effect Size (f^2)		Statistical Power	
	Predicted	**Actual**	**Predicted**	**Actual**
IVs – OCS-Plus impairments, Barthel Score, mental health history DV- PHQ-9	0.15	0.24	80%	95%
IVs – MoCA score, Barthel Score, mental health history DV- PHQ-9	0.15	0.27	80%	90%
IVs – OCS-Plus impairments, Barthel Score, mental health history DV- GAD-7	0.15	0.28	80%	97%
IVs – OCS-Plus impairments, Barthel Score, mental health history, PHQ-9 DV- GAD-7	0.35	1.11	99%	100%
IVs – MoCA score, Barthel Score, mental health history DV- GAD-7	0.15	0.18	80%	75%
IVs – MoCA score, Barthel Score, mental health history, PHQ-9 DV- GAD-7	0.35	1.48	99%	100%
IVs – OCS-Plus impairments, Barthel Score, mental health history DV- Perceived threat (SIPQ)	0.15	0.29	80%	98%
IVs – Perceived threat, OCS-Plus impairments, Barthel Score, mental health history DV – PHQ-9	0.35	1.82	99%	100%
IVs – OCS-Plus impairments, Barthel Score, mental health history DV- Perceived control (SIPQ)	0.15	0.09	80%	73%
IVs – Perceived control, OCS-Plus impairments, Barthel Score, mental health history DV – PHQ-9	0.35	0.44	99%	100%
IVs – OCS-Plus impairments, Barthel Score, mental health history DV- Illness coherence (SIPQ)	0.15	0.15	80%	80%
IVs – Illness coherence, OCS-Plus impairments, Barthel Score, mental health history DV – PHQ-9	0.35	0.32	99%	98%

Table L3.

Correlations Between Primary Outcome Variables

	OCS-Plus	**PHQ-9**	**GAD-7**
Time since stroke (days)	.04	.18	.08
Stroke Type	.07	.02	.10
Stroke Side	.07	-.08	-.02
Stroke Severity (NIHSS score)	.19	.19	.29*
Degree of physical difficulty (Barthel)	-.35**	-.04	-.01
Past history of stroke	.07	.06	-.13
Reported history of mood difficulties	.08	.30**	.29*
Current depression (PHQ-9)	.32**	-	.70**
Current anxiety (GAD-7)	.36**	.70**	-

*p<.05, **p<.01.

Appendix M: Questions administered from the Stroke Illness Perception Questionnaire

YOUR VIEWS ABOUT YOUR HEALTH NOW

Listed below are a number of difficulties that you may or may not have experienced **since your stroke.** Please indicate by **circling *Yes* or *No***, whether you have experienced any of these difficulties since your stroke, and whether you believe that these difficulties are related to your stroke.

	Have you experienced this difficulty *since your stroke*?			**If YES, is this difficulty *related to your stroke?***	
Pain (not headaches)	No	Yes	→	Yes	No
Difficulty swallowing	No	Yes	→	Yes	No
Weight loss	No	Yes	→	Yes	No
Extreme tiredness	No	Yes	→	Yes	No
Stiffness in muscles	No	Yes	→	Yes	No
Headaches	No	Yes	→	Yes	No
Bladder problems	No	Yes	→	Yes	No
Bowel problems	No	Yes	→	Yes	No
Sleep difficulties	No	Yes	→	Yes	No
Dizziness	No	Yes	→	Yes	No
Poor balance	No	Yes	→	Yes	No
Weakness or paralysis in arm or leg	No	Yes	→	Yes	No
Difficulty seeing things	No	Yes	→	Yes	No
Difficulty speaking	No	Yes	→	Yes	No
Feeling forgetful	No	Yes	→	Yes	No
Difficulty writing things down	No	Yes	→	Yes	No
Crying or laughing inappropriately	No	Yes	→	Yes	No
Poor concentration	No	Yes	→	Yes	No
Difficulty reading what things say	No	Yes	→	Yes	No

	Have you experienced this difficulty *since your stroke*?			If YES, is this difficulty *related to your stroke?*	
What I'm like as a person has changed	No	Yes	→	Yes	No
Feeling low	No	Yes	→	Yes	No
Clumsiness	No	Yes	→	Yes	No
Hearing difficulties	No	Yes	→	Yes	No
Difficulty walking or getting around	No	Yes	→	Yes	No
Confusion	No	Yes	→	Yes	No
Tingling or numbness	No	Yes	→	Yes	No
Falling over	No	Yes	→	Yes	No
Stiff joints	No	Yes	→	Yes	No

If you have experienced any other difficulties recently which you believe are related to your stroke, please write them in the space below.

I have experienced _____

YOUR VIEWS ABOUT YOUR STROKE

We are interested in your own personal views of how you see your stroke. Please indicate how much you agree or disagree with the following statements about the effects of your stroke by circling the appropriate box. Some of the questions may sound similar. But it would be really helpful if you could try to decide on an answer for all of them. **Please answer all of the questions**

The following questions are about how long that the **effects of your stroke are likely to last**.

1. The effects of my stroke will last for a short time

Strongly Agree	**Agree**	**Don't know**	**Disagree**	**Strongly Disagree**

2. The effects of my stroke are more likely to be permanent than temporary

Strongly Agree	**Agree**	**Don't know**	**Disagree**	**Strongly Disagree**

3. The effects of my stroke will last for a long time

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

4. The effects of my stroke will pass quickly

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

5. I will have the difficulties from my stroke for the rest of my life

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

6. The effects of my stroke will improve in time

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

These questions are about the way your stroke has **affected your life**, including your relationships with others.

7. My stroke is a serious condition.

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

8. My stroke has a major impact on my life

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

9. My stroke does not have much effect on my life

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

10. Memory problems since my stroke are affecting my life

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

11. My stroke strongly affects the way others see me

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

12. Since my stroke I fear becoming a burden on others

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

13. My stroke causes difficulties for those who are close to me

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

14. My stroke has strongly affected how I see myself

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

15. My stroke has seriously affected how much money I have

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

16. My stroke has badly affected my relationship with my family

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

17. Emotional problems since my stroke are affecting my life

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

These questions are about how much you feel **able to manage the effects of your stroke.** For example, how much you can influence your ability to get better.

18. I need to avoid doing too much as this may cause another stroke

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

19. What I do can determine whether the effects of my stroke get better or worse

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

20. There is a lot which I can do to manage the effects of my stroke

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

21. I cannot prevent another stroke from occurring

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

22. Nothing I do will affect my recovery from stroke

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

23. The course of my recovery from stroke depends on me

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

24. I have the power to influence my recovery from stroke

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

25. My actions will have no effect on the outcome of my recovery from stroke

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

26. I am confident that I can manage my recovery well

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

The following questions are about your **treatment for stroke**. This includes any treatment that you may have had during your hospital stay, as well as things that you may be doing since returning home.

For example:

- Any tablets that you are taking for your stroke (e.g. tablets to lower blood pressure).
- Any therapy that you might be involved with (e.g., seeing a physiotherapist for exercises, and an occupational therapist for help with carrying out your daily activities). This also includes things that you are doing on your own.
- Any lifestyle advice that you may have had (e.g., stopping smoking, exercising more, and eating healthy meals).

27. There is very little that can be done to improve the difficulties from my stroke

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

28. My treatment will be effective in preventing another stroke from happening

Strongly Agree	**Agree**	**Don't know**	**Disagree**	**Strongly Disagree**

29. The negative effects of my stroke can be prevented (avoided) by my treatment

Strongly Agree	**Agree**	**Don't know**	**Disagree**	**Strongly Disagree**

30. My treatment can control the difficulties from my stroke

Strongly Agree	**Agree**	**Don't know**	**Disagree**	**Strongly Disagree**

31. There is nothing which can help my recovery from stroke

Strongly Agree	**Agree**	**Don't know**	**Disagree**	**Strongly Disagree**

These questions are about how much you **understand what has happened to you** (e.g., whether you are clear about the things that you can do to help yourself to feel better).

32. The effects of my stroke are confusing to me

Strongly Agree	**Agree**	**Don't know**	**Disagree**	**Strongly Disagree**

33. My stroke is a mystery to me

Strongly Agree	**Agree**	**Don't know**	**Disagree**	**Strongly Disagree**

34. I don't understand my recovery from stroke

Strongly Agree	**Agree**	**Don't know**	**Disagree**	**Strongly Disagree**

35. The effects of my stroke don't make any sense to me

Strongly Agree	**Agree**	**Don't know**	**Disagree**	**Strongly Disagree**

36. I have a clear picture or understanding of my stroke

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

These questions are about whether you feel that the effects of your stroke **change on a day to day basis** (e.g., whether you feel better on some days than on others).

37. The effects of my stroke are very unpredictable

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

38. The difficulties from my stroke change a great deal from day to day

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

39. I have good days with few or no difficulties and bad days, when I have a lot of difficulties

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

40. I go through cycles in which the effects of my stroke get better and worse

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

These questions are about how your **feelings and emotions** have changed since your stroke.

41. I do not feel in control of my emotions

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

42. I get embarrassed by the way I am since my stroke

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

43. My stroke does not worry me

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

44. Since my stroke I get depressed

Strongly Agree	Agree	Don't know	Disagree	Strongly Disagree

45. When I think about my stroke I get upset

Strongly Agree	**Agree**	**Don't know**	**Disagree**	**Strongly Disagree**

46. The effects of my stroke make me feel anxious

Strongly Agree	**Agree**	**Don't know**	**Disagree**	**Strongly Disagree**

47. My stroke makes me feel afraid

Strongly Agree	**Agree**	**Don't know**	**Disagree**	**Strongly Disagree**

48. My stroke makes me feel angry

Strongly Agree	**Agree**	**Don't know**	**Disagree**	**Strongly Disagree**

49. I feel lost since my stroke

Strongly Agree	**Agree**	**Don't know**	**Disagree**	**Strongly Disagree**

50. I have lost confidence in myself since my stroke

Strongly Agree	**Agree**	**Don't know**	**Disagree**	**Strongly Disagree**

Appendix N: Publication in the Review Journal of Autism and Developmental Disorders

Review Journal of Autism and Developmental Disorders
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REVIEW PAPER

Are Extended Reality Interventions Effective in Helping Autistic Children to Enhance Their Social Skills? A Systematic Review

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Abstract
Autistic children's social skills do not always align with those of their neurotypical peers and research suggests that this can negatively impact quality of life. This review aimed to assess the effectiveness of extended reality (XR) interventions in helping autistic children to enhance their social skills. Five electronic databases were systematically searched and seventeen studies were identified. The majority targeted social-emotional reciprocity and were of relatively low quality. There was insufficient evidence to determine whether effects were generalisable, sustained or important to autistic people. Research in this field is in its infancy and evidence of effectiveness should be viewed with caution. Future studies should aim for high-quality, theory-driven research, and involve autistic people to ensure meaningful outcomes.
PROSPERO ID: CRD42021229442

Keywords Autism · Social Skills · Augmented reality · Virtual reality · Assistive technology

Introduction

Autism and Social Differences

Effective social communication depends on a range of *social skills*, a term which is complex and has been variably defined across the literature, but is broadly considered to encompass verbal and non-verbal behaviours that enable positive social interactions (Gresham & Elliott, 1984). This multi-faceted concept includes innate skills, such as eye tracking and a preference for faces, as well as more complex, learned behaviours, such as adjusting behaviour to suit particular social contexts (Spence, 2003). An exhaustive list of skills considered to be *social* is difficult to establish, due to the number of cognitive and behavioural processes drawn upon in any given social situation.

Autistic[1] children's social skills do not always align with those of neurotypical people. Their skills are different in terms of social-emotional reciprocity (e.g. social approach and initiation of interactions), nonverbal communicative behaviours (e.g. eye contact, and use of facial expressions), and the development, maintenance and understanding of relationships (e.g. reduced interest in peers and difficulties making friends); Diagnostic and Statistical Manual of Mental Disorders, 5th ed.; DSM-5; American Psychiatric Association [APA], (2013). These differences manifest differently for each individual (Happé et al., 2006) and can have negative secondary effects on wellbeing, learning and relationships. For example, social skills which do not align with neurotypical people have been shown to predict social anxiety (Bellini, 2006) and are associated with social rejection from peers (Ochs et al., 2001). This is not through lack of interest in

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[1] Identity first language is used within this article as this is the preferred language of many adults on the autism spectrum, i.e. 'autistic individual' rather than 'individual with autism' (Kenny et al., 2016). However, there is a lack of information about the preferred language of autistic children.

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social contact (Jaswal & Akhtar, 2019); autistic young people often express a desire for social interaction and report being more lonely than their neurotypical peers (Bauminger & Kasari, 2000). Furthermore, social differences in autistic people can have a negative effect on academic and occupational outcomes (Howlin, 2003; Welsh et al., 2001).

Theories of Social Difference in Autism

A number of theories might explain the social differences observed in autism. One theory proposes that autistic people can find it harder to make predictions about others' mental states (theory of mind) and so may find it difficult to encode socially relevant information (Baron-Cohen et al., 1985). Alternatively, it may be that difficulty understanding others' interpretations of the world is due to an underlying difference in executive functioning, such as rule learning (J. Russell, 1997). This can account for the finding that some autistic children struggle with theory of mind style tasks, even when the social element is removed (Russell et al., 1991). In particular, predictive processing might be different in autism. Pellicano and Burr (2012) propose that autistic people perceive the world more accurately, as they are less biased by prior experience when making predictions. At times this may be a strength, but could make some elements of social interaction more difficult, such as predicting when another person will speak (Stark et al., 2021). Another cognitive theory of autism is 'weak central coherence' (Frith, 1989) which suggests that autistic people have a preference for local processing rather than striving for overall meaning. This may lead to differences in the interpretation of social behaviours. However, more recent evidence suggests that this theory sits alongside social differences, rather than explains them (Happé & Frith, 2006). These cognitive theories are in line with evidence that social desire is not affected by autism (Happé & Frith, 1996).

In contrast, social motivation theory proposes that autistic people have differences in the psychological dispositions and biological mechanisms that bias neurotypical individuals to orient towards social stimuli, seek and derive pleasure from social interactions, and work to maintain social bonds (Chevallier et al., 2012). However, this contrasts with many accounts of autistic people, overlooks the role of other people's perceptions and responses, and ignores the many other explanations of social differences in autism (Jaswal & Akhtar, 2019). Furthermore, this theory has led to differences such as limited eye contact being prioritised as intervention targets, despite evidence for the positive effects of gaze aversion on concentration and regulation of emotions (Robledo et al., 2012).

Social Skills Interventions

A number of interventions have sought to teach social skills to autistic children, due to the secondary benefits on wellbeing and quality of life shown by previous research (McConnell, 2002). A range of outcomes have been targeted, such as turn taking, initiating interaction, recognising emotions and understanding theory of mind (Begeer et al., 2011; Matson et al., 2007; Rieth et al., 2014). Well-established interventions include Social Stories™ (Karkhaneh et al., 2010), video modelling (Sng et al., 2014), pivotal response training (Koegel et al., 2016), social skills groups (Reichow et al., 2012) and peer-mediated behavioural interventions (Laushey & Heflin, 2000). There is increasing evidence that, at least in some contexts, these interventions can help autistic children to utilise neurotypical social skills but the duration, generalisability and precise mechanism of change remains unclear (McConnell, 2002). Furthermore, little acknowledgement has been given to the fact that it may be effortful, and perhaps emotionally draining for autistic people to operationalise these skills.

When considering social skills interventions, it is important to acknowledge that social differences in autism do not necessarily equate to a 'difficulty'. Many argue that difficulties arise due to a lack of accommodation of autistic differences in a neurotypical society (Brownlow, 2010), and emerging research is suggestive of a 'double empathy problem', whereby communication breakdowns between autistic and non-autistic people are a two-way issue resulting from disjuncture in reciprocity between the two differently disposed social actors (Edey et al., 2016; Milton, 2012; Sheppard et al., 2016). Social skills training can therefore perpetuate the idea that autistic people need to adapt, without consideration of the role of the contexts in which autistic people live, learn and work in understanding, supporting and responding to autistic communication. However, many autistic people already find ways to overcome social challenges and seek support in doing this (Cresswell et al., 2019). Social skills interventions are one way in which services can support people to overcome these challenges, particularly in circumstances where it is not possible to adapt the environment e.g. public places.

Extended Reality Technologies

Over the last three decades, social skills interventions have begun to capitalise on the potential advantages of digital technologies. Computerised approaches can support social skills in situ (Vygotsky & Luria, 1994) or improve traditional training approaches, due to the attentional and motivational advantages of interactive technologies (Golan & Baron-Cohen, 2006). More recently, these advantages have been exploited using extended reality (XR), i.e. the merging of physical and virtual realities (Riva et al., 2016). This encompasses augmented reality (AR), in which virtual information is overlaid onto the real world, virtual reality (VR), in which users are fully immersed in a simulated environment and

mixed reality (MR), in which digital and real-world objects interact in real-time. By merging physical and virtual realities, XR could reduce the salience of potentially distressing stimuli, making interactions less overwhelming for some autistic people, thus providing an effective space to experiment with social skills (Thye et al., 2018). In virtual social settings, there is no risk of embarrassment in front of one's peers, yet the degree of realism may increase the likelihood of generalisability (Strickland et al., 1996).

Extended reality interventions have been successfully implemented in a number of therapeutic contexts including stroke rehabilitation (Laver et al., 2017) and exposure therapies for anxiety disorders (Bouchard et al., 2017; Vincelli et al., 2003). Applications for autistic people are also emerging, although the majority of interventions to date have used virtual learning environments (VLEs), a non-immersive technology in which users interact with simulated social situations using a desktop computer (e.g. Didehbani et al., 2016). More immersive technologies tend to require a head-mounted display (HMD) or Smartglasses, computer glasses that change what the wearer sees. Despite the potential challenges for those with tactile hypersensitivities, they have been shown to be well-tolerated, enjoyable and engaging in this population (Keshav et al., 2017; Newbutt et al., 2016), and effective in teaching other skills, such as taking public transport (Simoes et al., 2018).

Aims of this Review

Previous reviews have shown that XR interventions are feasible and sometimes beneficial for teaching autistic children a range of skills, although recognise that the quality of the literature is varied (e.g. Berenguer et al., 2020; Mesa-Gresa et al., 2018). However, no review to date has investigated the specific potential of XR for social skills. Furthermore, previous reviews of VR interventions have tended to include non-immersive technologies, such as VLEs. This review takes a focused approach by reviewing the evidence base for interventions in which one's perception of reality is altered. Social skills interventions have most commonly been implemented in children but we chose to include young adults, based on evidence that brain maturation, particularly that of the prefrontal cortex which coordinates the goal directed behaviour needed to implement social skills, continues to develop into a person's early twenties (Johnson et al., 2009). This is also in line with current plans to extend children's mental health services in England up to 25 years (NHS Long Term Plan, 2019). Due to the infancy of this research area, we chose not to restrict inclusion on the basis of study design.

It was considered important to understand the effectiveness of these interventions because of the secondary benefits social skills can have on indicators of wellbeing, such as increased peer interactions (Mandelberg et al., 2014) and reduced anxiety and low mood (Hillier et al., 2011; Schohl et al., 2014). Secondly, the realism of immersive VR offers a unique potential to improve upon existing social skills interventions which do not always generalise to real contexts (Bellini et al., 2007). Thirdly, XR is at a point of rapid expansion, with VR headsets becoming increasingly widespread. If social skills interventions are going to capitalise on this expansion, it is important to consider whether there is any good quality evidence for their effectiveness and whether investment in these technologies will actually lead to meaningful benefits for autistic young people.

To this end, this review aims to answer the following questions:

1. Is there any evidence that extended reality interventions are effective in helping autistic children and young people to enhance their social skills?
2. What is the quality of the evidence for observed effects?
3. To what extent have observed effects been shown to lead to meaningful change for autistic people?

Method

This quantitative systematic review uses a synthesis without meta-analysis methodology and information is reported in line with Cochrane guidance (Garritty et al., 2021). The protocol was pre-registered with PROSPERO (ID: CRD42021229442; available at https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=229442). Five online databases (PsychInfo, Medline, Pubmed, EMBASE, CINAHL) were searched using key terms listed in Appendix 28. All retrieved datasets were downloaded into a Zotero library and exported to Excel for first author screening by title and abstract, then by full text. Another author (EM) screened 26.5% of studies at initial screening and 25% of studies at full text screening. All texts accepted at final screening were forward and backward citation searched using Scopus.

Screening and Selection

Studies were screened for eligibility according to the following inclusion criteria:

- The paper was published in English, in a peer-reviewed journal to ensure a minimum quality level.
- The paper was published after 1990, when AR was developed and VR headsets became widely commercially available.
- The paper included an active intervention using XR. Studies described as VR but which used a desktop computer or motion based video game were not included for

review as they do not to extend one's perception of reality.
- The study targeted one or more social skill.
- The paper reported a quantitative measure of social skills, at least pre and post intervention. This included in vivo outcomes, as well as validated and idiosyncratic measures of social skills.
- Study participants were children and young adults up to the age of 25.
- Study participants were autistic.

Where papers reported on more than one study, they were included based on the study which met the inclusion criteria. Borderline cases were discussed amongst the researchers.

Study Selection

The initial search identified 592 records, 324 of which were retained after duplicates were removed (Fig. 1). Two hundred seventy-five studies were excluded at the abstract stage, with a 96.5% inter-rater reliability (kappa = .87). Thirty-one studies were excluded at the full text screen, with a 91.7% inter-rater reliability (kappa = .83). Disagreements were resolved through further screening at the full text stage and discussion with other authors. Seven additional records were identified through hand searching and citation chaining (Scopus search conducted May 3, 2021). A total of 17 studies were included in the review and a number of data items were extracted from each study. Where it was not possible to extract data directly, study authors were contacted, or available information was used to calculate an effect size. 50% of data extraction was duplicated by a second reviewer (EM).

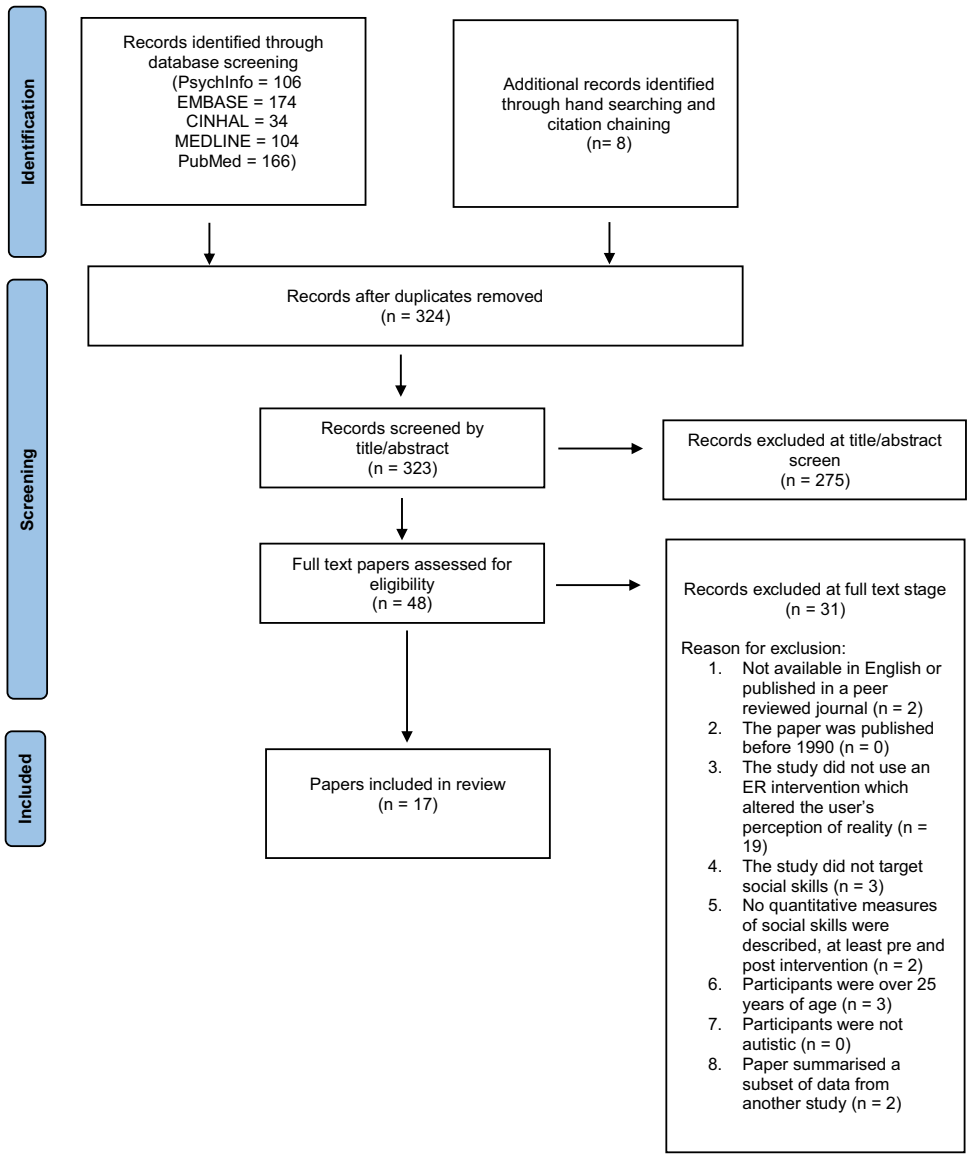

Fig. 1 Study selection process

Data Synthesis

Data were synthesised and organised according to the social communication criteria of DSM-5's autism diagnostic criteria (APA, 2000), as the effect of XR on specific social skills was the primary area of interest. In line with Siddaway et al. (2019), findings were integrated and critiqued, not merely summarised. The high number of single case studies meant that it was not possible to conduct a meta-analysis but key information is summarised in tables, with a summary of effect (Cohen's *d*) reported where enough information was provided in the paper or could be obtained through correspondence with the study authors. All studies were included for synthesis, irrespective of quality, but due to the range of study quality, summary plots were not developed, to avoid misrepresentation of the evidence base.

Quality Assessment

The quality and risk of bias of each study was evaluated using a quality checklist for healthcare intervention studies, developed by Downs and Black (1998). The tool was selected due to its applicability to both randomised and non-randomised studies. All papers were rated by the first author and 30% were duplicated by another author (EM), with an interrater reliability of 90.4% (kappa = 0.86). Discrepancies were resolved through discussion. Some items were not applicable to small-N research and so a summary score relative to the number of items assessed was generated for each paper to allow for direct comparison across studies. This was calculated by dividing the score obtained by the total possible score according to study design. A risk of bias score was generated by the same method, using only items assessing bias (14 to 26). A full summary of the assessment approach is provided in Appendix 31. Given that a number of items were not applicable to single case and small-N studies, the Single Case Experimental Design (SCED) Scale (Tate et al., 2008) was also used to qualitatively assess these studies (Appendix 32).

Results

Study Characteristics

Across the 17 studies (summarised in Table 1), 225 participants (15.5% female) aged 2 to 16 were included. Sample sizes ranged from one to 94 participants (M = 13.24, SD = 18.02). All participants were autistic however, only 11 studies reported that this was a clinical diagnosis according to DSM and only three confirmed diagnosis using a screening tool: Ip et al. (2018) using the Childhood Autism Spectrum Test (Williams et al., 2005); Liu et al. (2017) using the Social Communication Questionnaire (Chandler et al., 2007); and Lorenzo et al. (2019) using the Autism Spectrum Inventory (Rivière, 2002). Nine studies specified an intelligence quotient (IQ), with a mean or lower limit of at least 70. The studies used a range of designs, although a majority (10) were small-N, with either multiple baseline or pre-post measures. Only five studies used a control (Crowell et al., 2020; Herrero & Lorenzo, 2020; Ip et al., 2018; Lorenzo et al., 2016, 2019). Seven studies explicitly reported being at the preliminary or feasibility stage of the research.

Fourteen unique interventions were evaluated (seven VR; six AR; one MR) and are described in Table 2. Seven adapted existing social skills interventions, such as video modelling (Chen et al., 2016), collaborative game play (Mora-Guiard et al., 2017) and Social Stories™ (Herrero & Lorenzo, 2020). Rationale for the use of XR, included distraction reduction; the potential for engagement and sustained attention; freedom to practice social interactions without risk; and customisability i.e. the capacity to emphasise social stimuli and adapt difficulty level. Interventions most often took place in schools, and ranged from one 15 minute session (Crowell et al., 2020) to eighty 25-min sessions (Lorenzo et al., 2013). Some studies gave rational for these design decisions, such as short sessions due to attentional capacity, but the majority gave no justification for the frequency or duration of XR use. Adverse outcomes were not routinely assessed but one study commented that some children had difficulties with restlessness and tolerance of the headset (Ravindran et al., 2019) and another used 'reinforcement candy' to encourage 'emotional stability' during the intervention (Cheng et al., 2015).

Outcomes

Social-Emotional Reciprocity

Fifteen studies targeted social-emotional reciprocity: six using AR, seven using VR and two using MR. Specific targets included identifying and responding to social greetings; motivation to communicate with a conversation partner; understanding and use of social initiations; and sharing emotions and affect with others. Given the multiple countries in which these studies took place, it is possible that reciprocal behaviours considered socially appropriate differed according to cultural norms. Participants ranged from 6 to 16 years, although the majority of participants were primary school aged.

AR interventions included Smartglasses, which gave real-time prompts in social situations (Liu et al., 2017; Sahin et al., 2018; Vahabzadeh et al., 2018), a motion based game,

Table 1 Summary of included studies

Study	Design	Country of study	Name of intervention (AR/VR/MR)	Autism diagnosis and assessment criteria	Age range of participants (years)	IQ	Total sample size included in analysis (% female)	Social skills targeted	Measure of social skills (validated?)
Liu et al. (2017)	Single case series, pre-post	USA	Brain Power System - Face Game and Emotion Game (AR)	Clinical diagnosis according to DSM-5	8–9	Not specified	2 (0%)	Social emotional reciprocity and non-verbal communication—social withdrawal and non-verbal communicative behaviours e.g. eye contact	ABC (✓) and caregiver report (x)
Sahin et al. (2018)	Single case study	USA	Empowered Brain – Face-2Face Module (AR)	Clinical diagnosis but criteria not specified	13	Not specified	1 (0%)	Social emotional reciprocity—social awareness, cognition, communication, motivation and restricted interests	SRS-2 (✓)
Vahabzadeh et al. (2018)	Single case series, pre-post	USA	Empowered Brain - Face-2Face Module (AR)	Clinical diagnosis according to DSM-IV or DSM-V.	6.7–8.8	Not specified	4 (0%)	Social emotional reciprocity—a range of behaviours indicating social reciprocity	ABC (✓)
Lee (2020)	Multiple baseline experimental design	Taiwan	Real-time skeletal tracking system (AR)	Clinical diagnosis according to DSM-IV-TR	7–9	$M = 102.33$	3 (33.3%)	Social emotional reciprocity—understanding of social greetings	Novel situation based questions (x); Idiosyncratic ratings of role play behaviours (x)
Lee et al. (2018)	Multiple baseline experimental design	Taiwan	Augmented reality concept map training system (AR)	Clinical diagnosis according to DSM-IV-TR	8–9	$M = 93.3$	3 (33.3%)	Social emotional reciprocity—understanding of social greetings	Novel situation-based questions (x); idiosyncratic ratings of role play behaviours (x)

Table 1 (continued)

Study	Design	Country of study	Name of intervention (AR/VR/MR)	Autism diagnosis and assessment criteria	Age range of participants (years)	IQ	Total sample size included in analysis (% female)	Social skills targeted	Measure of social skills (validated?)
Lorenzo et al. (2019)	Case controlled, pre-post (non-randomised control)	Spain	Quiver Vision (AR)	Clinical diagnosis according to DSM-V and score > 30 on IAS	2–6	7 participants had a diagnosis of Developmental Delay, Intellectual Disability or Language Disorder	11 (9.1%)	Social emotional reciprocity—unclear target behaviour but domains of the IDEA include 'social relationship'; 'joint reference skills'; 'intersubjective and mentalistic skills'	IDEA[1]
Cheng et al. (2015)	Multiple baseline experimental design	Taiwan	3D-SU System (VR)	Diagnostic criteria not specified	10–12	$M = 82$	3 (0%)	Social emotional reciprocity—understanding of neurotypical social initiations	Novel situation-based questions (x)
Herrero & Lorenzo (2020)	Randomised controlled trial (parallel group design)	Spain	HMD Immersive VR System (VR)	Diagnostic criteria not specified	8–15	Not specified	14 (14.3% of experimental group, control not specified)	Social emotional reciprocity and non-verbal communication—social reciprocity, flexibility to change and non-verbal communication skills	Idiosyncratic ratings of in situ social reciprocity (x)
Ip et al. (2018)	Randomised controlled trial (parallel group design)	Hong Kong	Cave Automatic Virtual Environment (CAVE; VR)	Clinical diagnosis but diagnostic criteria unknown (mean score on Childhood Autism Spectrum Test above cutoff, 19.1)	7–10	95 (non-verbal)	72 (11.1%)	Social emotional reciprocity and understanding relationships—social reciprocity; emotion recognition; emotion regulation and everyday social skills e.g. maintaining relationships	PEP-3 (✓) ; Adapted Faces test (x); Eyes test (✓); ABAS-II (✓)

Table 1 (continued)

Study	Design	Country of study	Name of intervention (AR/VR/MR)	Autism diagnosis and assessment criteria	Age range of participants (years)	IQ	Total sample size included in analysis (% female)	Social skills targeted	Measure of social skills (validated?)
(Lorenzo et al., 2013)	Quasi-experimental, pre-post (no control)	Spain	Immersive Virtual Reality System (VR)	Asperger's syndrome, diagnostic criteria not specified	8–15	'Average Intelligence Levels' stated but not specified	20 (20%)	Social emotional reciprocity—no one primary outcome measure	Idiosyncratic ratings of in situ performance (x); Teacher ratings of classroom behaviours (x)
(Lorenzo et al., 2016)	Randomised controlled trial (parallel group design)	Spain	Immersive Virtual Reality System (VR)	Diagnosis according to DSM-V criteria	8–12	Not specified	40 (27.5%)	Social emotional reciprocity—a range of behaviours indicating social reciprocity	Idiosyncratic ratings of in situ performance (x); Teacher ratings of classroom behaviours (x)
Ravindran et al. (2019)	Single case series, pre-post	USA	Floreo's Joint Attention Module (VR)	Diagnosis confirmed by a family member	9–16	Not specified	12 (16.7%)	Social emotional reciprocity and non-verbal communication—frequency of interactions and eye contact	Joint attention assessment (x)
Crowell et al. (2020)	Randomised controlled trial (crossover[1] design)	Spain	Lands of Fog (MR)	Clinical diagnosis according to DSM -IV-TR	8–12	> 70	18 (16.7% assumed)	Social emotional reciprocity—social initiations and responses in a collaborative game	Frequency of in situ social initiations and responses (x)
Mora-Guiard et al. (2017)	Quasi-experimental, repeated measures (no control)	Spain	Lands of Fog (MR)	Clinical diagnosis according to DSM-IV-TR	10–14	> 70	10 (0%)	Social emotional reciprocity—social initiations, acts and responses in a collaborative game	Frequency of in situ social initiations, acts and responses (x)
Tsai et al. (2020)	Multiple baseline experimental design	Taiwan	Cave Automatic Virtual Environment (CAVE; VR)	Clinical diagnosis according to DSM-5 criteria	7–9	$M = 87$	3 (0%)	Social emotional reciprocity—understanding of social initiations in response to others' expressions	Novel situation-based questions (x); idiosyncratic rating of role play behaviours (x)

Table 1 (continued)

Study	Design	Country of study	Name of intervention (AR/VR/MR)	Autism diagnosis and assessment criteria	Age range of participants (years)	IQ	Total sample size included in analysis (% female)	Social skills targeted	Measure of social skills (validated?)
Chen et al. (2015)	Multiple baseline experimental design	Taiwan	Augmented Reality Self-Facial Modelling Learning System (AR)	Clinical diagnosis according to DSM-IV-TR	10–13	M = 101	3 (33.3%)	Understanding relationships—aimed to improve emotion recognition	Idiosyncratic measure of ability to identify facial expressions and emotions (x)
Chen et al. (2016)	Multiple baseline experimental design	Taiwan	AR with Video Modelling Storybook Learning System (AR)	Clinical diagnosis according to DSM-IV-TR	11–13	M = 103.66	6 (17%)	Understanding relationships—aimed to improve emotion recognition	Idiosyncratic measure of ability to identify facial expressions and emotions (x)

[1]It was not possible to determine whether this is a validated measure due to it having been published in Spanish

which provided opportunities to practice reciprocal behaviours (Lee, 2020), and a concept map enhanced with AR to aid learning (Lee et al., 2018). One study used an AR smartphone app to enhance child-therapist interactions, but the specific effects of AR were unclear (Lorenzo et al., 2019). VR interventions were less varied, with the majority simulating social situations in which participants could practice specific reciprocal behaviours (Cheng et al., 2015; Herrero & Lorenzo, 2020; Ip et al., 2018; Lorenzo et al., 2013, 2016; Ravindran et al., 2019). One study utilised a specific role playing game within the VR (Tsai et al., 2020). Conversely, the MR intervention (Crowell et al., 2020; Mora-Guiard et al., 2017) used mixed reality objects to digitally emphasise the benefits of collaboration with a partner.

Outcomes were most commonly measured by presenting participants with a range of social situations (sometimes in the form of Social Stories™) and asking them to describe, and/or demonstrate, how they might respond (Cheng et al., 2015; Herrero & Lorenzo, 2020; Lee, 2020; Lee et al., 2018; Tsai et al., 2020). These studies all concluded that social-emotional reciprocity could be improved using their specific XR intervention however, the criteria for rating responses was not validated or tested for age appropriateness using a non-autistic sample. Furthermore, one study reported that participants were given prompts when responding but did not specify a protocol for this (Cheng et al., 2015). Another reported improvements in social reciprocity but assessed behaviours using a novel and unvalidated joint attention assessment (Ravindran et al., 2019). A number of studies measured outcomes by observing changes in behaviours during the XR intervention. Again, it was unclear whether the coded behaviours were age-appropriate, or had any relation to social skills in the real world. Of studies measuring in situ behaviours, two concluded that behaviours improved after the intervention (Herrero & Lorenzo, 2020; Mora-Guiard et al., 2017) and one found no impact of XR on social reciprocity when compared to an active control (Crowell et al., 2020). Others did not set clear hypotheses regarding primary outcome measures, making it difficult to draw conclusions on their efficacy (Lorenzo et al., 2013, 2016). Only four of the studies targeting social-emotional reciprocity utilised validated outcome measures (Ip et al., 2018; Liu et al., 2017; Sahin et al., 2018; Vahabzadeh et al., 2018): the Aberrant Behaviour Checklist (ABC; Aman et al., 1985), Social Responsiveness Scale (SRS-2; Constantino & Gruber, 2012) and PEP-3 Psychoeducational Profile (Schopler et al., 2005). All of these studies showed an improvement in scores before and after the intervention, with a maximum follow up of 2 weeks (Ip et al., 2018). However, only in one instance was this in comparison to a control (Ip et al., 2018). One measure was unsuitable for the age of the study participants (Ip et al., 2018) and another indicated a 100% reduction in social difficulties (Liu et al., 2017) despite theoretical understandings

Table 2 Description of AR, VR and MR interventions and their reported purpose

Intervention	Description
AR	
AR based Self-Facial Modelling Learning System	This system is designed for children to interact with while wearing masks which represent particular emotions. AR superimposes facial expressions onto a participant's view of themselves, to show the emotion most appropriate to the situation. This augmentation of the participant's view of themselves is thought to promote learning.
AR Video Modelling With Storybook Learning System	A tablet computer is used to augment a physical storybook with short video clips, which interactively emphasise social details. This draws attention to social elements of the story and is thought to be more engaging than traditional video modelling approaches.
Real Time Kinect Skeletal Tracking System	This system augments a virtual character with a user's body gestures and superimposes a range of real life backgrounds. This enables children to practice a range of body gestures which might be appropriate to different situations e.g. shaking hands or waving arms. The primary benefit is that it is real time but without the anxiety associated with real world interactions.
AR Concept Map Training System	A tablet computer is used to augment a physical concept map with 3D greeting animations and graphics to indicate relationships between people and degrees of closeness in relationships. The purpose is to help children to visually learn about basic social relations and appropriate greeting behaviours.
Empowered Brain/ Brain Power System	Children wear a pair of Smartglasses which can evaluate looking behaviours and superimpose digital text and images onto the world, so that social cues are given in real time according to the user's ability. It includes a number of applications: *Face Game (also termed Face2Face)* uses an AR game to draw attention to human faces with visual overlays such as arrows and cartoon masks which gradually fade. Users score points by directing their gaze towards their caregiver's face. *Emotion Game* asks children to identify the facial expression of the person in front of them by tilting their head towards the correct emoticon. Points are gained for correct answers.
Quiver Vision	A smartphone application is used to augment real life pictures so that they are perceived as 3D objects. In Lorenzo et al. (2019), participants engage in activities which involve interacting with the AR components alongside the therapist, such as touching different AR objects according to the therapist's instructions.
VR	
3D-SU System	Users view and interact with animated social events using an immersive VR headset. Based on their responses to questions about the events, users are rewarded with a message and applause or instructed to try again. Using a headset is thought to reduce distraction and encourage sustained attention towards social events.
Cave Automatic Virtual Environment (CAVE) VR	Users are immersed in a number of social scenarios within a 'cave' of multiple large projection screens. This allows children to practice their responses free from risk of embarrassment. Ip et al. (2018) employ this technology to allow children to practice responses in a range of situations. They are supported by a trainer if needed and a relaxation scenario is available if the experience becomes distressing. Tsai et al. (2020) enhance the CAVE with motion control cameras. Users are encouraged to interact with avatars within the environment in specific role playing games.
Floreo - Joint Attention Module	A smartphone is fitted to a Google Cardboard headset, creating a VR experience. Users work through a number of 'learning cards' which require the demonstration of joint attention skills. A virtual avatar initiates and responds to joint attention bids and can prompt where necessary. A monitoring therapist can observe using a tablet computer and provide further support.
Immersive Virtual Reality System	Users are immersed in a number of scenarios using two large projection screens and movement detection software. Lorenzo et al. (2013) use this system to allow children to rehearse their responses to a range of classroom based tasks such as initiating conversation with a classmate. In Lorenzo et al. (2016), participants follow an evaluator's guidance on how to behave in a range of social situations e.g. attending a birthday party. This approach is thought to be beneficial due to its realism and the capacity to present information clearly without irrelevant distractors.

Table 2 (continued)

Intervention	Description
HMD Immersive VR System	Users are immersed in two school environments which are viewed through a head mounted display. Children can socially interact with a number of avatars, whose responses are controlled by the researchers. The system is reported to be highly immersive and flexible to allow for adaptation and promote real world generalisability.
MR	
Lands of Fog	Users collaborate with a partner to hunt for insects in a dense layer of virtual fog using a physical butterfly net. Explorative and collaborative efforts are rewarded with an upbeat tune or new feature. The approach is thought to be effective in encouraging collaboration as users can clearly see the actions of themselves and others, unlike in traditional collaborative game play.

of autism as a lifelong neurodevelopmental difference characterised by differences in social communication. One study measured outcomes using the Autism Spectrum Inventory (IDEA; Rivière, 2002). It is unclear whether this is validated as the measure is in Spanish, but no improvements were observed when compared with an active control, and again, the theoretical justification for use of this measure was limited (Lorenzo et al, 2019).

Overall, a number of studies have targeted elements of social-emotional reciprocity and almost all claim evidence of effectiveness (see Table 3). However, there is significant risk of bias in how outcomes were measured and the better quality studies tended to be single case. It is therefore difficult to draw conclusions about the efficacy of XR for improving this aspect of social communication. Furthermore, despite some qualitative reports, the studies did not attempt to determine generalisability and lack of longer-term follow-up makes it difficult to determine the extent to which improvements were sustained.

Non-verbal Communicative Behaviours

No studies targeted non-verbal communication in isolation but three studies (two VR and one AR) included non-verbal behaviours as outcome measures. Herrero and Lorenzo (2020) used an idiosyncratic measure of 'non-verbal behaviours' (e.g. use of facial cues, imitation and gestures) to suggest that use of their VR intervention was beneficial for 7- to 12-year-olds. However, all but one of the participants were rated as having 'fair' or 'good' non-verbal communication prior to the intervention and the measure used was not validated. Two studies concluded that their respective interventions increased eye contact: Ravindran et al. (2019) coded the amount of eye contact used by the 9- to 16-year-old participants during a joint attention assessment and Liu et al. (2017) utilised an idiosyncratic caregiver measure.

Given the limited number of studies which have investigated non-verbal communicative behaviours, it is difficult to draw conclusions about XR's efficacy for this domain of social communication. This is compounded by use of unvalidated measures, small sample sizes and unclear theoretical rationale for anticipated improvements in non-verbal behaviours. Only one study measured generalisability (caregiver report).

Developing, Maintaining and Understanding Relationships

Two AR studies primarily targeted the development, maintenance and understanding of relationships (Chen et al., 2015, 2016) and one VR study included some relational outcome measures (Ip et al., 2018). The AR studies targeted emotion recognition which was measured according to participants' ability to identify emotions in a story. According to an unvalidated assessment method, participants improved between baseline and follow up. Informal parent reports suggested that improvements corresponded with real world change in ability to identify emotions however, this was not formally measured. In contrast, Ip et al. (2018) found no evidence of improvements on this domain after use of VR, when compared with a waitlist control. They targeted the application of social skills to real life (e.g. ability to maintain relationships), measured using a domain of the Adaptive Behaviour Assessment System (ABAS-II; Harrison & Oakland, 2003), and emotion recognition, which was measured using the Eyes and Faces Tests (Baron-Cohen et al., 1997, 2001). Mixed outcomes from this small number of studies mean it is not possible to conclude whether XR is effective for improving relational social communication.

Related Skills

Two studies included outcomes which would theoretically benefit social skills but do not fall under the DSM-5 social communication criteria (APA, 2013), such as emotion regulation (Ip et al., 2018) and flexibility to changes during social situations (Herrero & Lorenzo, 2020). When compared with

Table 3 Summary of results

Study	Quality rating	Risk of bias rating	Effect of intervention on social skills (+ indicates improvement and – indicates lack of improvement)
Liu et al. (2017)	0.61	0.67	Observed reduction in social withdrawal after using the Brain Power System (+) Carer reported increases in non-verbal communicative behaviours post-intervention (+)
Sahin et al. (2018)	0.56	0.83	Observed reduction in ratings of social skills difficulties measured by the SRS-2 after using Empowered Brain (+)
Vahabzadeh et al. (2018)	0.61	0.67	Observed reduction in social withdrawal after using Empowered Brain, at two time points (+)
Lee (2020)	0.58	0.5	Statistically significant mean improvement in understanding of social greetings at maintenance in comparison to baseline (+)
Lee et al. (2018)	0.53	0.33	Statistically significant mean improvement in understanding of social greetings at maintenance in comparison to baseline (+)
Lorenzo et al. (2019)	0.67	0.62	No significant difference between pre- and post- scores across all dimensions of the IDEA (-) No significant difference in post-intervention ratings between the intervention group and non-digital control for all items compared (-)
Cheng et al. (2015)	0.56	0.5	Observed increases in ability to understand social initiations across baseline, intervention and maintenance (+)
Herrero & Lorenzo (2020)	0.31	0.23	Observed improvements in social reciprocity and inflexibility to change post-intervention for all participants (+) Observed improvements in non-verbal communication post-intervention for 6 of 7 participants (+)
Ip et al. (2018)	0.48	0.54	Statistically significant improvement social reciprocity post-intervention (+) ($d = 0.50$) Statistically significant improvement in emotion regulation post-intervention (+) ($d = 0.39$) No significant difference in emotion recognition (Faces Test) post-intervention (-) ($d = 0.26$) No significant difference in emotion recognition (Eyes Test) post-intervention (-) ($d = 0.19$) No significant difference in everyday social skills post-intervention (-) ($d = 0$) Statistically signification effect of group (intervention vs waitlist control) on improvements in emotion regulation (+) ($d = 0.54$) and social reciprocity (+) ($d = 0.67$)
Lorenzo et al. (2016)	0.37	0.46	No clear primary outcome measure. Study reported greater overall improvements in the intervention group, compared with the non-immersive control (+)
Ravindran et al. (2019)	0.42	0.33	Observed increases in frequency of interactions and amount of eye contact after using the VR system (+)
Crowell et al. (2020)	0.46	0.33	No significant difference in social initiations using MR, compared with non-digital control (-) ($d = 0.06$) Statistically significantly more social responses in control condition, compared with MR intervention (-) ($d = 0.14$)
Mora-Guiard et al. (2017)	0.39	0.33	Observed increases in initiations, acts and responses while using the MR system, between sessions 1 and 3 (+)
Tsai et al. (2020)	0.5	0.6	Observed improvements in understanding of social initiations between baseline and intervention for all participants. Sustained at maintenance for 2 of the 3 participants (+/-) Statistically significant increase in therapist ratings of role play performance at intervention and maintenance (+)
Chen et al. (2015)	0.5	0.43	Statistically significant mean improvement in emotion recognition at follow-up in comparison to baseline (+)
Chen et al. (2016)	0.5	0.43	Statistically significant mean improvement in emotion recognition at follow-up in comparison to baseline (+)

a waitlist control, the former showed an increase in emotion regulation ratings after use of VR, as measured by the PEP-3 (Schopler et al., 2005), and the latter showed improvements in flexibility to change across all participants in the intervention group, according to idiosyncratic measures.

Quality of Included Studies

Quality ratings ranged from 0.31 to 0.67 (where possible scores are between 0 and 1) with an average quality rating of 0.50. Risk of bias scores ranged from 0.17 to 0.83 with

an average of 0.47. Although the adapted method of scoring makes it difficult to give a precise assessment of quality, comparisons with previous applications of this checklist suggests that the studies were generally of poor to fair quality (Hooper et al., 2008). Higher scores were typically given for clarity of reporting, such as describing the main aims, interventions and findings. The majority of studies took place in representative locations, and there was little evidence of non-compliance. Lower quality studies tended to have non-representative samples, unclear recruitment strategies and lack of reporting on attrition. Furthermore, several studies did not attempt to blind those involved in rating outcomes. It was particularly notable that no studies gave justification for their selected sample size.

Further quality appraisal using the SCED Scale indicated that the majority of small-N studies were truly experimental in design (7 of 10) and conducted sufficient sampling at baseline and treatment phases; some of these studies also performed statistical analysis. However, a lack of precise and repeatable outcome measures was a significant weakness across small-N studies. In particular, outcomes tended to be rated by one, non-independent assessor and where there were multiple assessors, interrater reliability was not calculated. No attempt was made to show generalisation across settings or therapists.

Discussion

This systematic review identified 17 studies that investigated the potential of XR interventions to enhance the social skills of autistic children. To date, the majority of interventions have targeted social-emotional reciprocity, with relatively little attention given to the non-verbal and relationship aspects of social-communication. The overall quality of the research is relatively low, perhaps reflecting the infancy of the research area. The majority of studies have small sample sizes and a number are not truly experimental, making it difficult to draw firm conclusions about the efficacy of XR, particularly in comparison to current, less costly social skills interventions. Significant heterogeneity means it is not possible to determine the 'active ingredient' of XR interventions. Furthermore, only limited attempts have been made as yet to determine generalisability and there is limited exploration of whether the statistically significant changes observed by many of the studies were also clinically significant i.e. led to improvements in everyday social or occupational skills, or other important areas of functioning.

Limitations of the Current Literature

Given the relative novelty of this research, it is important to consider the limitations of the current evidence base, to be improved upon in future research. In particular, current evidence is limited in its ethical considerations, theoretical grounding, robustness of study design and sample representativeness. It is also unclear as yet, how easily and cost-effectively XR interventions could be scaled to clinical practice.

Ethical Considerations

Social skills interventions are primarily important because of the negative secondary effects of social difficulties and so quality of life has been an important outcome measure across the literature (e.g. Baghdadli et al., 2013; Mitchel et al., 2010). However, none of the studies included in this review measured quality of life, nor did they consider the importance of the intervention target to the participants themselves. Krasny et al. (2003) emphasise that interventions are most efficacious when children understand the relevance of the social skill and work towards individualised goals. Future interventions could capitalise on the customisability of XR to tailor interventions to individual needs. It is perhaps telling that the majority of interventions have targeted social reciprocity which, by definition, impacts those around autistic people. There are few attempts to directly target development and maintenance of relationships, despite this being what some autistic people have expressed as most important (Cresswell et al., 2019). Some studies did use participatory research methods, i.e. the incorporation of the views of autistic people into the design and implementation of the research (Cornwall & Jewkes, 1995), although the extent of involvement is somewhat limited. Increased meaningful involvement from autistic people within this field of research would help to ensure outputs are relevant and beneficial to the people for whom they are designed (Fletcher-Watson et al., 2019).

Future research should also make a more comprehensive attempt to measure adverse events, particularly given that one study which did comment on negative outcomes, noted problems with restlessness and discomfort, while another reported using positive reinforcement to keep children engaged. Moreover, commercially available VR headsets typically include safety notices regarding their use in children, emphasising the need for robust assessment of potential harm. Future studies could improve this through quantitative measurement of participant experience, such as whether the tool was straightforward to use and physically and sensorially comfortable, and analysis of the relationship to outcomes i.e. whether more positive experiences are associated with greater improvements. As well as the potential adverse effects of XR, future studies should consider the wider impact of teaching children to implement neurotypical social skills. Many autistic people already invest significant time and energy in monitoring and modifying their behaviour in order to align with social norms, known as

'social camouflaging'. Evidence suggests that this hiding of one's true self can be mentally and physically draining, and requires excessive concentration, self-control and discomfort (Hull et al., 2017; Mandy, 2019). Further exploration is needed of whether social skills training simply teaches children to mask their true selves, and if so, serious attention should be given to the potential deleterious impact on wellbeing. Perhaps greater research focus could instead be given to how settings, such as schools, could adapt to become more accommodating for autistic people, rather than placing all emphasis on changing autistic children to manage in a neurotypical world. This would be more consistent with the shared responsibility for communication between autistic and non-autistic people highlighted by the double empathy problem (Milton, 2012).

Theoretical Grounding

Despite the number of theoretical explanations for social differences in autism, the studies reviewed in this research give relatively little theoretical justification to the design of their interventions. As a result, XR has been applied in a number of different ways, often with no clear rationale for why design decisions were made. Greater attention to theory when designing interventions might further improve efficacy. This has proved beneficial for non-XR studies, such as the *Mind Reading* intervention which draws on the empathising-systemising theory by exploiting participants' relative strength in systematising when teaching emotion recognition (Golan & Baron-Cohen, 2006). Limited consideration of the mechanism by which change might occur may also account for lack of specificity of some of the research. In particular, some studies measured a number of outcomes with no clear indication of how these related to the purpose or design of the intervention. Theoretical understandings of the mechanism of change could also inform intervention duration, which varied significantly across the studies. While it seems theoretically unlikely that clinically significant changes could be made in the time of the shortest intervention (15 min), strong theoretical justification should be given before subjecting children to high numbers of intervention sessions.

Study Design

Quality appraisal highlighted a number of methodological limitations of the current research. For example, the majority of studies did not attempt to blind the individuals who coded behavioural outcomes and in fact, behaviours were often rated by researchers or parents who knew the purpose of the study, both of whom are likely to have been highly invested in positive outcomes. In particular, parents of autistic children have been shown to report positive effects when told their child is receiving treatment, even in the absence of an intervention (Jones et al., 2017). It is also possible that improvements occurred as a result of repetition of the outcome measures themselves. For example, a number of studies used questions based on Social Stories™ to measure outcomes, and as a social skills intervention in itself, this may have contributed to improvements. Similarly, when outcomes were measured by in vivo performance, improvements may have been due to practice effects, rather than social skill development. Greater use of validated measures of social skills, applied by raters who are blinded to the study purpose and intervention allocation, would improve some of these issues. Future research should also ensure that potential confounders, such as whether participants are receiving any other therapies, are fully considered.

Appropriately for the stage of research, the majority of studies were small-N. However, in larger trials, no details were given about the determination of sample size, and so it may be that those which did not find an effect were underpowered. In one study, a significant proportion of the sample were excluded from analysis based on age, however, the purpose of this is not explained, diminishing the validity of their results. To determine effectiveness, more large-scale trials are required: these should be sufficiently powered and ideally pre-registered, to reduce the likelihood of data dredging and subsequent false positives. More experimental, controlled studies would also make it easier to determine, not only whether XR interventions are effective, but whether they offer any real improvement to social skills, over and above current methods.

Representativeness of Sample

Current research is limited in the extent to which participants are representative of the autistic population as a whole. Around 25% of autistic people are female (Loomes et al., 2017) but only five of the included studies met this threshold. Similarly, up to 50% of the autistic population are estimated to have an Intellectual Disability (ID; Charman et al., 2011; Loomes et al., 2017) yet a number of the included studies excluded participants with an ID or stated a mean IQ within the average range. Limited justification is given for this and it compounds evidence that people with ID are consistently excluded from autism research (G. Russell et al., 2019). Future research should seek to recruit participants who truly reflect the autistic population as a whole, or provide strong justification for exclusion.

Limitations of the Review

The findings of this review should be viewed in the context of its limitations. Firstly, an effect size could only be given

for a limited number of studies making it impossible to compare the efficacy of different interventions using meta-analysis. Instead, this study used a systematic but non-analytic approach to synthesis. Vote counting and summary plots were avoided to reduce the potential misrepresentation of efficacy, given the number of low quality studies. This could perhaps have been improved by only including studies which meet a particular quality threshold. However, this was not deemed feasible for this review given the small number of studies. As the research develops and more experimental, controlled studies are carried out, more systematic methods of synthesis will be possible. Furthermore, publication bias will have significantly impacted the number of studies showing positive effects. Grey literature was not included, to ensure included studies were of peer-review quality, but this may have increased the positive results bias. When determining studies to be included, there were some borderline cases, where insufficient descriptions of study interventions made it difficult to establish whether the XR was immersive. This was resolved through discussion amongst researchers but, could have been improved by contacting study authors for more information.

The conclusions that can be drawn from this review are also constrained by the limited amount of data, both in terms of number of studies and overall sample size. The maximum age of participants across studies was 16 and so it is unclear whether XR interventions could be effective for helping older teenagers and young adults to enhance their social skills. Additionally, only VR studies which altered one's perception of reality were included and so the efficacy of immersive VR could not be compared to that of VLEs, which may be more cheaply and easily implemented in clinical practice.

Conclusion

The potential to digitally alter how one perceives the world offers exciting new possibilities in developing effective and engaging social skills interventions for autistic children. However, enthusiasm for XR interventions should be viewed with caution. While there is some indication that interventions are feasible to implement with autistic children, limited evidence exists for their effectiveness in bringing about meaningful, longstanding improvements in everyday functioning. Furthermore, no assessment has been made of the potential emotional cost of autistic children implementing behaviours which do not come naturally to them. This review demonstrates the need for theoretically grounded interventions, designed with the interests of autistic people at the forefront. Controlled trials and larger sample sizes, as well as other improvements to study design, are required to draw firm conclusions about the efficacy of XR interventions and their generalisability, before potentially high cost scaling to routine clinical services is considered.

Appendix 1

Search terms*

1. 'Extended Reality' OR 'Virtual Reality' OR 'Virtual Environment' OR 'Augmented Reality' OR 'Augmented Environment' OR 'Simulat* Reality' OR 'Mixed Reality' OR 'VR' OR 'AR' OR 'XR' OR 'MR'
2. 'Child*' OR 'Young' OR 'Youth*' OR 'Adolescen*' OR 'Student*' OR 'Teen*' OR 'Boy*' OR 'Girl*' OR 'P?ediatric*' OR 'Minor*'
3. "Social Skill*' OR "Social Interacti*' OR "Social Competence' OR "Social Responsiveness' OR 'Social Motivation' OR 'Social Abilit*' OR 'Social Awareness' OR 'Conversation Skill*' OR 'Conversation Abilit*' OR 'Friendship Skill*' OR 'Peer interacti*' OR 'Emotion* Recognition' OR 'Facial Recognition' OR 'Perspective Taking' OR 'Turn Taking' OR 'Turn-Taking' OR 'Theory of Mind' OR 'Communicat*' OR 'Interact*'
4. 'Autis*' OR 'Asperger*' OR 'ASD' OR 'ASC' OR 'Pervasive Developmental Disorder' OR 'PDD' OR 'HFA'
5. 1 AND 2 AND 3 AND 4

*Keyword and thesaurus searches were also conducted for each of the above terms, where possible within each database.

Terms considered but excluded

'TOM', 'Toddler*', 'Baby', 'Babies', 'Newborn*', 'Preschool*', 'Preschool*'

Appendix 2

The Downs and Black Checklist (below) was used to assess study quality. The scoring of item 27, referring to the power of the study, was modified. Instead of rating according to an available range of study powers, we rated whether or not a power calculation was performed. Items which were not applicable to single case and small-N studies are marked*. Items which were not applicable to studies which did not use statistical analysis are marked †. Questions 14 and 22 were not applicable to studies which did not have a control intervention. All other items were rated as *Yes* (scored as 1) or *No* (scored as 0) or *Unable to Determine* (scored as 0). Items 14 to 26 specifically assess bias and so were used to generate a risk of bias score.

1. *Is the hypothesis/aim/objective of the study clearly described?*

2. *Are the main outcomes to be measured clearly described in the Introduction or Methods section?* If the main outcomes are first mentioned in the Results section, the question should be answered no.

3. *Are the characteristics of the participants included in the study clearly described?* In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case-control studies, a case-definition and the source for controls should be given.

4. *Are the interventions of interest clearly described?* Treatments and placebo (where relevant) that are to be compared should be clearly described.

5. *Are the distributions of principal confounders in each group of subjects to be compared clearly described?* A list of principal confounders is provided.

6. *Are the main findings of the study clearly described?* Simple outcome data (including denominators and numerators) should be reported for all major findings so that the reader can check the major analyses and conclusions.

7. *Does the study provide estimates of the random variability in the data for the main outcomes?* In non-normally distributed data the inter-quartile range of results should be reported. In normally distributed data the standard error, standard deviation or confidence intervals should be reported. If the distribution of the data is not described, it must be assumed that the estimates used were appropriate and the question should be answered Yes.

8. *Have all important adverse events that may be a consequence of the intervention been reported?* This should be answered yes if the study demonstrates that there was a comprehensive attempt to measure adverse events. (A list of possible adverse events is provided).

9. *Have the characteristics of patients lost to follow up been described?* This should be answered yes where there were no losses to follow up or where losses were so small that findings would be unaffected by their inclusion. This should be answered no, where the study does not report the number of patients lost to follow up.*

10. *Have actual probability values been reported (e.g. 0.035 rather than <.05) for the main outcomes except where the probability value is less than 0.001?* †

11. *Were the subjects asked to participate in the study representative of the entire population from which they were recruited?* The study must identify the source population for participants and describe how the participants were selected. Participants would be representative if they comprised the entire source population, an unselected sample of consecutive participants, or a random sample. Random sampling is only feasible where a list of all members of the relevant population exists. Where a study does not report the proportion of the source population from which the patients are derived, the question should be answered as unable to determine.

12. *Were those subjects who were prepared to participate representative of the entire population from which they were recruited?* The proportion of those asked who agreed should be stated. Validation that the sample was representative would include demonstrating that the distribution of the main confounding factors was the same in the study sample and the source population.

13. We*re the staff, places and facilities where the patients were treated, representative of the treatment the majority of patients receive?* For the question to be answered yes the study should demonstrate that the intervention was representative of that in use in the source population. The questions should be answered no if, for example, the intervention was undertaken in a specialist centre unrepresentative of the hospitals most of the source population would attend.

14. *Was an attempt made to blind study subjects to the intervention they have received?* For studies where the patients would have no way of knowing which intervention they received, this should be answered yes.*

15. *Was an attempt made to blind those measuring the outcomes of the intervention?*

16. *If any of the results of the study were based on 'data dredging', was this made clear?* Any analysis that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analyses were reported, then answer yes.

17. *In trials and cohort studies, do the analyses adjust for different lengths of follow up of patients, or in the case-control studies, is the time period between the intervention and outcome the same for cases and controls?* Where follow-up was the same for all study patients, the answer should be yes. If different lengths of follow-up were adjusted for by, example, survival analysis the answer should be yes, studies where differences in follow-up are ignored should be answered no.**

18. *Were the statistical tests used to assess the main outcomes appropriate?* The statistical techniques must be appropriate to the data. For example non-parametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered yes. If the distribution of the data (normal or not) is not described it must be assumed that the estimates used were appropriate and the questions should be answered yes.

19. *Was compliance with the intervention/s reliable?* Where there was non-compliance with the allocated treatment or where there was contamination of one group, the question should be answered no. For studies where the effect of any misclassification was likely to bias any association to the null, the question should be answered yes.

20. *Were the main outcome measures used accurate (valid and reliable)?* For studies where the outcome measures are clearly described, the question should be answered yes. For studies which refer to other work or that demonstrated the outcomes measures are accurate, the question should be answered as yes.

21. *Were the participants in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?* For example, patients for all comparison groups should be selected from the same hospital. The question should be answered unable to determine for cohort and case control studies where there is no information concerning the source of patients included in the study.* †

22. *Were study participants in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?* For a study which does not specify the time period over which patients were recruited, the question should be answered as unable to determine.*

23. *Were study subjects randomised to intervention groups?* Studies which state patients were randomised should be answered yes except where method of randomisation would not ensure random allocation. For example, alternate allocation would score no because it is predictable.* †

24. *Was the randomised intervention assignment concealed from both patients and health care staff until recruitment was complete and irrevocable?* All non-randomised studies should be answered no. If assignment was concealed from patients but not from staff, it should be answered no.* †

25. *Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?* This question should be answered no for trials if: the main conclusion of the study were based on analyses of treatment rather than intention to treat; the distribution of known confounders in the different treatment groups was not described; or the distribution of known confounders differed between the treatment groups but was not taken into account in the analyses. In non-randomised studies, if the effect of the main confounders was not investigated or confounding was demonstrated but no adjustment was made in the final analyses the question should be answered as no.* †

26. *Were losses of patients to follow up taken into account?* If the numbers of patients lost to follow-up are not reported, the question should be answered as unable to determine. If the proportion lost to follow up was too small to affect the main findings, the question should be answered yes.

27. *Is there any evidence that a power calculation or reasonable equivalent was used to determine sample size?*

Appendix 3

The SCED scale (below) was used to qualitatively assess the quality of the single case and small-N studies included in this review. All items are scored as *Yes* or *No*.

1. Clinical history was specified. Must include age, sex, aetiology and severity?

2. Target behaviours. Precise and repeatable measures that are operationally defined. Specify measure of target behaviour.

3. Design 1: 3 phases. Study must be either A-B-A or multiple baseline.

4. Design 2: Baseline (pre-treatment phase). Sufficient sampling was conducted.

5. Design 3: Treatment phase. Sufficient sampling was conducted.

6. Design 4: Data record. Raw data points were reported.

7. Observer bias: Inter-rater reliability was established for at least one measure of target behaviour.

8. Independence of assessors.

9. Statistical analysis.

10. Replication: either across subjects, therapists or settings.

11. Evidence for generalisation.

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Declarations

Conflict of Interest The authors declare no competing interests.

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