Exploring domains of 'frontal dysfunction' relevant to everyday life following acquired brain injury.

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ABSTRACT

Damage to the frontal areas of the brain is associated with alterations in cognitive, social, and emotional regulation abilities. These neuropsychological consequences present challenges to ecologically valid assessment (difficulties in everyday life being poorly predicted by traditional neuropsychological test performance) and far transfer of rehabilitation gains to everyday life. In this thesis, the literature on 'cold' and 'hot' cognitive, social and emotional frontal functions is discussed in relation to these challenges. Gaps in the research are identified relating to 1. Associations between specific 'hot' and 'cold' cognitive processes, 2. Association of 'hot' and 'cold' cognitive processes with everyday outcomes. Four studies are presented each addressing a different aspect of these gaps: 'cold' executive and 'hot' emotion regulation abilities and peer relationships following paediatric acquired brain injury (ABI); patterns and predictors of performance of people with traumatic brain injury (TBI) on a modified gamble task compared with healthy controls; interaction between coping style and specific executive functions in association with emotional outcomes after ABI; and the effect of brief goal management training (GMT) and periodic alerts on achievement of everyday intentions following ABI. Results indicate variation in the extent to which 'hot' and 'cold' frontal functions are associated with each other and everyday outcomes. A 'frontal-contextual system' model in which performance characteristics

arise from the dynamic interaction between 'hot' and 'cold' frontal systems and everyday practical and social contexts is presented as a way of understanding everyday difficulties. Application of novel methodologies that can sample the interactions between system components and are sensitive to inter-individual variability may be useful for advancing understanding of the links between frontal dysfunction and everyday life. Implications for intervention that are similarly focused on the interactions between components and facilitation of social or physiological conditions that give rise to optimal adaptation in everyday life are discussed.

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“Essentially, all models are wrong, but some are useful”

George Box

Box, G. E. P. & Draper, N. R. (1987) *Empirical Model-Building and Response Surfaces*. John Wiley & Sons. p. 424

CHAPTER 1

1 Frontal functions and challenges in everyday life after Acquired Brain Injury

1.1 Background to the clinical problem: from clinic to everyday challenges in life after ABI

People with acquired brain injury (ABI, secondary to trauma, infection, stroke, tumour) are more likely to experience significant psychosocial problems related to acquired cognitive and other deficits, increased risk of behavioural or mental health problems (Deb, Lyons, Koutzoukis, Ali, & McCarthy, 1999; Morrison, Pollard, Johnston, & MacWalter, 2005) and changes to self-identity (Beadle, Ownsworth, Fleming, & Shum, 2016). People can experience considerable difficulties in resuming pre-injury roles or adjusting their lives to achieve meaningful engagement in new roles (Olver, Ponsford, & Curran, 1996). Whilst recovery trajectories may vary, a significant number survive brain injury with relatively low levels of medical or physical dependency, and intact ability to complete simpler everyday living tasks independently, but struggle with everyday life, psychiatric symptomatology, accessing and maintaining stable employment, and family and social relationships (Hoofien, Gilboa, Vakil, & Donovick, 2001; Jourdan et al., 2016). The term ‘hidden disability’ (Simpson, Simons, & McFadyen, 2002) has been used to describe the cognitive and emotional sequelae that can be devastating but are often not recognised by others, including services (Gladman et al., 2007). Enduring cognitive difficulties commonly include executive or higher attentional functions, a set of related functions typically implicated in the management of goal directed behaviour across varied contexts including: self-awareness and self-monitoring, problem solving, cognitive flexibly, attentional control, prospective memory, sequencing and ability to respond appropriately in a given situation (Ashman, Gordon, Cantor, & Hibbard, 2006; Diamond, 2013; Zinn, Bosworth, Hoenig, & Swartzwelder, 2007). There has been a growing trend for investigation of social and emotional processes that are also associated with frontal areas of the brain. As a result, a distinction has been made between 'cold executive' processes, which do not require subjective experience of emotion, and 'hot executive' processes, such as emotion-based decision-making or aspects of empathy, which do (Takeuchi et al., 2013). Difficulties with 'hot' and 'cold' executive functions are also amongst the most commonly reported consequences of traumatic brain injury (TBI) in childhood during the first year post injury (Sesma, Slomine, Ding, & Mccarthy, 2008) and longer-term (Ganesalingam, Sanson, Anderson, & Yeates, 2007; Hawley, Ward, Magnay, & Long, 2004). Amongst children who have sustained a brain injury, the effects of injury on 'higher' executive and self-regulatory functions need to be considered within a developmental context (Catroppa & Anderson, 2010). A child may not have yet reached the stage of development where a specific cognitive function is 'on line' and so may not show a deficit until later in childhood, termed 'neurocognitive stall' by Chapman (2006). This appears to be especially so in the domains of social and executive cognitive functioning (Catroppa & Anderson, 2010).

From a patient perspective Pollock, St George, Fenton, & Firkins (2012), identified stroke survivors' and healthcare professionals' top two priorities as improvement of cognition and helping people come to terms with the long-term consequences of stroke as the second priority. Winter et al. (2016) found that for veterans with TBI, cognitive and physical difficulties ranked most problematic, whereas for caregivers emotional and interpersonal domains were ranked most problematic.

Despite this clear priority to address higher cognitive, social and emotional self-regulatory functions in clinical practice, assessments of executive function are notoriously poor at predicting everyday outcomes following ABI (Manchester, Priestley, & Jackson, 2004). For social and emotional regulatory impairments, there are significantly fewer suitable standardised measures for assessing these domains objectively as compared with 'cold' cognitive domains (Spikman, Timmerman, Milders, Veenstra, & van der Naalt, 2012). Whilst there is a developing evidence base for cognitive rehabilitation (Stamenova & Levine, 2018; Tate et al., 2014), challenges remain in the meaningful transfer and maintenance of gains beyond clinic-based programmes into everyday life, in part due to the very nature of difficulties in EF that impact upon goal directed behaviour, adaptation and self-awareness (Gracey, Fish, et al., 2016; Tate et al., 2014). Together, this highlights the need to improve understanding and assessment of, and interventions for, the everyday cognitive and social neuropsychological difficulties people face, alongside adaptation to their changed circumstances over time.

1.2 Defining 'frontal functions'

In this section a summary of issues in defining 'frontal functions' is presented, after which links between everyday life (social, emotional and participatory outcomes) and processes typically associated with frontal systems (including prefrontal cortex, medial temporal and limbic structures and anterior parietal areas), will be explored. Note: Whilst anatomical localisation will be discussed where relevant to consideration of the organisation and development of frontal functions, discussion of the anatomical substrates of these functions is not the primary focus of this thesis.

1.2.1 Introduction to theories of frontal functions

Luria (1995) held a hierarchical view of the organisation of brain functions, describing the frontal lobes as serving an overarching function above all other domains of cognition, for the 'programming, controlling and verification' of behaviour. Influenced by Vygotsky, Luria saw these 'higher cognitive functions' as having developed from the interplay between biology and social context, for example through the internalisation of language or other practices that enable performance of more complex tasks (Vygotsky, 1978). Ardila (2008) elaborates this account, proposing that the higher metacognitive functions associated with goal directed behaviour and problem solving arise from increasingly elaborate (as in speech) and internalised (as in thought) motor sequencing. Lezak (1982) described executive functions (EFs) as relating to 'how' a person does something (initiation, consistency, self-correction, response to changes etc) whereas other domains of cognition are concerned with 'what' the person can do, or 'what' the material being processed is (words, objects etc). Although Lezak's account refers to component processes, the view is held, consistent with Luria, that EFs are 'supramodal', having an overarching effect on 'all aspects of behaviour' (Lezak, 1982; p. 283). Baddeley (2003) also described a supramodal central executive involved in the management of verbal and visual short-term memory 'slave' systems, which retain information long enough for consolidation into long term memory. Contemporary models of the functions of frontal functions have moved away from unitary accounts towards a multi-component architecture of at least partially dissociable neuropsychological functions. Miyake and colleagues (Miyake et al., 2000; Miyake & Friedman, 2012) for example define the processes comprising the central executive, initially identifying inhibiting (as tapped by go, no-go tasks for example), shifting (as when the category rule changes in a card sort test) and updating (shown in tasks generating interference, such as the Brown-Peterson paradigm). Miyake et al. (2000) found these functions contributed unique variance to task performance but were also correlated, which they termed the 'unity / diversity' hypothesis. Miyake & Friedman's (2012) later analysis suggested 'inhibition' comprised the 'supramodal' component of executive working memory, with inhibition and shifting separate components.

Distinct but related networks of frontal brain activation have also been identified from research employing functional neuroimaging; the 'default mode network' (DMN; see Andrews-Hanna, 2012 for a review), and a stimulus driven network (SDN; Corbetta and Shulman, 2002). The DMN appears to be involved in self-reflective or evaluative tasks including those that involve comparison of self with others, or retrieval of autobiographical memories. The SDN serves to orient our attention to the occurrence of a salient stimulus in the environment, and then evaluate the stimulus to determine and initiate the relevant course of action. These large brain networks appear to be composed of component hubs associated with further subdivision of functions. Although the SDN and DMN networks appear as independent and associated with distinct types of task, recent findings suggest these networks interact during more complex tasks, for example when engaged in a cognitive transition between tasks (Smith, Mitchell, & Duncan, 2018).

Despite evidence for at least partial dissociation of localised functions, and evidence for 'network' models, this historical notion of EF's representing a common overarching function has led to the description of a wide range of difficulties arising from frontal lesions being described in terms of 'executive' deficits. The legacy of this is that in clinical practice and research, the terms 'frontal functions', 'executive functions' and 'dysexecutive syndrome' have often been used synonymously and have become somewhat problematic to define due to the wide range of uses and definitions in the literature. The term 'dysexecutive syndrome' was coined by Baddeley & Wilson, 1988) to account for the diverse failures in cognitive and self-regulatory behaviour observed in patients with frontal lesions and arises from the assumption of a supramodal frontal 'executive' process, which if impaired leads to a 'dysexecutive' presentation. However, executive functions are now seen as not synonymous with frontal functions, which comprise a range of specific cognitive, meta-cognitive and socio-emotional functions (as argued by Ardila, 2008; Stuss, 2011). A corollary of this is therefore that dysexecutive syndrome cannot be understood as a failure specifically of executive functions.

1.2.2 'Hot' and 'cold' frontal functions

Descriptions of executive or frontal functions have labelled systems involving more logical processing as 'cold' and those involving arousal or affect as 'hot' (Chan, Shum, Toulopoulou, & Chen, 2008; Takeuchi et al., 2013). Ardila (2008), drawing on neuropsychological research as well as evolutionary theory, argues for two dissociable frontal systems – a dorsolateral executive / metacognitive system (cognitive processes associated with regulation of thought and behaviour, such as problem solving), and an orbitofrontal and medial social and emotional system (for regulation of emotional reactions and social behaviour). Extending this dichotomous approach, Stuss (2011) distinguishes between (cold) executive functions (such as task setting and monitoring, equivalent to the component functions of the 'central executive'), and (hot) emotional regulation processes (associated with reversal learning, reward processing as well as behavioural inhibition). Stuss also describes integrative 'metacognitive' functions (associated with self-reflection and conscious awareness, empathy and integration of multiple 'hot' and 'cold' processes as is often necessary in everyday life), and 'energisation' (ability to initiate and sustain a response). Stuss' anatomical distinctions differ from some accounts, and conceptualisation of component functions could be further elaborated. For example, there are well developed models accounting for specific social cognitive processes (Frith & Frith, 2003; Salas & Yuen, 2016) and emotion regulation processes (Gross, 2007; Ochsner et al., 2008). Social processes might not be solely 'hot' as it has been argued that certain processes involved in empathy (taking another's perspective for example) are 'cold' (Chavez-Arana et al., 2018; McDonald, 2013). The metacognitive domain in Stuss' model appears to cover a number of functions that others have argued are further dissociable - e.g. emotion-based decision making, Bechara, Damasio, & Damasio (2000); multi-tasking situations/demands, Shallice & Burgess (1991) - and whilst Stuss implicates (largely right) frontal polar regions in self-reflection, others localise such functions to medial frontal areas (Frith & Frith, 2003; Salas & Yuen, 2016) or systems (Andrews-Hanna, 2012). Along similar lines as Stuss' distinction between cognitive EFs and metacognition, Diamond (2013) draws helpful distinctions between 'core' executive functions (inhibition, working memory, flexibility) that underpin 'higher-order' executive functions such as problem solving, abstract reasoning or complex goal-directed behaviour. Diamond also describes a developmental shift from 'reactive' deployment of EFs in younger children to more anticipatory use of EFs progressively through childhood and adolescence. Taken together, these distinctions between 'core' and 'higher' executive functions, and between 'hot' and 'cold' functions, and Stuss' delineation of anatomically distinct 'frontal functions' perhaps provide the most helpful framework for understanding what is meant by 'frontal', 'executive' and 'dysexecutive'.

1.2.3 Executive functions and everyday life: the problems of ecological validity in assessment and 'far transfer' in rehabilitation

Another common issue in clinical neuropsychology has been the poor ecological validity of traditional 'frontal' / executive functioning tests such as the Wisconsin Card Sort Test (WCST), Trail-making test, Stroop test and Tower of London tests (Ardila, 2008; Manchester, Priestley, & Jackson, 2004), and problems with transfer of clinic-based rehabilitation gains into sustained changes in everyday life.

Ecological validity is the extent to which structured clinical or experimental tasks are associated with behaviours in real-world, everyday or naturalistic settings (Chaytor & Schmitter-Edgecombe, 2003). Two types of ecological validity have been described

(Franzen & Wilhelm, 1996), those concerned with assessments that mimic or match a specific, everyday or naturalistic task (verisimilitude), or those that strongly predict everyday task performance (veridicality). Historically there was an absence of tests specific to EF's that could be used to identify domains of impairment and predict everyday behaviour (Lezak, 1982). This situation has evolved with an increasing number of tests of cognitive EF and of assessment tools designed to be ecologically valid. Burgess and colleagues (Burgess, Alderman, Evans, Emslie, & Wilson, 1998) collected data using 6 different tests of executive functions and looked at which tests were most associated with relative or carer's ratings of everyday 'dysexecutive' difficulties. WCST 'categories achieved' (concept formation) and 'perseverative errors' (shifting), Trails and an ecologically valid multi-component task (the 'six elements test') were associated with informant, but not self-rated everyday dysexecutive problems. The authors concluded that, at the behavioural level, dysexecutive problems following brain injury can be fractionated into 5 domains: inhibition, intentionality (including ability to organise goal-directed behaviour), executive memory (which appeared uniquely associated with WCST 'perseverative errors'), 'positive' emotion (referring to strong positive or negative emotional behaviour such as euphoria or aggression) and 'negative' emotions (referring to poverty of affect). Neither of the latter two factors showed any association with the executive functioning tests, consistent with a 'hot' versus 'cold' distinction of frontal functions. The DEX questionnaire used in this study has been validated in relation to measures of everyday functioning including neurobehavioural symptoms and activities of daily living (Azouvi et al., 2015). The measure has also been subject to Rasch analysis and revisions to improve fit with Stuss' delineation of frontal functions by Simblett and colleagues (Simblett & Bateman, 2011; Simblett, Ring, & Bateman, 2017). There is potential for this revised version to contribute to improvements in both ecological validity and neuropsychological basis of clinical assessment, although to date this has not been evaluated systematically.

Shallice's (Shallice, 1982; Shallice & Burgess, 1991) 'supervisory attentional system' (SAS) model attempts to account for failures in goal-directed behaviour in non-routine novel or complex tasks in everyday life for patients with frontal damage. The model highlights how much of our behaviour is relatively automatic, not requiring control by the 'supervisory system'. However, under certain non-routine (complex or novel) circumstances often characterised by multiple potentially competing sub-goals, the supervisory system is required to optimise outcome in the situation. The model is well illustrated in the paper by Shallice & Burgess (1991) who described 3 patients with everyday difficulties following frontal brain damage. These patients performed at average or above average levels on traditional tests of intellectual functioning and attention or executive functioning. However, impaired performance is described on the 'six elements test' and the 'multiple errands test'. These tasks were designed to be more ecologically valid (based on verisimilitude) and involve development and application of a plan to address a set of competing task goals within a set of rules or constraints. Specific 'supervisory attentional system' processes implicated in non-routine everyday tasks are: identifying and specifying a goal; developing a plan or new temporary behavioural schema; implementing the schema; and monitoring the outcome. Repeated performance of a new behavioural schema will result in it becoming automatic so no longer involving the SAS. The model also highlights how ‘delayed intention markers’ for engagement of the supervisory system at a later point or future circumstance, and level of motivational significance, may also be set. This enables an individual to then interrupt ongoing routine behaviour to change course of action according to an initial plan (e.g. remembering to stop a phone call in time to take the dinner out of the oven). However, the delaying, and later action upon, intentions in everyday life typically occur over much longer time periods than occur within assessments such as the modified six elements or multiple errands tests. A further challenge to ecological validity is, therefore, an individual’s prospective memory ability – their ability to encode both the goal and the type of trigger required to remember to enact the goal at a later time (e.g. if associated with another action, at a specific time or place; Fish, Wilson, & Manly, 2010). Related to this is the individual’s awareness of their strengths and difficulties in relation to a specific task or situation, and ability to draw on this to adapt behaviour accordingly (Crosson et al., 1989; Toglia & Kirk, 2000) for example by thinking to set a reminder or write a note.

The SAS model has also informed rehabilitation, particularly problem solving and goal management type approaches (Evans, 2009; Krasny-Pacini, Limond, et al., 2014; Levine et al., 2000, 2011; Miotto, Evans, de Lucia, & Scaff, 2009). The original formulation of these approaches is derived from problem solving therapy (Bell & D’Zurilla, 2009), and takes patients through a systematic process of identifying the goal, weighing options and selecting a method for achieving it, identifying the steps, then learning and implementing these. Group-based attention and goal management interventions also incorporate development of self-awareness through discussion with others, and self-monitoring of problems and successes and factors impacting everyday performance such as mood and fatigue. Addressing the issue of ‘far transfer’ from rehabilitation to everyday life, the idea of adding ‘content free’ cues to maintain attention to the goal and sub-steps has been explored with some success (Fish et al., 2007; Gracey, Fish, et al., 2016; Manly et al., 2004; Tornås et al., 2016).

Network accounts as described earlier (Andrews-Hanna, 2012; Corbetta & Shulman, 2002) also provide a possible explanation for poor predictive value of assessments, and difficulty with transfer of rehabilitation into everyday life. Difficulty managing demands in everyday life might arise secondary to focal damage in any aspect of a relatively wide network required to perform a task in everyday life, or in white matter connectivity / integrity, which may or may not be so clearly associated with performance on tests sensitive to focal frontal cortical damage, such as the Stroop or Controlled Oral Word Association Test. This would account for the good sensitivity but poor specificity of traditional EF tests for detecting everyday difficulties. Similarly, specific cognitive EF skills (or their improvement through rehabilitation) may be necessary but not sufficient to achieve optimal performance in everyday life.

1.2.4 Models of frontal systems and emotional functioning

Neuroscience models of affect regulation highlight the role of the amygdala in processing emotional significance and emotional learning, and connections with frontal areas relating to orientation of attention and inhibition of reactions (e.g. Ledoux, 2000; Phelps & Anderson, 1997; Pessoa, 2010). In addition to these relatively automatic processes, other cognitive EF processes involving medial and lateral prefrontal areas have been implicated in consciously mediated emotion regulation processes such as verbal reappraisal or labelling.

Ochsner & Gross's (2005) review of neurosciences research into the down-regulation of emotional reactions identified two broad categories of process, both of which implicate executive processes: attentional control (targeting the automatic orientation of attention to affectively salient stimuli, involving orbito-frontal cortex (OFC), anterior cingulate cortex (ACC), medial and lateral PFC) and cognitive control (targeting the anticipation and/or later (re)appraisal of the stimulus, also involving ACC, dorsal medial and ventral lateral PFC). In a later review paper (Ochsner et al., 2008) describe how verbal labelling and reappraisal appear to reduce activation in amygdala (detection and encoding) and insula (affective experience) emotional response areas. Attempts at regulation through suppression however resulted in increased insula reactivity (Goldin, McRae, Ramel, & Gross, 2008). They conclude that a number of 'cold' cognitive processes are involved in effective emotion regulation through verbal reappraisal, including dorsal PFC areas (involved in working memory), ventral inhibitory processes, ACC (involved in monitoring and direction of attention), and dorso-medial PFC (involved in reflection on one's own or other's emotional states).

Hofmann, Schmeichel, Friese, & Baddeley (2010) set out a similar account for the role of the component functions of the central executive of working memory in emotional self-regulation. They propose that an emotional reaction may enter working memory if salient or strong enough to recruit attentional resources. The supervisory attentional system may be called upon in more demanding situations where a new behavioural schema needs to be developed to successfully override any impulses and maintain focus on goal-directed behaviour over time. Evidence in support of this comes from Lindstrom & Bohlin (2012) who studied working memory performance manipulated with threat-relevant stimuli. They interpret their findings as consistent with the ‘dual-competition framework’ model (Pessoa, 2009), which states that if the emotional stimulus is not pertinent to the goal, it will compete for limited resources and impair capacity of the higher-level goal directed system. However, if there is congruence between an emotional stimulus and the goal, emotional salience will enhance this higher-level cognitive functioning.

Williams & Thayer’s (2009) brief review similarly argues that individual differences in ‘cold’ EFs are associated with individual variability in a range of stress-regulatory processes including susceptibility to exposure to stressful events, magnitude of stress reaction, recovery from stress and restoration. They draw upon findings of impaired EF’s in research with patient populations and genotypic variation in specific EF abilities (notably the Attention Network Test and inhibition, switching and updating). They argue this genotypic variation links variations in dopaminergic system responses to stress, parasympathetic vagal nervous system modulation of PFC activity and EF abilities and stress-related early life experiences to behavioural outcomes in terms of stress, health and wellbeing. Studies of people with brain injury that address these specific mechanisms is lacking although Gyurak et al. (2009), comparing people with frontal deficits secondary to dementia and healthy controls, found verbal fluency scores (but not other EF tests) were predictive of emotional regulation in trials where participants were warned about the stimulus (but not un-warned trials). This seems partially consistent with Ochsner & Gross' (2005) view of EF involvement in later controlled, rather than earlier automatic, downregulation of emotion.

There is also growing interest in parasympathetic vagal nervous system activation involvement, as reflected in heart rate variability (HRV), in both 'hot' and 'cold' frontal processes. The vagally mediated parasympathetic nervous system is thought to be activated under conditions of novelty, challenge or threat and is indicated by reduced HRV, whereas higher HRV arises under conditions of social engagement and safety (Porges, 2009; Thayer & Lane, 2000). Evidence is generally supportive of this claim. Studies indicate that greater resting HRV is associated with better performance on traditional executive tests such as WCST, n-back and Stroop colour word interference tests (Hovland et al., 2012; Thayer, Hansen, Saus-Rose, & Johnsen, 2009). In addition to these 'cold' cognitive functions, associations have also been identified between HRV and social and emotional processes such as depression (Kemp et al., 2010), perseverative cognition (such as worry or rumination; Ottaviani et al., 2016); emotion identification, social inference, empathy and alexithymia amongst people with TBI (Francis, Fisher, Rushby, & McDonald, 2016a), trait reactivity and executive functions (Bailey, Potter, Lang, & Pisoni, 2015). Byrd, Reuther, McNamara, DeLucca, & Berg (2015) investigated HRV in children and adults exposed to varying demands of specific inhibition, working memory and problem-solving tasks. In both children and adults, all tasks except problem solving were associated with on-task HRV suppression, which the authors interpret as indicating that vagally mediated regulatory demands only arise under conditions of time-response pressure, and not frontal tasks per se.

Individual differences in parasympathetic response to specific types of subjective (rather than purely cognitive) challenge such as time-response pressure, might therefore modulate capacity to apply higher-level regulatory skills. In addition to improved regulation of negative affective states, increased HRV is thought to be associated with higher levels of wellbeing (Kemp, Koenig, & Thayer, 2017) comprising positive emotional states, sense of mastery and social connection (Seligman, 2018), and broadening and flexibility of attention (Fredrickson, 2004).

Therefore, in everyday life, deviation from behaviours observed in clinic-based assessment or rehabilitation might arise if an emotionally salient aspect of a situation, with goals that are incongruent with, or represent threat to the person's overarching intentions, captures attentional or regulatory resources. Against this, positive psycho-social processes such as social connection, sense of mastery and positive affect might also optimise frontal functions, as might congruency between intentions and a goal of high motivational value. The behavioural outcome will not simply be predicted by performance on assessments of EF implicated in self-regulation but would also need to consider the salience of the real-world emotional 'trigger', the salience or motivational value of the person's intention, and their capacity to manage any competing demands that arise. This itself might be complicated by different regulatory processes being implicated across early/automatic regulation and later conscious or strategic attempts to cope or adapt effectively.

1.2.5 Models of frontal systems and social processing

Spikman et al (2012) defined social cognition as ‘comprising the capacities of individuals to understand the behaviour of others and to react adequately in social situations’. The concept covers several separate functions. Crick & Dodge’s (1994) model of social information processing in children sets out a series of processing steps in response to a social ‘cue’ or stimulus: 1. Encoding the to-be-perceived stimulus or social cue, 2. Representing and interpreting the social cue, 3. Searching and generating responses, 4. Deciding upon or selecting the appropriate response (based on representation of potential consequences), and 5. Enactment of the social response, in which implementation of the social skill as well as monitoring and self-regulation are required. Similarly, Frith & Frith (2003) set out social processes in a dyadic interaction as follows: 1. Predicting the intentions of others, 2. Aligning to the other’s subjectivity, and 3. Influencing the subjectivity and behaviour of the other. Frith & Frith (2010) argue for the parallel operation of a ‘cold’ mentalisation process which can represent the mental states or goals of others (theory of mind) as well as an affectively ‘hot’ mirroring system which enables subjective experiencing of, and empathy with, another’s emotional state.

A further model of ‘hot’ social and behavioural regulation has been proposed by Damasio and colleagues (Bechara, Damasio, et al., 2000; Damasio, Tranel, & Damasio, 1991). Based on the study of individuals with focal orbitofrontal or ventromedial lesions (Bechara, 2004) they have proposed a model of emotion-based decision making in which interoceptive signals (‘somatic markers’) learned through previous long and short-term reward / punishment contingencies, guide choice of different response options in complex, fast moving situations such as social interactions. The automatic 'marking' of the possible risk / reward associated with conscious consideration of a choice within working memory has also been termed the 'what if' system (Bechara, Damasio, Damasio, & Anderson, 1994). The 'somatic marker' hypothesis has been tested using a specific paradigm, the Iowa Gambling Task (IGT). This purports to measure the ability to identify and select more favourable responses over time as a result of different reward and punishment contingencies. Bechara (2004) argues that patients with ventromedial PFC lesions have a specific deficit in decision-making requiring weighing of longer-term consequences, which is associated with social disturbances in the context of retained ability to perform 'cold' executive tests. Whilst there is a clinical coherence to the somatic marker hypothesis, and some patient-based evidence that these social difficulties can be made sense of in this way, a number of authors have critiqued both the model and the research (Dunn, Dalgleish, & Lawrence, 2006; Maia & McClelland, 2004).

Taking a developmental approach to understanding these issues (Beauchamp & Anderson, 2010)'s 'SOCIAL' model highlights underpinning biological as well as 'hot' and 'cold' cognitive and social processes and environmental factors. Consistent with ideas emerging in the adult literature, the model indicates that 'cold' cognitive processes such as control of attention, inhibition, and self-monitoring, likely support specific social processes and skills such as theory of mind, empathy and mentalisation. In children, development of executive and social cognitive skills typically occurs trough childhood and into early adulthood as cortical myelination moves from anterior primary motor cortex and frontal polar regions to dorsolateral prefrontal cortex

(Gogtay et al., 2004), consequently younger age at injury might be associated with delayed onset of higher executive or social difficulties (Chapman, 2006).

1.2.6 Conclusion

In conclusion, frontal functions appear to comprise distinct ‘cold’ executive processes and ‘hot’ emotion and social regulation processes consistent with (Chan et al., 2008; Takeuchi et al., 2013). A simplified schematic summarising organisation of frontal functions is presented in Figure 1. The overarching integrated sets of processes that include frontal and other brain areas (including parietal and temporal and limbic structures, and the parasympathetic nervous system) are described as frontal systems, and ‘frontal functions’ as specific types of hot or cold processing ability. The term ‘executive function’ relates to ‘cold’ lower-level, component cognitive processes such as attention control, and functions that comprise executive working memory (e.g. shifting, inhibiting, updating). Specific social and emotional functions have also been identified such as use of interoceptive signals to guide decision-making, a ‘cold’ perspective taking aspect of empathy or mentalisation and ‘hot’ mirroring or feeling for the other person. The metacognitive or higher-order frontal functions refer to a set of supramodal processes involved in non-routine tasks with multiple or competing sub-goals, requiring integration of component processes or conscious reflection, potentially across ‘hot’ and ‘cold’ executive, social and emotional processes. This is consistent with contemporary accounts that blend supramodal and component process accounts of frontal functions (Diamond, 2013; Miyake & Friedman, 2012; Stuss, 2011). The literature appears to refer to metacognition in terms of integration of core executive processes into higher order functions required for management of complex goal directed behaviour. However, in everyday life such metacognitive skills might also conceivably reach across executive, social and emotional systems. Understanding certain categories of function as sub-served by networks of component functions provides a basis for the partial associations seen between some component processes and everyday life, but poor predictive validity of tests. Potential interactions between 'hot' and 'cold' systems also provides a basis for understanding issues of poor transfer of rehabilitation into everyday life.

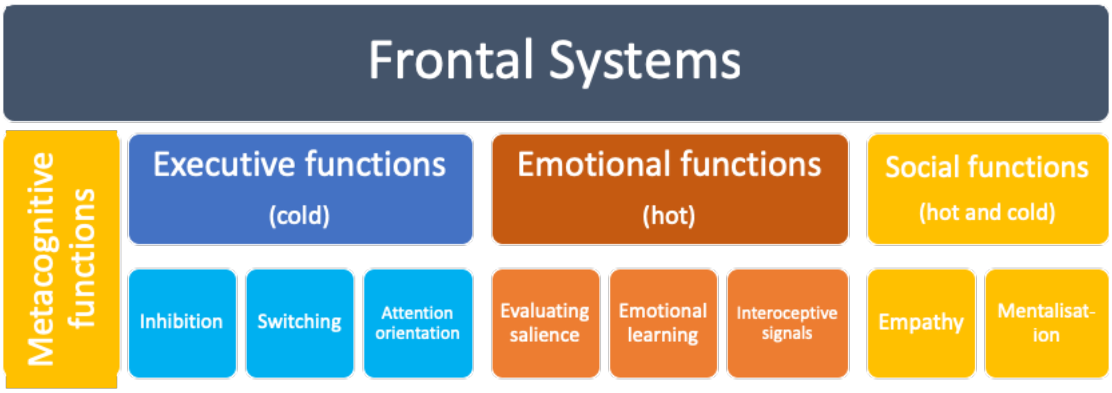

Figure 1: Simplified schematic illustrating examples of core 'hot' and 'cold' executive, social and emotional functions within a hierarchical structure.

1.3 Frontal systems and 'everyday' psychosocial outcomes in people with ABI

The following sections will explore the evidence relating specific frontal functions to everyday participatory outcomes. In addition to practical or participatory outcomes, social and emotional outcomes will also be covered as these are considered strongly associated with everyday outcomes (Azouvi et al., 2015; Mauri, Paletta, Colasanti, Miserocchi, & Altamura, 2014). Given the relevance of rehabilitation to longer term participatory outcomes, rehabilitation and other supportive healthcare interventions will be considered an important aspect of everyday life following brain injury (Nalder et al., 2012). For the purposes of this thesis, when considering 'everyday life' the focus will be particularly on those aspects that require some degree of 'frontal systems' involvement, in that they present social, emotional or cognitive demands, rather than the more automatic or routine aspects (Takeuchi et al., 2013).

1.3.1 Cognitive factors associated with everyday participatory outcomes

A relatively large number of studies have attempted to identify predictors of psychosocial and participatory outcomes following stroke and brain injury, with many variables and outcomes included. In general, pre-injury factors, severity of injury, language impairment as well as social and emotional issues and domains of cognitive functioning emerge as significant predictors of outcomes, (Barker-Collo & Feigin, 2006; Sherer et al., 2015) consistent with the World Health Organisation-International Classification of Functioning (WHO-ICF; Üstun, Chatterji, Bickenbach, Kostanjsek, & Schneider, 2003).

Some studies particularly focus on EF or social processing variables relevant to outcomes. Ponsford, Draper and Shönberger (2007) found several cognitive measures including EF were associated with poorer outcome although digit-symbol coding, a measure of speed of processing (SoP), was the strongest cognitive predictor. In terms of Stuss' (2011) model, SoP could be related to impairment of the 'energization' aspect of frontal functioning, particularly if performance indicated a relatively rapid drop in performance after a promising start, or a failure to initiate action. However, it is also possible that SoP is a marker for a higher level of global impairment, or for diffuse axonal injury disrupting white matter connections and therefore network integrity. A later study, employing a wider range of cognitive assessments, found the addition of cognitive (memory and EF) measures significantly improved predictive models of psychosocial outcomes, in particular the Behavioural Assessment of the Dysexecutive Syndrome (BADS) Zoo Map test, Trails-B and Wechsler Memory Scale (WMS) III working memory index scores (Spitz, Ponsford, Rudzki, & Maller, 2012). However, Wood & Rutterford (2006) found that, of several cognitive predictors, working memory emerged as the only measure significantly associated with community integration.

Mani, Cater, & Hudlikar (2017) conducted a systematic review of studies looking at associations between cognition and return to work post TBI. They conclude that EFs emerged as the most common significant predictor, with other domains of cognitive functioning including attention and memory also contributing, echoing findings in stroke (Barker-Collo & Feigin, 2006). However, the review did not contrast specific domains or measures of executive functioning. Ownsworth & McKenna's (2004) review of cognitive predictors of return to work also highlights the contribution of EF. They suggest the impact of cognitive difficulties on vocational outcomes might be mediated by metacognitive (associated with awareness and application of strategies to manage cognitive problems) and emotional or motivational variables in addition to access to rehabilitation/therapy and other factors such as financial incentives or environmental supports. Yeates et al. (2016) also explored executive and social cognitive predictors of workplace supervisors' evaluation of the work-related social skills of people with brain injury. The study found that of the social and executive cognitive measures included, mentalisation (ability to hold the states and intentions of others in mind; the ‘Faux Pas’ test) and plan formulation and implementation (BADS modified six elements task) emerged as significant contributors to supervisors’ evaluations of social skills. It has also been argued that variability in job demands such as complexity can impact upon the extent to which EF difficulties are associated with outcome (Matheson, 2010).

1.3.2 Factors impacting engagement in and response to rehabilitation

Frontal dysfunction might impact participation directly, or indirectly by affecting ability to access services or benefit from interventions. A review of studies of rehabilitation outcome predictors by Whyte, Skidmore, Aizenstein, Ricker, & Butters, (2011) identified various ways in which cognition might impact upon rehabilitation outcomes, suggesting capacity for full engagement or participation in rehabilitation might be lower for those with deficits in EF and self-awareness. They also suggest deficits in flexibility, learning and prospective memory are likely to hinder attempts at compensatory adaptation, whilst deficits in sustained attention, processing speed and learning might hinder efforts at improving functioning through intensive practice. From this model they then identify intervention targets that might enhance response to rehabilitation amongst those with these deficits, such as metacognitive strategy training to address EF and self-awareness issues (Skidmore et al., 2011). Similar arguments that longer term favourable outcomes are dependent on an individual’s ability to adapt flexibly to their new circumstances, problem solve, develop awareness of their difficulties, cope in an active problem-oriented manner and remember to implement required strategies at the appropriate time have been made (Anson &

Ponsford, 2006; Cicerone et al., 2011; Crosson et al., 1989; Fish et al., 2010; Judd & Wilson, 2005).

The identification of a range of metacognitive, executive and emotional functions likely associated with rehabilitation outcomes (including self-awareness, learning, mood or motivation and prospective memory) gives a clear indication of the need to include relevant evidence-based interventions within a wider rehabilitation effort to improve far-transfer of interventions from clinic to everyday life. Increasingly, interventions addressing goal management skills (concerned with improving goal directed behaviour, problem solving and self-monitoring) are being adapted to include mindfulness (Alfonso, Caracuel, Delgado-Pastor, & Verdejo-García, 2011; Levine et al., 2011), implicit or errorless learning (Bertens, Kessels, Fiorenzato, Boelen, & Fasotti, 2015), emotion regulation strategies (Tornås et al., 2016), self-awareness (Cantor et al., 2014), autobiographical memory supports (Cuberos-Urbano et al., 2016) and periodic alerts or reminders (Gracey et al., 2017; Tornås et al., 2016).

In summary, once global levels of impairment, and severity of injury have been considered, EF's emerge as a significant cognitive predictor of a range of outcomes for people with stroke and other ABIs, alongside other factors such as spatial neglect / inattention (in stroke), memory, communication and speed of processing. In terms of rehabilitation, deficits in EF may present a barrier to transfer of strategies or learning into everyday life and may also compromise other health behaviours related to outcome. Social processing skills, mood and motivation are also likely to contribute to outcomes indicating a dynamic, unfolding trajectory in which both ‘hot’ and ‘cold’ processes associated with frontal functions may play a key role.

1.3.3 Evidence for frontal systems associated with emotional outcomes following brain injury

Poor emotional outcomes are a significant issue for 20-40% of people post stroke or brain injury (Ayerbe, Ayis, Wolfe, & Rudd, 2013; Ayerbe, Ayis, Crichton, Wolfe, & Rudd, 2014; Hart et al., 2016; Matheson, Wohl, & Anisman, 2009; Osborn, Mathias, & Fairweather-Schmidt, 2014) with depression and suicidality appearing to increase over time post injury (Fordyce, Roueche, & Prigatano, 1983; Teasdale & Engberg, 2001; Tsaousides, Cantor, & Gordon, 2011). Children are also especially vulnerable to negative emotional and behavioural outcomes including anxiety, depression, personality changes and attention deficit hyperactivity disorder (ADHD; Max et al., 1997, 2004, 2011) with associated peer relationship problems (Tonks, Yates, Williams, Frampton, & Slater, 2010). Causes of emotional distress are likely complex and multi-factorial. Gainotti (2001) describes how emotional consequences might arise from ‘psychological’ effects relating to losses and changes, as well as organic effects or damage to brain systems involved in emotion regulation. Goldstein (1952) described 3 possible routes to changes in everyday life following brain injury: the potential direct effects of brain damage on abilities; the potential for a strong negative emotional reaction to these changed circumstances which he described as the ‘catastrophic reaction’; and further loss of abilities through attempts to avoid the ‘catastrophic reaction’, compounding the effects of the initial injury. These ideas have been further developed in our models (Gracey, Evans, & Malley, 2009; Gracey, Longworth, & Psaila, 2016; Gracey, Olsen, Austin, Watson, & Malley, 2015; Wilson, Gracey, Evans, & Bateman, 2009), which emphasise the two-way interaction between stresses or challenges in everyday life (and regulation of these responses), enduring sense of self and attempts to maintain continuity of self, and longer-term coping, in turn impacting everyday life (for example through loss of social networks or employment opportunities). Gracey, Olsen, et al. (2015) extended these ideas to the understanding of emotional outcomes in childhood ABI. Emotional issues might arise directly from the injury, from the child's appraisals of changes and losses, and appraisals and coping responses of others in the child's context which might significantly impact upon development through disrupted attachment relationships and / or cognitive scaffolding for neurodevelopment.

Hackett et al (2005)'s systematic review of predictors of post stroke depression found that out of many possible predictor variables studied, level of physical disability, stroke severity, cognition and social isolation were consistently associated with level or presence of post-stroke depression. Ayerbe et al (2013) drew similar conclusions in their more recent review and meta-analysis. In TBI, the studies of Malec, Brown, Moessner, Stump, & Monahan (2010) and Ownsworth et al. (2011) both highlight severity of injury as important, in contrast to (Spitz, Schönberger, & Ponsford, 2013), and appraisals of post-injury ability as related to post-injury depression, with accuracy of perceived functioning at discharge from hospital a possible moderating factor.

Focusing on cognitive predictors, Jorge & Robinson (2004) found greater impairments in problem solving and cognitive flexibility in people with major depression and TBI, and that EF was more significantly impaired in TBI depressed than non-depressed, although it emerged that depression itself might contribute to the deficits in problem solving, rather than vice versa. Involvement of EFs in anxious versus non-anxious people with TBI have also been found by Gould, Ponsford, & Spitz (2014). Anxiety disorders were associated with reduced processing speed, working memory and EF, with EF best differentiating anxious from non-anxious TBI participants, but again direction of causality is not clear.

Given the theoretical links between emotion activation and regulation, and ‘cold’ cognitive working memory or supervisory attentional processing discussed previously, there may also be a role for cognition, coping style and appraisal in predicting community functioning and emotional outcomes. The association between working memory (but not other cognitive factors) and emotional outcomes identified by Wood & Rutterford (2006) appeared accounted for by self-efficacy, suggesting greater working memory deficit might directly impact unfavourably on community outcomes, and over time, the resulting loss of confidence reduces self-efficacy, further impacting emotional outcomes. Spitz et al. (2013) found both memory and executive functions were associated with depression post-injury, alongside less adaptive and more maladaptive coping styles.

Taken together, these findings suggest more severe injury and poorer self-awareness of deficits early post-injury may increase vulnerability to perceptions of loss of function / greater changes early post-discharge, which in turn impact upon self-efficacy and emotional outcomes. Cognitive factors, particularly working memory, other EFs and speed of processing appear associated with emotional outcomes, possibly through effects on participatory outcomes, and resulting lowered self-efficacy and increased awareness of losses and changes. Considering these challenges, less adaptive and more maladaptive coping styles are further associated with worse emotional outcomes. Despite a theoretical basis for specific executive deficits contributing to emotional vulnerabilities, few studies were designed to address this question. However, some evidence suggests possible direct contribution of EF or self-regulatory deficits to emotion regulation and emotional outcomes. There is also some indication that where depression and EFs appear associated, it is the depression that at least partially contributes to EF (problem solving) difficulties. Therefore, whilst there is some lack of certainty regarding directions of causality and specific mechanisms involved, specific frontal functions appear at least indirectly, and possibly directly associated with greater vulnerability to poorer emotional outcomes.

1.3.4 Relationships between social cognitive processes and social outcomes

Several studies have explored social cognitive processes after ABI and factors associated with social outcomes. Spikman et al. (2012) evaluated social cognitive abilities relating to emotion identification, ‘cold’ theory of mind, and ‘hot’ empathic ability, alongside non-social cognitive tests in people with TBI and controls. They found a substantial level of impairment in three domains of social cognitive functions in TBI (emotion identification, theory of mind and empathy) which was not attributable to performance in other cognitive tasks. Struchen et al.’s (2008) study focused on social cognitive and executive functions in the prediction of occupational and social outcomes following TBI. They found that executive functioning contributed significantly to occupational and social outcomes, with social cognitive tasks accounting for unique variance in outcomes, albeit at a lower level than EF. Unlike Spikman et al, they did find correlations between ‘cold’ EF’s and the social cognitive tasks they employed, perhaps attributable to their use of the La Trobe questionnaire as an outcome measure, which includes items relating to a range of cognitive and social communication problems. The conclusion here is that both ‘cold’ EFs and ‘hot’ social cognitive processes contribute to occupational and social outcomes, and that whilst there is some association between EFs and social processes, social processes also make a unique contribution to outcomes.

In the study of Yeates et al. (2016) the greater contribution of ‘cold’ executive functioning over specific ‘hot’ social cognitive tests (other than mentalisation), to perceived social skills was not in keeping with the authors’ hypotheses that a wider range of social cognitive abilities (rather than ‘cold’ executive skills) would be the stronger predictors of workplace social skills. In the context of study limitations, the authors suggest that supervisors may perceive, and rate workplace related social interaction issues secondary to executive deficits, for example seeing poor organisational skills in terms of how respectful or considerate of others the person is, rather than as a ‘cognitive deficit’. An alternative account is that many aspects of social communication can be considered as complex goal-directed behaviours sitting under wider executive control (Struchen et al, 2008), or that management of the demands of meeting work related goals in a social interactional context require metacognitive integration of component ‘cold’ and ‘hot’ frontal functions.

Social competence following childhood brain injury is significantly lower than typically developing children (Rosema, Crowe, & Anderson, 2012) and children with brain injury are more likely to have problems with peer relationships and loneliness (Ross, McMillan, Kelly, Sumpter, & Dorris, 2011; Tonks et al., 2010) with these problems increasing over 1-2 years post injury (Ryan et al., 2016). In children who sustain brain injury, the age and developmental stage at which an injury is sustained may have developmentally specific effects, and as previously discussed the child may grow into deficits as the downstream effects of foundation skills or neuroanatomical disruption become apparent. In keeping with these ideas, research indicates contributions of socio-emotional processes (Anderson et al., 2013; Ganesalingam, Sanson, Anderson, & Yeates, 2006; Rosema et al., 2012; Tousignant et al., 2018) and cognitive factors including executive functions and speed of processing (Anderson et al., 2013) to social competence in childhood brain injury. Parent or family factors also emerge as significant predictors of social and emotional outcomes (Rosema et al., 2012; Shari L Wade et al., 2011). In adolescents, loss of 'hot' empathy abilities as shown in sensitivity to others appears to be a particular issue (Tousignant et al., 2018). Whilst Rosema et al (2012) and Ciccia, Beekman, & Ditmars (2018) highlight weaknesses in research in this field, including need for more work on age or developmental stage at injury, measures of social and emotional processing domains, and environmental factors the research is indicating potential targets for intervention including 'hot' and 'cold' processes and family functioning (Chavez-Arana et al., 2018).

1.3.5 Summary and conclusions

The preceding review concludes that anatomically and neuropsychologically dissociable yet linked frontal functions are of key relevance in successful adaptation to life post-injury and potential to benefit from rehabilitation. Whilst many factors impact on outcomes (including severity of injury, communication and memory), it could be argued that the frontal brain areas and associated systems and functions represent a 'common pathway' by which goal directed and social behaviours are expressed, and responses to novelty, challenges or social and emotional salience, in the ongoing stream of life post-injury are processed and regulated. Evidence for benefits of interventions for frontal deficits transferring into everyday life is limited perhaps because the deficits people have are the very skills needed for better adaptation and outcome. These include use of problem-solving ability in the face of stresses or inflexibility in thinking, abilities which themselves might interact with emotional state. Again, however, the evidence concerning the interplay of processes such as coping style and executive functioning is mixed.

Children with acquired brain injury are particularly vulnerable to mental health problems and peer relationship changes likely due to disruption of development of executive, social and emotional, processes although further testing of these hypothesised relationships is required. In adults with ABI problems with executive functioning appear at least partially associated with negative emotional outcomes, although mechanisms remain unclear. However, the evidence base for psychological therapies to help people adjust and adapt is mixed (e.g. Waldron, Casserly and O'Sullivan, 2013) and further work is required to develop clinical models to guide intervention and identify viable adaptations to therapy to address cognitive problems that can affect memory and problem solving (Gracey, Ford, & Psaila, 2015; Ownsworth, Gooding, & Beadle, 2018).

The work reviewed in this chapter has identified several ways in which frontal functions and everyday outcomes might influence each other both directly (in terms of cognitive limitations to participation and rehabilitation) but also through multiple indirect routes. This review also highlights methodological issues and lack of theoretical coherence to help select and test variables and relationships, with clinical studies generally failing to test the specific models being developed in cognitive and affective neuroscience research. This is particularly so with the conceptualisation and measurement of specific aspects of frontal functions such as metacognition, emotionally 'hot' processes as well as interactions between coping style and executive functions and metacognition. These challenges of ecological validity of assessments and of transfer from rehab to everyday life could be accounted for by considering 1. Improved measurement of specific component processes, 2. Testing model-based hypotheses regarding interactions between component processes, including direct, indirect and bidirectional relationships, and relationship to everyday outcomes and 3. Evaluation of processes and mechanisms that facilitate far transfer of rehabilitation from the clinic into everyday life. Addressing these gaps might contribute to improving prediction of difficulties in everyday life from clinical neuropsychological assessment, and generalisation of rehabilitation to everyday life.

1.4 Questions being addressed in this thesis

This thesis aims to address some of the challenges to understanding and rehabilitating problems arising due to frontal systems damage. The specific overarching question is as follows: Can specific 'hot' or 'cold' neuropsychological frontal functions be identified that are associated with social processes, or social, emotional and participatory indicators of everyday functioning? The question will be addressed through a specific focus on:

1. Cognitive and emotional frontal functions associated with social relationship outcomes in children and young people who sustained an ABI at different ages (Chapter 2, Paper 1).
2. Cognitive functions associated with social-emotional functions in adults with TBI compared with non-injured controls (Chapter 3, Paper 2).
3. Executive functions associated with emotional outcomes (Chapters 4 and 5, papers 3 and 4).
4. Use of periodic alerts and brief goal management training to improve achievement of everyday intentions in adults with everyday executive difficulties following ABI (Chapter 5, paper 4).

CHAPTER 2

2 Which specific frontal functions are associated with social peer relationship problems following childhood acquired brain injury?

Gracey, F, Watson, S, McHugh, M, Swan, A, Humphrey, A and Adlam, A. (2014) Age at injury, emotional problems and executive functioning in understanding disrupted social relationships following childhood acquired brain injury. *Social Care and Neurodisability* 5(3), 160-170.

2.1 Introduction to the paper

Prior research clearly indicates associations between social outcomes (such as peer relationships, or social behaviour regulation) and frontal functions in children with brain injury. However, there is variation across studies as to which functions have emerged as significant, including 'cold' EFs such as working memory, as well as solely 'hot' self-regulatory or social processes. Studies have also varied regarding time since injury and age / developmental stage of the sample. In this paper we sought to further explore associations between age at injury, various 'hot' and 'cold' frontal functions (as rated by parents on the Behavior Rating Inventory for Executive Function, BRIEF; Gioia, Isquith, Guy, & Kenworthy, 2000) and peer relationship problems in a clinical sample covering a wide range of age at injury and age at assessment. We also sought to further compare mental health needs of children with brain injury referred for rehabilitation with those referred for mental health problems but no brain injury.

Age at injury, emotional problems and executive functioning in understanding disrupted social relationships following childhood acquired brain injury

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Abstract

Purpose – *Clinically significant childhood acquired brain injury (ABI) is associated with increased risk of emotional and behavioural dysfunction and peer relationship problems. The purpose of this paper is to determine how emotional and peer related problems for children with ABI compare with those of children referred to mental health services, and to identify clinical predictors of peer relationship problems in a heterogeneous sample typical of a specialist community rehabilitation setting.*

Design/methodology/approach – *Participants were 51 children with clinically significant ABI (32 traumatic brain injury; 29 male) referred for outpatient neuropsychological rehabilitation. Emotional, behavioural and social outcomes were measured using the Strengths and Difficulties Questionnaire (SDQ), and executive functioning was measured with the Behaviour Rating Inventory of Executive Functions. Correlational analyses were used to explore variables associated with peer relationships. A subgroup ($n = 27$) of children with ABI were compared to an age and sex matched mental health group to determine differences on SDQ subscales.*

Findings – *The SDQ profiles of children with clinically significant ABI did not significantly differ from matched children referred to mental health services. Time since injury, peer relationship problems, metacognitive, and behavioural problems correlated with age at injury. These variables and SDQ emotional problems correlated with peer relationship problems. Linear multiple regression analysis indicated that only metacognitive skills remained a significant predictor of peer relationship problems, and metacognitive skills were found to significantly mediate between age at injury and peer relationship problems.*

Research limitations/implications – *The study confirms the significant effect of childhood ABI on relationships with peers and mental health, those injured at a younger age faring worst. Within the methodological constraints of this study, the results tentatively suggest that age of injury influences later peer relationships via the mediating role of poor metacognitive skills within a heterogeneous clinical sample.*

Originality/value – *This is the first study to examine the roles of emotional, behavioural and executive variables on the effect of age at injury on peer relationship problems in a sample with a wide range of ages and ages of injury.*

Keywords *Children, Acquired brain injury, Mental health problems, Executive function, Social outcomes, Peer relationships*

Paper type *Research paper*

Introduction

Sustaining an acquired brain injury (ABI) predominantly resulting from traumatic injury (TBI), brain infections, vascular problems, hypoxia, surgery or tumour, in childhood is known to be associated with a wide range of poor outcomes more than six months post-injury and across the range of severity. In recognition of the uncertain relationships between injury severity and psychosocial outcomes, guidelines on rehabilitation of ABI have opted for use of the term "clinically significant" ABI, reflecting the heterogeneous population seen in rehabilitation services (Turner-Stokes, 2003). Poor outcomes following childhood ABI have been found in the domains of cognition (Anderson *et al.*, 2005; moderate-severe TBI), educational attainment (e.g. Ewing-Cobbs *et al.*, 2004; moderate-severe TBI; Hawley *et al.*, 2004; mild-moderate-severe TBI), emotional and behavioural problems (Max *et al.*, 1998, 2005; moderate-severe TBI). Whilst children typically recover well from mild TBI (mTBI; Carroll *et al.*, 2004), a proportion also experience enduring psychological changes impacting on participation (Yeates and Taylor, 2005).

Research indicates increased likelihood of poor social relationship outcomes following childhood ABI, although conclusions vary about the mechanisms by which these poor outcomes are linked with the injury. Limond *et al.* (2009), found that quality of life was significantly lower in 13 times more ABI children up to five years post-injury than a normative sample and there were significant emotional, cognitive, and behavioural problems in almost half of the brain injured sample. For those sustaining moderate-severe TBI, or multiple mTBIs, difficulties such as these have in turn been associated with poor vocational outcomes and increased risk of criminal behaviour in adulthood (Williams *et al.*, 2010). Ross *et al.* (2011) investigated the domains of friendship quality (as measured by the Friends Quality Questionnaire – Revised (FQQ-R; Parker and Asher, 1993), rates of loneliness (as measured by the Loneliness and Social Dissatisfaction Scale; Asher and Wheeler, 1985), and general psychosocial functioning (as assessed by the Strengths and Difficulties Questionnaire, SDQ; Goodman, 2001). Children with moderate to severe TBI aged seven to 13 years were rated as having much greater difficulties than matched non-injured controls, particularly relating to emotional problems and attention/hyperactivity. Ross *et al.* did not find evidence of significant difficulties in peer relationships (SDQ, FQQ-R) or loneliness, but the mean age of the TBI group at time of assessment was just over ten years, leading to a hypothesis that social vulnerabilities might not be evident until later in adolescence. Consistent with this hypothesis, Tonks *et al.* (2010) found evidence of significant peer relationship problems (assessed using the SDQ) amongst slightly older brain injured children aged nine to 15 years compared with healthy controls. These difficulties with peer relationship problems and emotional distress did not significantly differ from children with mental health problems. However, failure to include a matched comparison group means the findings might be confounded by age or sex.

Studies looking at factors influencing these negative social outcomes amongst children with moderate-severe TBI, or other clinically significant ABI, have included executive functioning (flexibility, inhibition, behaviour regulation), in addition to specific social processing deficits (e.g. social inference or theory of mind) and contextual factors (age at injury, developmental stage, parenting factors). Yeates *et al.* (2004) found that long-term social outcomes (mean follow up of four years) following moderate-severe paediatric TBI were accounted for, in part, by executive functions, pragmatic language, and social problem-solving. The findings were consistent with those of Dennis *et al.* (2001) who reported that deficits in executive function are predictive of social outcome following paediatric TBI. However, Ganesalingam *et al.* (2007) found deficits in self-regulation of emotion, but not other cognitive executive problems to mediate between history of brain injury and negative social outcomes in a sample of children aged between six and 11 years with and without brain injury.

Anderson and colleagues (Anderson *et al.*, 2009; Anderson and Catroppa, 2005) have considered the impact of developmental context on injury outcomes, arguing that problems relating to executive functions do not emerge until later in childhood or early adulthood, in line with both brain maturation and social/cultural expectations of increased independence. Support for this was found by Sesma *et al.* (2008), who showed that executive function difficulties increased over a 12-month period post-injury, in a group of children aged 5 to 15 years who had survived a TBI (mild, moderate, and severe) relative to an orthopaedic injury comparison group. Younger age at injury has also been associated with increased risk of social problems later in childhood (e.g. Karver *et al.*, 2012). It is therefore possible that any influence of age at injury, executive and self-regulatory skills on social outcomes varies according to (i.e. is moderated by) age or developmental stage.

The finding that executive functioning plays a role in social processing is consistent with the developmental social cognition theoretical literature. For example, based on the Social Information Processing (SIP) framework, Crick and Dodge (1994) proposed that SIP involves: the interpretation of social cues, clarification of goals, generating alternative responses, selecting and implementing a specific response, and evaluating the outcome. The role of executive processes such as inhibitory control, working memory, cognitive flexibility, planning and problem-solving, and self-monitoring are hypothesised to underpin these stages in SIP. This is elaborated upon in the integrated models proposed by Yeates *et al.* (2012) to understand social behaviour (including peer relationships), and Beauchamp and Anderson (2010) to understand social skills (i.e. social competence, social interaction, and social adjustment) following paediatric ABI. Both models suggest that development of social skills following ABI is mediated by social/affective and cognitive/executive processes. They also highlight the important influence of the external environment, including relationships with others, and brain development (Beer and Ochsner, 2006).

Therefore problems with mental health, social functioning, and peer relationships have been identified for children with clinically significant ABI. In addition' specific executive and self-regulatory skills have emerged as being associated with poor peer relationship outcomes. However, there are inconsistencies in study findings regarding the presence of social problems in younger children with ABI, and whether cognitive executive functioning, emotion regulation or behavioural inhibition predict poor social outcomes. The present study aims to replicate and extend the study of Tonks *et al.* (2010) by including an age and sex matched mental health comparison group. We also aim to identify which of the factors identified in prior research into social outcomes has predictive value in a heterogeneous clinical sample of children identified by the referrer as having neuropsychological needs. Given the relatively narrow age and age at injury ranges of samples in some prior research, we made use of a clinical sample with a wide range of ages at injury and time since injury. Based on prior studies and the SIP model, we predict that children who sustained their injury when younger, who are presenting to services later in childhood or adolescence, with poorer executive function, behaviour regulation and emotional problems, are those who will have more peer relationship problems.

The specific research questions are as follows:

1. How do emotional and peer related problems for children with clinically significant (Turner-Stokes, 2003) ABI compare with age and sex matched, non-brain injured children referred to mental health services?
2. Do age at injury, age, emotional and behavioural problems, and executive functioning predict extent of problems in peer relationships in a heterogeneous clinical sample of children with ABI?

Method

Ethical approval

Approval was provided by the local Research and Development office for the analysis of anonymised routine clinical data collected within the organisation.

Participants

1. ABI groups. Data were collected through consecutive accepted referrals to a regional specialised childhood ABI neurorehabilitation service providing a sample of 51 children who had experienced ABI after a period of typical development and a further two who had suffered birth trauma. Of the 51 participants with ABI, 29 (56.9 per cent) were male and the mean chronological age was 13 years three months (SD three years four months; range five years four months-17 years nine months). The mean chronological age at time of injury was ten years six months (SD four years seven months; range three months-17 years three months). The time between injury and assessment ranged from 26 days to nearly 13 years (SD three years two months). In all, 33 (63.5 per cent) sustained a TBI (road traffic accidents, 21; falls, eight; sporting injuries, three; assaults, one). In total, 18 experienced other types of ABI: illness/infection (encephalitis, meningitis, NMDA encephalopathy (nine); tumour (six), vascular condition/event (two), and hypoxic-ischaemic episode (one).

Severity of injury data was available for 26/33 (79 per cent) of the children who sustained a TBI. In all cases severity of injury was defined by the lowest recorded Glasgow Coma Scale (mild 13-15, moderate 9-12, and severe injury 3-8; Teasdale and Jennett, 1974) score. In total, 4/33 (12 per cent) of the TBI children had sustained a mild brain injury, 4/33 (12 per cent) had sustained a moderate brain injury and the remaining 18/33 (54.55 per cent) had sustained a severe brain injury. Information on duration of post-traumatic amnesia was not available from records. Medical records indicated that no children included in the study had evidence of pre-morbid history of learning disability or developmental disorder or behavioural problems, including substance misuse.

Data from consecutive referrals to the service comprising a subgroup of 25 from this larger sample and two children with neuropsychological problems resulting from brain injury at birth, were used for comparison with a matched mental health control group to address the first research question. In total, 19 (67 per cent) of these ABI participants were male, the mean age of the ABI group was 13 years and four months (SD three years and seven months), and 15/27 sustained a TBI, 5/27 had illnesses (meningitis, encephalitis), other causes being tumour (three), stroke (one), anoxia (one) and birth trauma (two).

2. *Mental health comparison group*. Routine data were collected from consecutive accepted and assessed referrals to a range of core and specialist services forming the Child and Adolescent Mental Health Service (CAMHS) serving a largely rural UK county with a population of approximately 600,000. An age- (within six months) and sex-matched group was identified on a case-by-case basis for comparison with the group of 27 children with ABI. These data were anonymised and provided for analysis. In total, 19 participants (67 per cent) in this group were male, and mean chronological age of the comparison group was 13 years six months (SD three years six months).

Measures

1. *Mental health and peer relationship problems*. The SDQ (Goodman, 2001) is a 25 item measure of childhood psychiatric symptoms, comprising subscales addressing emotional (depression, anxiety), conduct, attention/hyperactivity, peer relationship problems and pro-social behaviours. The "Impact" supplement provides ratings of the impact of the child's problems on domains of participation. The scale is used as a standard screening measure across CAMHS. It can be rated by parents or teachers of three- to 16-year olds, or self-rated by 11-17 year olds. Parent rated data was used for 47 (92.1 per cent) of the children. Self-rated data were used for the remaining four children, two of whom were 17 and two were 16 years, as parent rated data were not available. Goodman *et al*. (1998) has shown the self-rated SDQ to have good reliability and validity for measuring the same constructs as measured by the parent-rated version. However, Goodman notes a tendency for 11-17 year olds' self-ratings of problems to be lower than those of parents. The mean test-retest reliability after four to six months for parent rating is reported as 0.62 whereas the self-rating is less stable with a mean correlation of 0.51(Goodman, 2001).

2. *Executive function*. The Behaviour Rating Inventory of Executive Function (BRIEF; (Gioia *et al.*, 2000) is an 86 item measure of executive functioning of children in home and school environments. BRIEF subscales relate to specific domains of executive functioning: inhibition, shifting and emotional control (making up the Behaviour Regulation Index, BRI), initiation, planning/organising, working memory, organisation of materials and monitoring (making up the Metacognitive Index, MI). A total score can also be derived, the General Executive Composite (GEC). The measure is rated by parents or teachers of 5- to 18-year olds and can be self-rated by 11-18 year olds. The mean test-retest correlation for the parent form with a clinical sample has been reported as 0.79 (range 0.72-0.84) over an average of three weeks. Test-retest correlation for the BRI was 0.83, and for MI and GEC 0.81 (Gioia *et al.*, 2000). In the current study, parent rated data was used for 50 (98 per cent) of the children. Self-rated

data were used for one child aged 17 years and seven months at time of assessment as parent rated data was not available.

Analyses

1. Comparison of ABI and age, sex matched non-ABI accepted CAMHS/mental health referrals on SDQ subscale scores. A power calculation was conducted to detect a medium effect size (based on Tonks *et al.*'s, 2010 identification of differences between ABI and a healthy control group) using a dependent measures *t*-test in a between-groups analysis with probability 0.05. This indicated that a sample size of 23 in each group ($n = 46$) was required. Between-groups analysis was carried out to test for differences in SDQ scores between accepted referrals to a childhood ABI service and age and sex matched non-brain-injured children accepted for assessment in mental health services.

2. Associations between age at injury, executive, social and emotional functioning and peer relationship problems. Power calculation indicated that, to detect a large effect size using linear multiple regression (five predictor variables correlating 0.3 with the outcome variable) with probability 0.05, a total sample size of 63 is required. Correlations between variables of interest were used to identify significant associations between age at injury, cognitive, social and emotional variables and peer relationships for entry into the multiple regression. A linear multiple regression analysis was used to test variables associated with peer relationship problems, and mediation analysis using bootstrapping conducted to test indirect effects of variables following the recommendations of Baron and Kenny (1986), using appropriate methods for investigating mediation in small samples (Hayes, 2009). Hayes (2009) argued that bootstrapping procedures are more appropriate than parametric statistics for investigating the indirect effect within mediation models because assumptions regarding normality are not necessary and these methods are more powerful. We, therefore, made use of the methods and macros described by Preacher and Hayes (2008) for investigating mediation models using bootstrapping. We generated 5,000 bootstrap samples for the mediation model. Bias Corrected confidence intervals (BCa CI) were calculated to examine the significance of the indirect effect (the extent of mediation) within the model.

Results

How do emotional and peer related problems for children with ABI compare with age-and sex-matched, non-brain injured children referred to mental health services?

Between-groups comparisons (dependent measures *t*-tests) were conducted to test for differences between ABI and age and sex matched non-ABI (mental health) referrals on SDQ subscales and overall distress. There were no statistically significant differences between the two groups on SDQ total difficulties ($t = -0.221$, $df = 51$, $p > 0.05$), nor on any subscale measures (see Table I).

Frequencies of ABI and non-ABI children falling into "normal", "slight problem" and "significant problem" clinical categories on each subscale are shown in Figure 1. Proportions of children in both groups falling into the "clinically elevated" category on the SDQ were high. In the ABI group, 59 per cent were rated with high levels of distress, 76 per cent experiencing significant impact of their difficulties on domains of participation such as home life, school, leisure, and friendships. In all, 48 per cent were rated as having peer relationship problems. There appeared to be a tendency for children referred to mental health services to have more emotional problems than behavioural, this being reversed in children with ABI, but this was not statistically significant ($\chi^2 = 5.94$; $df = 4$; $p > 0.05$).

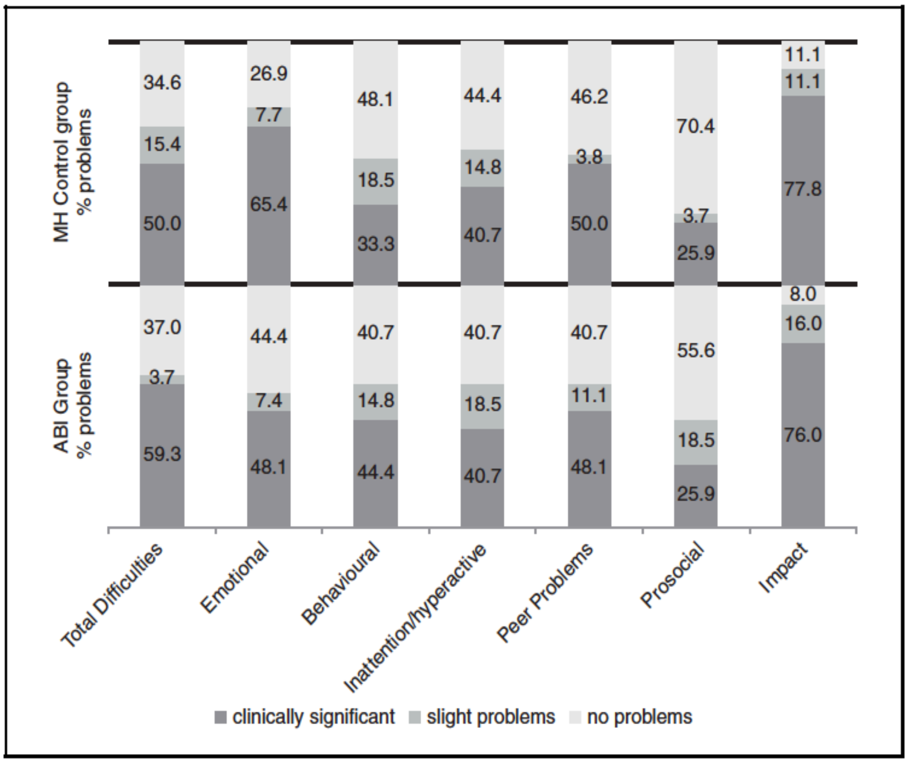
Do age at injury, age, social and behavioural problems, emotional, and executive factors predict extent of problems in peer relationships?

To test hypotheses regarding prediction of peer relationship problems, data from 51 children with ABI were analysed using linear multiple regression. First, significant correlations (at $p < 0.01$) between variables of interest were sought for inclusion in later analysis, with SDQ peer relationships subscale as the outcome variable. Age at injury was significantly associated

Table I Comparison of ABI group with mental health control group across SDQ total and subscale scores							
SDQ subscale	*Group*	*n*	*Mean*	*SD*	*T*	*df*	*Sig. (two-tailed)*
SDQ total difficulties	ABI	27	17.44	8.07	−0.221	51	0.826
	Mental health control	26	17.92	7.66			
SDQ emotional	ABI	27	4.37	2.78	−1.167	51	0.248
	Mental health control	26	5.23	2.58			
SDQ behavioural	ABI	27	3.56	2.81	0.522	52	0.604
	Mental health control	27	3.15	2.93			
SDQ attention hyperactivity	ABI	27	5.85	2.81	−0.096	52	0.924
	Mental health control	27	5.93	2.84			
SDQ peer problems	ABI	27	3.67	2.56	−0.036	51	0.971
	Mental health control	26	3.69	2.59			
SDQ prosocial	ABI	27	5.96	2.86	−0.595	52	0.554
	Mental health control	27	6.44	3.08			
SDQ impact	ABI	25	4.16	2.90	−0.481	50	0.632
	Mental health control	27	4.56	3.02			

Note: SDQ, Strengths and Difficulties Questionnaire

Figure 1 Proportions of children falling into no, slight or clinically significant categories on parent-rated SDQ total and subscale scores (per cent, $n=54$) for the mental health control (MH; $n=27$) and acquired brain injury (ABI; $n=27$) groups



with time since injury, SDQ behavioural problems and BRIEF BRI and MI subscales ($p<0.01$). Associations between peer relationship problems and the following variables were all significant ($p<0.01$): age at injury, time since injury, SDQ emotional problems, SDQ behavioural problems and BRIEF BRI and MI subscales. Age at assessment did not emerge as significant. Therefore the following variables were included in the multiple regression analysis: age at injury, time since injury, SDQ behavioural problems and BRIEF BRI and MI subscales.

The backward method was used in which all variables were entered together, the variable with the weakest correlation with the outcome variable removed, and each model thus derived tested until the most parsimonious model was achieved. As the correlation matrix contained a number of significant results, co-linearity diagnostics were included in the analysis. The results yielded satisfactory tolerance levels ranging from 0.38 to 0.84, and acceptably low variance inflation factors, highest value 2.9. As shown in Tables II and III, a model with only one predictor variable, BRIEF MI, emerged as significantly accounting for peer relationship problems ($\beta = 0.585$; $p < 0.001$). This model accounted for 32.8 per cent of the variance (adjusted $R^2 = 0.328$).

To further investigate possible mediation following the guidelines of Baron and Kenny (1986) and Hayes (2009), correlations (two-tailed) among age at injury, metacognition (BRIEF MI), and peer relationships (SDQ) were examined. There were significant negative relationships between age at injury and peer relationships, $r(51) = -0.35$, $p = 0.02$), and between age at injury and metacognition, $r(48) = -0.40$, $p = 0.005$. There was a significant positive relationship between metacognition and peer relationships, $r(48) = 0.59$, $p < 0.001$. When investigating the relationships among age at injury, metacognition, and peer relationships, using the mediation analysis described above, we found that age at injury significantly predicted both metacognition ($p = 0.005$) and peer relationships ($p = 0.013$). Metacognition also significantly predicted peer relationships, controlling for age at injury ($p < 0.001$). The indirect effect was significant ($z = -2.41$, $p = 0.02$), and this was confirmed by the results yielded using the bootstrapping method (95 per cent BCa CI = −0.21 to −0.04) and is illustrated in Figure 2.

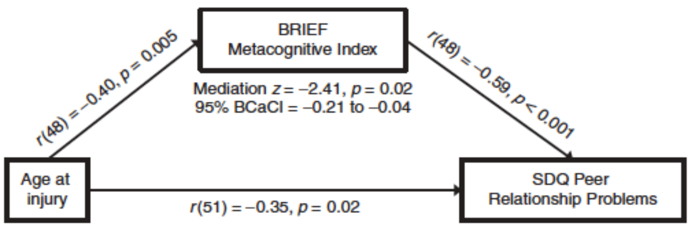
Table II Summary of multiple regression analysis for the model including BRIEF metacognitive index and constant only

Model	*Sum of squares*	*df*	*Mean square*	*F*	*p*
Regression	100.04	1	100.04	23.97	0.000
Residual	191.96	46	4.17		
Total	292	47			

Table III Summary of multiple regression model for the BRIEF MI and peer relationships

Predictor	*Adjusted R^2*	*β*	*T*	*p*
BRIEF MI	0.328	0.585	0.49	0.000

Figure 2 Diagram showing the significant correlations between predictor (age at injury), mediating factor (BRIEF MI), and peer relationship problems



Notes: BCa CI, Bias corrected confidence intervals. Mediation statistics are also provided, indicating the indirect effect of BRIEF MI to be significant

To further check possible contributions of BRIEF BRI and time since injury, we ran the above analysis to test for mediation of these variables between age at injury and peer relationship problems. No significant results were yielded. This indicates that within our data BRIEF MI significantly and fully mediated the relationship between age at injury and peer relationship problems. Our hypothesis that children who sustained their injury when younger, who are now older, with executive, emotional and behaviour regulation problems, are those who will have more peer relationship problems, is therefore only partially supported.

Discussion

This study sought to extend previous research concerned with understanding poor social outcomes that are common following childhood ABI. First, consistent with the finding of Tonks *et al.* (2010), the level and nature of social and mental health problems amongst children with ABI were comparable with those of a matched group of children with mental health problems across a range of ages. Although children with ABI appeared to have relatively higher levels of behavioural problems compared to their non-brain injured counterparts who tended to have higher levels of emotional problems, these differences were not significant. Second, correlational analyses suggested that younger age at injury was significantly associated with more peer relationship problems. Age at injury and peer relationship problems were both associated with time since injury, behavioural problems, metacognitive abilities and behavioural regulation. However, when entered together into a multiple regression the best fit for the data was achieved with a model including BRIEF MI scores only. We further explored the nature of the relationships between these variables using mediation analysis. This indicated that the effects of age at injury on peer relationship problems could be fully attributed to parent-rated metacognitive ability.

Our findings are broadly consistent with the SIP framework (Crick and Dodge, 1994) wherein social processes are considered to be underpinned by executive processes such as flexibility, working memory and self-monitoring. The theoretical models proposed by Yeates *et al.* (2012) and Beauchamp and Anderson (2010) suggest an integration of social/affective and cognitive/executive processes in social skills. However, the failure to identify emotional or behavioural regulation skills or prosocial behaviours as significant within the model means the current study's findings are only partly consistent with previous research (Dennis *et al.*, 2001; Yeates *et al.*, 2004; Ganesalingam *et al.*, 2007). In contrast to the current study, Ganesalingam *et al.* (2007) found emotion self-regulation to be a significant mediator between the presence of ABI and negative social and behavioural outcomes, but not behavioural self-regulation or cognitive (executive) functioning, although this was in a younger age group.

The results presented here should be interpreted with caution. The sample size for the multiple regression analysis falls short of that required (63) as indicated by the power calculation. Whilst this indicates that the significant association between metacognitive functioning and peer relationship problems identified here can be considered robust, it also means that the study is under powered to detect potentially significant effects of other variables. Sources of additional error variance that contribute to risk of false negative results include the wide ranges in injury severity, age, age at injury, time since injury, and use of both self and parent rated questionnaires. This may account for the difference in outcome between our study and that of Ganesalingam *et al.* (2007). The specific processes affecting development of peer relationships are likely to be different across different ages at injury and of assessment, with potential for a wider range of factors to influence outcome as time post-injury increases (Anderson *et al.*, 2009; Wade *et al.*, 2011). The current study included a wide range of age and time since injury, and results indicated age at assessment not to be significantly associated with peer relationships. However, the sample size overall was possibly too small and heterogeneous to detect age or developmental stage specific effects. Furthermore, the data consisted of parent-rated questionnaire measures, raising the risk of inflated correlations due to shared method variance. In defence of this, the analysis found acceptable levels of inter-correlations between measures. The variable included in the final model (BRIEF MI) does not appear to overlap in terms of item content with the outcome variable (i.e. metacognitive skills, such as working memory, self-monitoring, planning abilities vs relationship issues such as being bullied, having friends). Finally, the measures were taken at the same time point which means these results cannot be interpreted as indicating that problems with metacognition are a cause of peer relationship problems.

Access to rehabilitation for children in the UK is limited and children with brain injuries or their family members often have to reach crisis point before they qualify for community mental health services. Although we did not test hypotheses regarding effectiveness of interventions, our study highlights the need for an understanding of the neuropsychological deficits associated with a child's brain injury in order to deliver the most effective interventions that anticipate likely later negative outcomes. Attention to the development of executive skills and social needs of children with young age at injury may be especially important. For example, where children have executive difficulties with self-monitoring, initiating, planning and remembering about consequences of their behaviour, antecedent approaches to support self-regulation rather than the more widely used contingency based approaches are indicated (Wade *et al.*, 2006b; Ylvisaker *et al.*, 2007). Given the relationship between development of self-regulation skills and attachment (Wade *et al.*, 2011), these results are consistent with, if not supportive of, the proposal that rehabilitation of childhood brain injury should pay close attention to family functioning, parental adjustment, and attachment relationships within the family (e.g. Wade *et al.*, 2006a, 2011; Laatsch *et al.*, 2007).

In summary, and with the study limitations in mind, our results confirm the significant negative psychosocial outcomes, including poor peer relationships, associated with childhood ABI, which are not significantly different to those experienced by non-injured children presenting to community mental health services. Whilst children who acquired their brain injuries across a range of ages are at high risk of having internalising and externalising disorders, younger age at injury specifically jeopardises peer relationship problems via the mediating effect of metacognitive difficulties. Future research should include sufficiently large samples to allow for the possible moderating effects of age and developmental stage to be fully tested along with both social/affective and cognitive/executive variables.

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2.2 Summary of key findings

In the context of methodological limitations (notably selection bias, cross sectional design, small sample size, shared method variance), the study suggests that younger age at injury may be associated with poorer social relationship outcomes. The study also suggests that this relationship is mediated by metacognitive functions (as rated by parents on the BRIEF), in contrast to some previous research. The metacognitive index of the BRIEF covers everyday examples of 'cold' executive functions such as working memory, planning and problem solving. Somewhat surprisingly, 'hot' behavioural and emotional regulation scores were not associated with peer relationships in the final regression model.

CHAPTER 3

3 Do people with TBI show a 'signature' pattern of deficit on the Bangor Gambling Task assessment of emotion-based decision making compared with non-injured controls, and does this relate to cognitive and executive functions?

Adlam, A.-L. R., Adams, M., Turnbull, O., Yeates, G., & **Gracey, F.** (2017). The Bangor Gambling Task: Characterising the Performance of Survivors of Traumatic Brain Injury. *Brain Impairment*, *18*(1), 62–73. doi:10.1017/BrImp.2016.30

3.1 Introduction to the paper

There is a relative gap in formal assessment tools for 'hot' social and emotional aspects of frontal functions for people following brain injury, by comparison with assessment tools for 'cold' executive functions. One measure that is becoming well established and is published as a standardised assessment tool is the Iowa Gambling Task (IGT) (Bechara, Tranel, & Damasio, 2000). The task purports to measure ability to use an emotional biasing signal, or 'somatic marker' to guide decision making in fast moving, complex situations such as social interactions. It could therefore be considered to have potential ecological validity, insofar as scores on the measure correlate with difficulties in everyday social and emotional domains. However, there is a body of literature that is critical of the 'somatic marker hypothesis' and provides alternative hypotheses to account for findings from IGT studies. It has also been argued that in clinical practice interpretation of scores of tests in the domain of 'hot' frontal functions is complicated by much greater individual variability in the general population making development of assessments that are sensitive to detection of problems arising post-injury more difficult. To address the issue that IGT performance might be associated with working memory ability, the Bangor Gambling Task (BGT) was developed, and an initial study demonstrated similar patterns of performance on the BGT to the IGT in a group of healthy controls. However, the performance of people with TBI who are vulnerable to impairment on this measure had not previously been explored in terms of either patterns, or cold cognitive correlates, of performance.

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The Bangor Gambling Task: Characterising the Performance of Survivors of Traumatic Brain Injury

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The Bangor Gambling Task (BGT, Bowman & Turnbull, 2004) is a simple test of emotion-based decision making, with contingencies varying across five blocks of 20 trials. This is the first study to characterise BGT performance in survivors of traumatic brain injury (TBI) relative to healthy controls. The study also aimed to explore sub-groups (cluster analysis), and identify predictors of task performance (multiple regression). Thirty survivors of TBI and thirty-nine controls completed the BGT and measures of processing speed, pre-morbid IQ, working memory, and executive function. Results showed that survivors of TBI made more gamble choices than controls (total BGT score), although the groups did not significantly differ when using a cut-off score for 'impaired' performance. Unexpectedly, the groups did not significantly differ in their performance across the blocks; however, the cluster analysis revealed three subgroups (with survivors of TBI and controls represented in each cluster). Findings also indicated that only age and group were significant predictors of overall BGT performance. In conclusion, the study findings are consistent with an individual difference account of emotion-based decision making, and a number of issues need to be addressed prior to recommending the clinical use of the BGT.

Keywords: Executive, social cognition, emotion processing, measurement, psychometric, traumatic brain injury

Introduction

Traumatic brain injury (TBI) particularly affects the frontal lobes with many survivors suffering cognitive, social, and emotional difficulties, including poor or risky decision making (Hellawell, Taylor, & Pentland, 1999; Salmond, Menon, Chatfield, Pickard, & Sahakian, 2005). These difficulties can lead to a failure to return to employment and the breakdown of interpersonal relationships (Ownsworth & McKenna, 2004).

Damasio and colleagues (Bechara, Damasio, Damasio, & Anderson, 1994; 1996; Bechara, Tranel, Damasio, & Damasio, 1996; Damasio, 1994; Eslinger & Damasio, 1985) have long argued that emotion-based decision-making deficits experienced by individuals following frontal lobe lesions, particularly the ventromedial prefrontal cortex (VMPfC), are due to an inability to use emotion-based biasing signals generated from the body (somatic markers), when appraising

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different response options. It is hypothesised that decision making in complex and uncertain situations, such as social situations, involves a combination of carrying out a logical cost-benefit analysis of a given action and responding to somatic markers indicating how rewarding or punishing an action is likely to be (somatic marker hypothesis, Damasio, 1994; 1996). It is also suggested that the somatic markers lead to an 'emotional hunch' or 'gut feeling', which can guide cognitive decision making (Damasio, Adolphs, & Damasio, 2003).

Empirical support for the somatic marker hypothesis has largely stemmed from the Iowa Gambling Task (IGT), an experimental paradigm designed to mimic real-life decision making by factoring in complexity, uncertainty, reward, and punishment (see Bechara, Damasio, & Damasio, 2000). In the IGT, participants either win or lose money (real money or facsimile) by selecting cards from four separate decks. Two of the card decks have high wins and high losses ('risky' or 'bad' decks), and two of the decks have small wins and small losses ('safe' or 'good' decks). Participants are not told which decks are risky/bad or safe/good, and their choice of deck selection is recorded across 100 trials. A key feature of this task is that participants learn to forego short-term benefit for long-term profit over a long and complex (multiple decks with varying contingencies) reinforcement history. Changes in anticipatory skin conductance response (SCR) have been associated with successful learning, that is, making fewer 'bad' choices, and instead selecting from the safe decks, supporting the role of somatic markers in task performance (Bechara et al., 2000). It is argued, therefore, that to do well on this task participants must rely on 'intuitive' decision making processes, in particular, the activation of somatic marker biasing signals.

Consistent with the notion that the IGT mimics real-life complex decision making, individuals with frontal lobe damage failed to adopt optimal strategies when performing the IGT leading to a continued preference for the 'risky' decks (Bechara et al., 1994). More importantly, these same individuals failed to show the anticipatory physiological changes in SCRs found in healthy controls (Bechara et al., 1996). Similar findings have been shown in individuals following TBI (Cotrena et al., 2014; Fujiwara, Schwartz, Gao, Black, & Levine, 2008; Levine et al., 2005). Despite the wealth of evidence supporting the IGT as a test of emotion-based decision making (for a review, see Bechara, 2004), studies have questioned the role of somatic markers and whether alternative mechanisms can account for performance on the IGT (for review, see Dunn, Dalgleish, & Lawrence, 2006). For example, some studies using dual-task methods (Hinson et al., 2002; Jameson et al., 2004; but see Turnbull et al., 2005) and neurological studies (Bechara, Damasio, Tranel, & Anderson, 1998) have found that performance on the IGT is influenced by working memory capacity. Other studies have questioned the assumption that tasks need to be as complex as the IGT to capture emotion-based decision making. For example, Bowman and Turnbull (2004) designed the Bangor Gambling Task (BGT) to have a similar structure to the IGT, that is, a gambling task with explicit financial reward/punishment and with varying contingencies, but to be a simpler version of the task. In contrast to the four-deck IGT, the BGT involves using a single deck of cards; therefore, varying contingencies across time rather than time and space as in the IGT. It also has a simple 'gamble/no gamble' response and either a win or loss on each trial. In the original IGT, participants receive both win and loss feedback on each trial, which potentially increases the working memory demands of the task. In their study, comparing performance on the BGT directly with performance on the IGT, Bowman and Turnbull (2004) found that undergraduate participants showed the same incremental learning across both tasks, with a significant correlation in overall performance. Furthermore, the same individuals were impaired using the IGT cut-off of +9 or below (Bechara et al., 2001) on both tasks. Based on these findings, Bowman and Turnbull (2004) concluded that not only does the BGT have similar structural properties to the IGT, but participants also perform similarly on both tasks, thus tasks do not need to be complex to measure emotion-based decision making. The authors suggested that the BGT potentially offers a simple measure of emotion-based decision making, removing the complex instructions and complex feedback of the IGT, which can be used with neurological patients.

Following this suggestion, we report the first study to examine the performance of individuals with TBI relative to healthy controls on the BGT. This study aimed to characterise BGT performance in survivors of TBI with the predictions that (i) survivors of TBI will make more 'gamble' choices than controls (total BGT score); (ii) more survivors of TBI than controls will fall below the Bowman and Turnbull (2004) suggested cut-off of +9 or below; and (iii) survivors of TBI will have difficulty learning to change their 'gamble' response in the face of increasing losses. Specifically, there will be a significant Group by Block interaction, with no group difference on Block 1 (i.e., the first 20 trials, consistent with previous IGT studies see, for example, Tranel, Bechara, & Denburg, 2002).

Given the novelty of this study, we also set out to explore patterns of performance across blocks using cluster analysis to identify sub-groups. This is of interest given the individual differences shown in control performance on emotion-based decision tasks (e.g., Dunn et al., 2006; Horstmann, Villringer, & Neumann, 2012; Steingroever, Wetzels, Horstmann, Neumann, & Wagenmakers, 2013), and the heterogeneity in TBI (e.g., pre-morbid ability, mechanism of injury, time since injury, extent brain damage, etc.).

Finally, given the literature exploring alternative mechanisms for performance on emotion-based decision making tasks (e.g., for a review, see Dunn et al., 2006), the current study aimed to explore possible predictors of performance. Whole group multiple regressions were conducted with working memory and executive function ability as predictors. The additional variables of presence of injury, speed of processing, estimated pre-morbid IQ, age, and gender were included as possible predictors.

Method

Participants

Thirty survivors of TBI (25 males, 5 females; mean age: 34 years, range: 20–52 years; mean years of education: 13 years, range 12–17 years), recruited from a regional specialist neurorehabilitation centre, participated in this study (see Table 1 for details of sample characteristics). All participants survived a closed head injury, were at least 6 months post injury (mean time since injury: 51.4 months, range: 11–192 months) and had emerged from post-traumatic amnesia. According to the Glasgow Coma Scales (GCS, Teasdale & Jennett, 1974) severity classifications, 23 of the survivors suffered severe TBI (GCS 3–8), 2 suffered moderate TBI (GCS 9–12), and 2 suffered mild TBI (GCS 13+), with missing data for 3 participants. Despite the missing data and the variability in injury severity, all participants showed clinically significant impairments in everyday functioning consistent with referral to the specialist neurorehabilitation service.

Thirty-nine healthy control participants (17 males, 22 females; mean age: 38 years, range: 18–65 years; mean years of education: 14 years, range 12–18 years) were recruited from the MRC Cognition and Brain Sciences Unit's volunteer panel.

The groups did not significantly differ on age ($p = .27$) or years of education ($p = .13$), but there were significantly more females in the healthy control sample than the TBI sample ($\chi^2(1) = 11.25$; $p = .001$).

This study had ethical approval from the University of Cambridge local research ethics committee and written informed consent was obtained.

Measures

Bangor Gambling Task (BGT). The BGT was administered in accordance with the procedures outlined by Bowman and Turnbull (2004). Using a deck of 100 playing cards, 9 were labelled as 'win 20p', 29 'win 10p', 35 'lose 20p', and 27 'lose 10p'. Participants were given written instructions, which were also read aloud by the examiner, informing them that the aim was to make as much money as possible. It was at the discretion of the player whether to gamble or not, but they were instructed to inform the experimenter prior to turning over the top card of the deck. At the start of the game, all participants were given £2.00 and told that they could keep any money they won. Unknown to the participants, the deck was split into five blocks of 20 card selections. If participants gambled on every card, they would win £1.00 on Block 1 (15 win, 5 lose cards), neither win nor lose on Block 2 (10 win, 10 lose), lose £1.00 on Block 3 (5 win, 15 lose), lose £2.00 on Block 4 (5 win, 15 lose at a higher value), and lose £3.00 on Block 5 (3 win, 17 lose). The cards were administered in the same order to each participant. The task took approximately 15 minutes to complete (with instructions).

Consistent with the IGT and the Bowman and Turnbull (2004) original study, performance was calculated as the number of 'no gamble' minus the number of 'gamble' decisions made per block and overall. A negative score indicates more 'gamble' responses.

It is important to note that in the BGT to gamble in the first block is advantageous; therefore, choosing to 'gamble' in Block 1 could be interpreted as exploratory behaviour rather than impaired decision making. As with other measures of emotion-based decision making (e.g., the IGT), participants would not be aware of this without first sampling the cards; therefore, Block 1 is included in the main analysis with the prediction of no group difference on Block 1.

Characterisation measures and potential predictors of BGT performance. Survivors of TBI and controls completed the following measures: The Speed and Capacity of Language Processing test (SCOLP, Baddeley, Emslie, & Nimmo-Smith, 1992) measured speed of processing (Speed of Comprehension) and estimated pre-morbid IQ (Spot the Word). Working memory was measured using two subtests of the Wechsler Adult Intelligence Scales 3rd edition (WAIS-III, Wechsler,

TABLE 1
Demographics, Cognitive Functioning, and Injury Characteristics for Control and TBI Groups

	Control			TBI		
	N	Mean (range)	*SD*	*N*	Mean (range)	*SD*
Male**	39	17	–	30	25	–
Age (years)	39	38.18 (18–65)	–	30	34.73 (20–52)	–
Years of education	39	14.20 (12–18)	–	19	13.21 (12–17)	–
BGT Total score*	39	2.41 (–56–130)	28.54	30	– 14.51 (–100–44)	27.86
BADS Six Elements (ps)		3.59	0.64	26	3.23	0.86
BADS Six Elements rules broken	39	0.23	0.49	26	0.38	0.75
Digit Span Backwards (rs)	39	7.03	2.80	24	6.63	2.83
Letter Number Sequencing (rs)	39	10.79	2.86	22	9.86	3.47
SCOLP Speed of Comprehension (ss)**	39	13.51	3.22	26	6.23	2.46
SCOLP Spot the Word (ss)**	39	11.54	3.19	26	8.38	2.70
Time since injury (months)		n/a		30	51.40 (11–192)	–
GCS on admission (min. 3, max.15)		n/a		27	6.31 (3–15)	–
GCS classification (Rimel, Giordani, Barth, & Jane, 1982)		n/a		27	–	–
Severe (–8)				23	–	–
Moderate (9–12)				2	–	–
Mild (13+)				2	–	–
DEX patient total		n/a		24	36.42	19.52
DEX carer total		n/a		21	38.76	16.38

N = sample size; *SD* = standard deviation; BGT = Bangor Gambling Task; BADS = Behavioural Assessment of the Dysexecutive Syndrome; SCOLP = Speed and Capacity of Language Processing; GCS = Glasgow Coma Scale (Teasdale & Jennett, 1974); DEX = Dysexecutive Syndrome Questionnaire.
*$p < .05$; ** $p < .01$
ps = profile score as derived from manual; ss = scaled score as derived from manual; rs = raw score.

1997), digit span and letter–number sequencing, and executive function was measured using the Modified Six Elements (6 Elements) subtest of the Behavioural Assessment of the Dysexecutive Syndrome (BADS, Wilson, Alderman, Burgess, Emslie, & Evans, 1996). In addition, survivors of TBI and their relatives completed the Dysexecutive Syndrome Questionnaire (DEX, Wilson et al., 1996) to characterise behavioural symptoms of everyday executive function difficulties.

A measure of attention, the Sustained Attention to Response Task (SART, Robertson, Manly, Andrade, Baddeley, & Yiend, 1997), was originally included in the study protocol; however, only 56% ($n = 22$) of the controls and 16.6% ($n = 5$) of the TBI group completed this measure. Due to the large amount of missing data, the SART was excluded from subsequent analysis.

Plan of Analysis

A Student's T test was used to test for group differences on total BGT score, and a Chi-Squared test was used to test for differences in the proportion of survivors of TBI performing below the cut-off (+9 or below) relative to the proportion of controls.

To test for the predicted Group (TBI, Control) by Block (1, 2, 3, 4, 5) interaction, a mixed-model ANOVA with a between-subjects factor of Group and a within-subjects factor of Block was conducted. Box's test of equality of variances was not significant; however, Mauchly's test of sphericity was significant; therefore, the Greenhouse–Geisser correction was applied. G-Power indicated that to detect a significant two-way interaction (Group by Block) with a medium effect size, at alpha .05 and power .85, a total sample size of 36 would be required.

To explore individual differences in performance across Blocks, and potential sub-groups, a hierarchical cluster analysis was conducted using the squared Euclidian distance measure of difference between cases. The clustering variables were scores for performance on each of the five blocks. As these measures are on the same scale, it was not necessary to standardise the scores prior to clustering. Identified clusters were compared in terms of proportion of TBI vs. controls, demographics (age and gender) and cognitive variables (SCOLP Speed of Comprehension, SCOLP Spot the Word, BADS 6 Elements, letter-number sequencing, and backwards digit span).

Finally, to explore predictors of total BGT performance and to test contribution of specific executive and working memory variables, a stepwise hierarchical multiple regression was conducted with overall BGT score as the outcome variable, demographic variables in Model 1 (age, gender), injury group, SCOLP Speed of Comprehension scaled score (processing speed), and SCOLP Spot the Word scaled score (estimated pre-morbid IQ) in Model 2, and letter–number sequencing and backwards digit span (working memory), and BADS 6 Elements profile score (executive function) in Model 3.

Results

Table 1 shows the participant demographics, characteristics, and performance on the measures included in the hierarchical multiple regressions.

Hypotheses 1 to 3: Survivors of TBI Will be Impaired Relative to Controls on the BGT

As predicted (Hypothesis 1), overall, survivors of TBI made more gamble choices than controls (total BGT score: $t(67) = 2.47$, $p = .02$; see Table 1). However, when the suggested cut-off of +9 or below (Bowman & Turnbull, 2004) was applied, the number of survivors of TBI (80%) classified as ‘impaired’ was not significantly greater than the number of controls (66.7%; Hypothesis 2: $\chi^2(1) = 1.51$; $p = .22$).

Unexpectedly, the prediction that survivors of TBI will have difficulty learning to change their ‘gamble’ response in the face of increasing losses (Hypothesis 3) was not supported (Group by Block interaction: $F(2.19, 67) = 1.98$, $p = .14$; $\eta_p^2 = .03$, small effect). The mixed-model ANOVA revealed, however, a significant main effect of Group ($F(1, 67) = 6.41$; $p = .01$; $\eta_p^2 = .09$, medium effect) and a significant main effect of Block ($F(2.19, 67) = 23.74$, $p = .0001$; $\eta_p^2 = .26$, large effect). Paired t-tests (Bonferroni correction, $p \leq .005$) revealed the following pattern of performance across the Blocks: $1 = 2 = 3 < 4 < 5$, indicating that in Blocks 4 and 5 participants were selecting more advantageously than in Blocks 1–3; therefore, participants showed evidence of learning across blocks (see Figure 1).

Exploring Individual Differences and Sub-Groups on BGT Performance

Inspection of the dendrogram derived from the hierarchical cluster analysis identified a parsimonious cut-off of +10, which yielded three clusters. These clusters were very clearly distinguished by pattern of performance, as shown in Figure 2. Cluster 1 ($n = 50$) showed a tendency to gamble more initially over Blocks 1–3 and reversal of this from Block 4 to 5. Cluster 2 ($n = 9$) largely refrained from gambling throughout, showing a tendency towards gambling on Block 3, and then reversal of this pattern. Cluster 3 ($n = 7$) was similar to Cluster 2 at Block 1 but showed a contrasting pattern of increased gambling over time, with little or no reversal over the later blocks. The performance of three participants was not classified by the analysis.

Post-hoc analyses were not performed due to the small sample sizes in the clusters (particularly Clusters 2 and 3); however, as shown in Table 2, descriptively, Cluster 3 appeared to be characterised by more males and more survivors of TBI.

Exploring Predictors of BGT Performance

Model 1 (age and gender) was significant ($F(2, 55) = 4.31$; $p = .02$), with addition of group, processing speed, and estimated pre-morbid IQ significantly improving the model (Model 2, Adjusted $R^2 = .20$; R^2 change = .13, $F(3, 52) = 3.14$; $p = .03$). Addition of working memory and executive variables did not significantly improve variance accounted for in Model 3 (R^2 change = .01, $F(3, 49) = .31$; $p = .82$). Model 2 accounted for the greatest amount of variance in BGT performance (20%), with only age ($\beta = -.28$; $t = -2.28$; $p = .03$) and group ($\beta = -.54$; $t = -2.67$; $p = .01$) remaining significant.

Discussion

This study aimed to characterise BGT performance in survivors of TBI. Findings suggest that consistent with previous studies examining emotion-based decision making (e.g., IGT; Cotrena et al., 2014; Fujiwara et al., 2008; Levine et al., 2005), overall survivors of TBI made more gamble choices than controls.

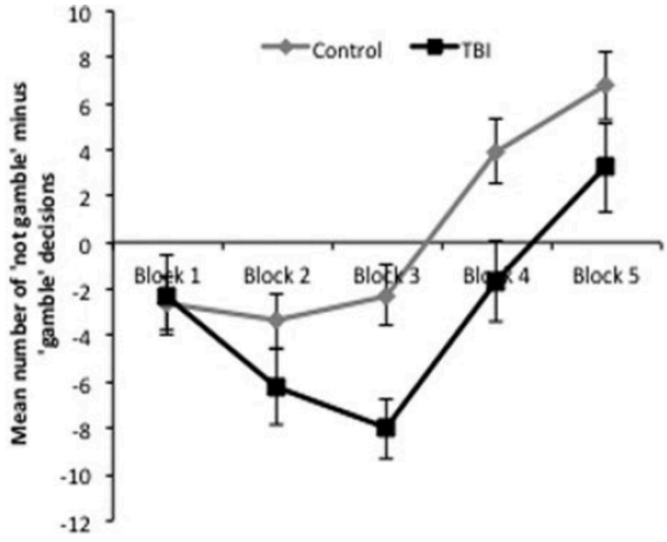

FIGURE 1

Performance (mean number of 'not gamble' minus 'gamble' decisions per block, with standard error bars shown) of each group (Control, TBI) on the BGT across the five blocks.

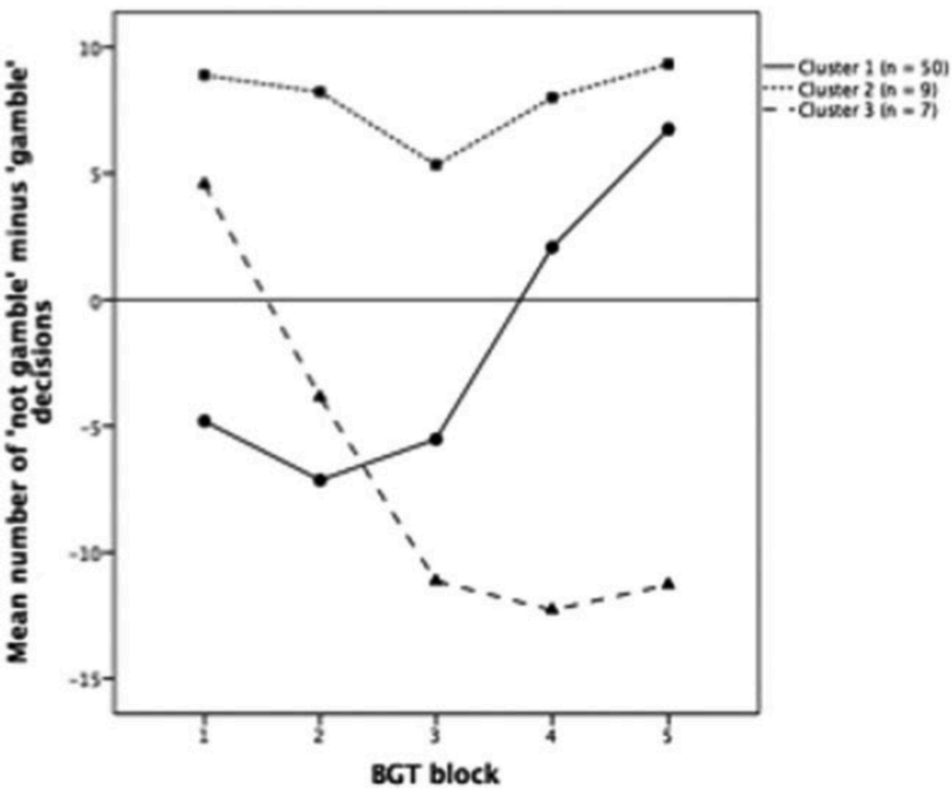

FIGURE 2

Chart showing mean performance for each of the three clusters on the BGT across the five blocks.

TABLE 2
Characteristics of the Three Clusters Differentiated by Block-by-Block Pattern of BGT Performance

Variable	Cluster	*N* (%)	Mean	*SD*
Gender (male)	1	28 (56)	–	–
	2	6 (66.70)	–	–
	3	6 (85.70)	–	–
Group (TBI)	1	22 (44)	–	–
	2	3 (33.30)	–	–
	3	4 (57.10)	–	–
Age (years)	1	50	36.10	13.23
	2	9	36.22	11.09
	3	7	39.86	13.08
SCOLP Speed of Comprehension (ss)	1	47	10.55	4.93
	2	8	11.25	4.59
	3	7	10.43	3.95
SCOLP Spot the Word (ss)	1	47	9.87	3.56
	2	8	12.25	2.12
	3	7	10.29	2.98
BADS Six Elements (ps)	1	48	3.40	0.79
	2	8	3.50	0.76
	3	6	3.67	0.52
Letter-Number Sequencing (rs)	1	46	10.20	3.19
	2	6	12.17	3.06
	3	6	11.33	1.63
Digit Span Backwards (rs)				
	1	47	6.53	2.54
	2	7	8.71	4.31
	3	6	7.67	1.97

N = sample size; *SD* = standard deviation; BGT = Bangor Gambling Task; SCOLP = Speed and Capacity of Language Processing; BADS = Behavioural Assessment of the Dysexecutive Syndrome. ps = profile score as derived from manual; ss = scaled score as derived from manual; rs = raw score.

The groups did not significantly differ, however, when the Bowman and Turnbull (2004) suggested cut-off of +9 or below to indicate 'impaired' performance was applied, due to a high proportion of controls (66.7%) being classified as 'impaired'. It is not uncommon to find emotion-based decision making 'impairments' in controls. For example, Steingroever et al. (2013) found that up to a third of controls failed the IGT, with some authors suggesting that individual differences in task approach and strategy use can account for this (e.g., Horstmann, Villringer, & Neumann, 2012). Interestingly, more controls were classified as impaired in the current study (66.7%) compared to the undergraduate sample reported in the original BGT study (35%; Bowman & Turnbull, 2004). There are many possible reasons for the difference in performance between the two control groups and a direct comparison is not possible. However, from examining the reported demographic data, the controls in the original study were younger than those in the current study and age has been shown to be associated with both performance (Cauffman et al., 2010) and strategy use (Wood, Busemeyer, Koling, Cox,

& Davis, 2005) on emotion-based decision making tasks. Also, the control group in the current study had a higher proportion of male participants (44%) compared to the original study (30%), and gender has also been associated with performance on emotion-based decision making tasks (van den Bos, Homberg, & de Visser, 2013). It is also worth noting that the BGT cut-off suggested in the original study was taken from Bechara et al. (2001), who used the performance of individuals with VMPfC damage on the IGT to guide the criteria for impairment in other patient groups (e.g., substance use). It is possible, therefore, that the suggested cut-off of +9 or below is not the most sensitive measure of impairment on the BGT.

Based on previous studies (e.g., Bechara et al., 1994), it was expected that survivors of TBI would show difficulty learning to reduce their 'gamble' responses in the face of increasing losses and thus show a different pattern of performance compared to controls across the five blocks. This prediction was not supported, and despite an a priori power calculation indicating that the current study had a large enough sample size to detect a medium effect size (Group by Block interaction), the actual effect obtained was small, suggesting a larger sample size might have been needed. The results were consistent with the findings on overall BGT score, in that the TBI group made more gamble choices than the controls (main effect of Group); however, both groups showed the general pattern of making more gamble choices on Blocks 1–3, and then reversing this on Blocks 4 and 5 (main effect of Block). Analysing performance between-groups and across blocks of 20 trials is consistent with previous studies of emotion-based decision making tasks (e.g., Bechara et al., 2001; Bowman & Turnbull, 2004; Tranel et al., 2002); however, some authors have questioned the sensitivity of this approach, particularly for patient populations (e.g., Dunn et al., 2006; Ryterska, Jahanshahi, & Osman, 2013).

We explored the data further to identify potential sub-groups with broadly similar patterns of performance. Three clusters were identified, with both survivors of TBI and controls being represented in each cluster. The largest group (Cluster 1) tended to gamble initially (which is favourable in Block 1), increase tendency to gamble slightly in Block 2 (which would lead to a neutral outcome), but reduce gambling over Blocks 3–5. This pattern of performance suggests evidence of learning in response to the change in contingencies. Taking this a step further, given that 22 of the 30 survivors of TBI were represented in this cluster, this suggests that survivors of TBI can show learning on an emotion-based decision making task.

Participants in Cluster 2 appeared to avoid gambling throughout the task, even in Block 1 where gambling is advantageous, suggesting a risk-avoidant strategy, whereas Cluster 3 increased gambling throughout the task and did not change their behaviour in response to the change in contingency. The pattern of performance in Cluster 3 suggests an inability to forgo short-term gain for long-term profit, although the profile is even more extreme than that typically shown by individuals with bilateral damage to the VMPfC (e.g., Bechara et al., 1994; Tranel et al., 2002). Although not tested here, alternative mechanisms might also account for this finding including adopting a risk-taking strategy or being sensation seeking, apathy or insensitivity to negative outcomes, impaired reversal learning, or an inability to inhibit a response (see Dunn et al., 2006 for detailed discussion).

Differences between clusters on performance during Block 1, and prior to any manipulation of contingencies, indicate potential for large individual differences in approach to the task, which may make identification and interpretation of performance differences following contingency changes more problematic. Furthermore, although overall differences in the BGT between controls and survivors of TBI could be accounted for by the TBI group having an acquired impairment in emotion-based decision making, an alternative account is that individual differences in risk-taking behaviour might underlie the likelihood of sustaining a TBI, further confounding interpretation of performance on gambling tasks such as the BGT.

Given the small group sizes it was not possible to analyse differences on demographic or cognitive variables; however, descriptively, Cluster 3 appeared to have more survivors of TBI than controls and more males than females. Despite the caveats of this being an exploratory analysis with the cluster yielding a small group size, the higher proportion of survivors with TBI than controls in Cluster 3 is consistent with the main prediction that survivors of TBI will fail to learn from increasing losses, although an individual differences account cannot be ruled out.

In relation to the sex-difference found in Cluster 3, there is a growing body of literature suggesting that men and women perform emotion-based decision making tasks differently. These studies generally show that men focus on the long-term gain, whereas women focus on both the long-term gain and the win/loss frequency when performing tasks such as the IGT (for review, see van den Bos, Homberg, & de Visser, 2013). Sex differences in performance (or task strategy) on the BGT have not been examined; therefore, it is not possible to conclude that males and females perform the task differently, and instead, it is possible that the higher number of males in Cluster 3 is accounted for by the higher number of survivors with TBI, who were predominantly male (83.3% of the sample) in the current study. Related to this possible explanation, presence of injury was significant in the multiple regression analysis when examining predictors of overall BGT performance, whereas gender was not.

The multiple regression analysis also found no significant influence of processing speed or premorbid IQ on overall BGT performance, despite group differences on these variables. The finding that processing speed did not influence overall BGT performance is consistent with the recent literature examining predictors of IGT performance (Gansler, Jerram, Vannorsdall, Schretlen, 2011). Similarly, a review of 43 studies of gambling task performance indicated that only a small number of studies found significant effects of IQ on IGT performance and, in these studies, effect sizes were small (Toplak, Sorge, Benoit, West, & Stanovich, 2010).

There was also no significant influence of working memory or executive function ability on overall BGT performance, which is in keeping with some previous studies (e.g., Bechara et al., 1998; Turnbull et al., 2005). It is worth noting that there was also no significant difference between the survivors of TBI and controls on these measures which, although inconsistent with some studies of neurocognitive performance following TBI (e.g., Dunning, Westgate, & Adlam, 2016; Zimmermann et al., 2015), is consistent with findings reported in patients with anterior VMPfC (i.e., poorer emotion-based decision making relative to controls but intact working memory, Bechara et al., 1998).

Finally, the multiple regression analysis suggested that age influenced overall BGT performance. As with gender (see above), age differences in performance on the BGT have not been directly studied; however, studies using the IGT have found age differences in performance between adolescents and adults up to the age of 30, with avoidance of the disadvantageous decks improving with age (i.e., make fewer 'bad' choices, Caufmann et al., 2010). This is inconsistent with our finding of poorer overall performance (i.e., make more 'gamble/bad' choices) with increasing age. It is possible that differences in strategy use might have influenced performance (e.g., Wood et al., 2005; although their study found no corresponding difference in task performance); however, age effects should be directly studied before firm conclusions can be drawn.

Limitations and Future Directions

As discussed above, the cut-off used to classify 'impaired' vs. 'not impaired' performance on the BGT was not directly derived from BGT data and, therefore, might not be sensitive to group differences in performance (as suggested by the finding of a significant difference in total score, but not when using the cut-off to classify total score). Future research might want to consider alternative approaches to identifying impaired performance on the BGT, and total score might in itself not be a sensitive measure (see Dunn et al., 2006 for discussion). Related to this, given the lack of a significant Group by Block finding, future research (particularly with clinical groups) might want to examine linear contrasts/trends in performance rather than performance across blocks of 20 trials (see Dunn et al., 2006). The exploratory cluster analysis raised some interesting findings; however, the small sample sizes in the clusters limited the use of post-hoc tests to further examine their characteristics. Future research might want to extend this approach to analysing emotion-based decision making performance in a larger sample of participants and identifying models that can best account for individual differences (e.g., Franken & Muris, 2005), including pre-injury factors and acquired neurocognitive changes in survivors of TBI.

A further limitation of our study is that estimated IQ, as measured using the SCOLP Spot the Word subtest, significantly differed between the TBI and control groups and is, therefore, a potential confound. Future studies might want to address this by including groups matched on IQ.

Finally, the current study, consistent with the original BGT study (Bowman & Turnbull 2004), did not test whether the BGT relies on somatic markers (emotion biasing signals) to guide decision making. Future studies might want to examine this more directly using psychophysiology methods.

Clinical Implications

Despite the attractiveness of its simplicity over other tests (e.g., the IGT), and that it is free to use (compared to IGT (prices in 2016): £472 in the UK; $574 in the USA; $1072.50 in Australia), the current study raises a number of issues that need to be addressed prior to recommending use of the BGT as a measure of emotion-based decision making for survivors of TBI. First, a meaningful, reliable, and sensitive approach to classify task performance as being impaired or not needs to be developed. This may, for example, model individual differences in patterns of performance rather than classify a total score. Second, the mechanisms underpinning task performance (e.g., somatic markers, reversal learning, or individual differences in sensation-seeking behaviours, etc.) need to be confirmed. Third, the relationship between performance on the BGT and real-life emotion-based decision making needs to be confirmed. Finally, if the BGT is to replace the IGT, then it needs to be confirmed that the BGT shares similar properties to the IGT when performed by survivors of TBI, for example, by a direct comparison as tested in the original Bowman and Turnbull (2004) study.

Conclusion

Despite survivors of TBI making more gamble decisions compared to controls on the BGT, this study suggests considerable overlap between survivors of TBI and controls in their individual patterns of performance. These findings are in keeping with an individual difference account of emotion-based decision making, and it is suggested that future research focuses on developing models to best capture performance on emotion-based decision making tasks in survivors of TBI. It is also suggested that future research examines the mechanisms underpinning performance on emotion-based decision making tasks, and the neural correlates associated with performance, in survivors of TBI. In conclusion, the current study raises a number of issues that need to be addressed prior to recommending use of the BGT as a measure of emotion-based decision making for individuals with neurological conditions.

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Conflict of Interest

None.

Ethical Standards

The authors assert that all procedures contributing to this work comply with the ethical standards of the relevant national and institutional committees on human experimentation and with the Helsinki Declaration of 1975, as revised in 2008.

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3.2 Summary of key findings

It can cautiously be concluded that the BGT performance is more impaired in those with TBI than healthy controls, and that it is not associated with cold cognitive processes but may be associated with general processing abilities as reflected in estimated IQ and speed of processing. Furthermore, the cluster analysis indicates that an apparent group difference between TBI and controls seems more likely attributable to individual differences in general strategies for the gamble task, with 2 small groups showing almost complete avoidance of gambling throughout, or a strategy of mostly choosing to gamble, respectively.

CHAPTER 4

4 Do executive functions and coping styles interact to determine different emotional outcomes amongst people with everyday EF problems following ABI?

Gracey, F., Fish, J. E., Wagner, A. P., Simblett, S. K., Bateman, A., Malley, D., Evans, J. J. & Manly, T. (submitted / under review). The influence of executive functioning on the relationship between coping style and emotional outcomes in the chronic phase following acquired brain injury. *Accepted (with revisions) to Neuropsychology, November 2018*

4.1 Introduction to the paper

Research in the field of affective neurosciences has identified shared processes related to 'hot' regulation of emotion and 'cold' cognition. For example, an influential model implicates executive attention early in the processes of triggering of a negative emotion, working memory in early coping responses such as verbal reappraisal, and involvement of the supervisory attentional system in devising a new 'behavioural schema' for responding adaptively to the emotion. This has been explored a little in ABI populations, in studies focussing on potential interactions between coping style and EFs. However, there have been methodological and conceptual limitations to study design and interpretation, resulting in variability in findings. In this study we therefore set out to identify the presence of interactions between 'cold' frontal functions (maintaining attention to a simple goal, and supervisory functions involved in developing and implementing a plan), coping styles and emotional outcomes across anxiety, depression and anger in a heterogenous sample of people with clinician-identified everyday executive difficulties.

Neuropsychology

The influence of executive functioning on the relationship between coping style and emotional outcomes in the chronic phase following acquired brain injury

--Manuscript Draft--

Manuscript Number:	
Full Title:	The influence of executive functioning on the relationship between coping style and emotional outcomes in the chronic phase following acquired brain injury
Article Type:	Data-Driven Article
Abstract:	Background Weaknesses in executive functioning (EF) have been proposed to create a general vulnerability to increased emotional distress, and to interact with coping style. However, studies of coping and EF following acquired brain injury have failed to consistently support hypothesized interactions. The current study sought to identify contributions of coping styles and metacognitive and cognitive EF to models of depression, tension-anxiety and anger-hostility in people with ABI. Method 69 people with ABI (43 Male, 34 TBI, mean age 47.8 years) were recruited and completed demographic and clinical measures, the Coping Inventory for Stressful Situations, the Profile of Mood States and The Hotel Task and Sustained Attention to Response Test (SART) at the baseline phase of a separately reported trial of an EF rehabilitation intervention. Results Multiple regression analyses employing a model fit approach (Akaike's Information Criterion, AIC) identified Hotel Task * task-oriented coping interaction terms in models of anxiety and depression, and SART * emotion focused coping in the anxiety model, consistent with poorer EF being associated with a greater impact of coping style. Fatigue and emotion-focused coping style emerged as associated with all three emotional outcomes. Conclusions The current study is the first to identify an effect of interaction between EF and coping style on emotional outcomes in ABI. It is proposed that future studies include measures of metacognitive or higher EF rather than more circumscribed cognitive EF measures, explore mechanisms by which fatigue is associated with emotional outcome, and employ longitudinal designs.
Keywords:	Brain Injuries; Executive Functions; Coping; Emotions
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The influence of executive functioning on the relationship between coping style and emotional outcomes in the chronic phase following acquired brain injury.

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The influence of executive functioning on the relationship between coping style and emotional outcomes in the chronic phase following acquired brain injury.

Abstract

Background

Weaknesses in executive functioning (EF) have been proposed to create a general vulnerability to increased emotional distress, and to interact with coping style. However, studies of coping and EF following acquired brain injury have failed to consistently support hypothesized interactions. The current study sought to identify contributions of coping styles and metacognitive and cognitive EF to models of depression, tension-anxiety and anger-hostility in people with ABI.

Method

69 people with ABI (43 Male, 34 TBI, mean age 47.8 years) were recruited and completed demographic and clinical measures, the Coping Inventory for Stressful Situations, the Profile of Mood States and The Hotel Task and Sustained Attention to Response Test (SART) at the baseline phase of a separately reported trial of an EF rehabilitation intervention.

Results

Multiple regression analyses employing a model fit approach (Akaike's Information Criterion, AIC) identified Hotel Task * task-oriented coping interaction terms in models of anxiety and depression, and SART * emotion focused coping in the anxiety model, consistent with poorer EF being associated with a greater impact of coping style. Fatigue and emotion-focused coping style emerged as associated with all three emotional outcomes.

Conclusions

The current study is the first to identify an effect of interaction between EF and coping style on emotional outcomes in ABI. It is proposed that future studies include measures of metacognitive or higher EF rather than more circumscribed cognitive EF measures, explore mechanisms by which fatigue is associated with emotional outcome, and employ longitudinal designs.

Keywords

Brain Injuries, Executive Functions, Coping, Emotions

Public significance statement

Brain injury causes a mix of problems with thinking skills and emotions that can interact to influence how people adjust in their everyday lives. This study shows problems with particular higher-level cognitive skills, executive functions, can make coping with the stress of life after brain injury much more difficult. Therefore, in clinical services for people with brain injuries, assessing and supporting executive functioning difficulties alongside emotional difficulties is important.

Introduction

The elevated incidence and prevalence of emotional disorders following ABI (Fann, Katon, Uomoto, & Esselman, 1995; Hart et al., 2016; Tsaousides, Cantor, & Gordon, 2011), improved understanding of the psychological impact of ABI, and the need for integration of psychological factors into rehabilitation is increasingly well documented (Ownsworth, Fleming, Haines, Cornwell, Kendall, Nalder & Gordon, 2011; Ownsworth & Haslam, 2014; Wilson, Gracey, Malley & Bateman, 2009). Progress is being made with the development and evaluation of psychological interventions that might both treat (Ashman, Cantor, Tsaousides, Spielman, & Gordon, 2014; Bradbury, Christensen, Lau, Ruttan, Arundine, & Green, 2008; Hsieh, Ponsford, Wong, Schoenberger, Taffe & Mckay, 2012; Waldron, Casserly, & O'Sullivan, 2012; Watkins, Auton, Deans, Dickinson, Jack, Lightbody, Sutton, van den Broek & Leathley, 2007) or prevent later development of (Backhaus, Ibarra, Klyce, Trexler, & Malec, 2010; Hackett, Anderson, House, & Halteh, 2008; Watkins, Wathon, Leathley, Auton, Deans, Dickinson, Jack, Sutton, van den Broek & Lightbody, 2011) psychological problems. However, the emerging findings are mixed for both intervention studies (Ownsworth and Gracey, 2017) as well as studies seeking to identify those factors that serve as vulnerabilities for later problems, making it difficult to develop and apply individually tailored psychological interventions based upon less well-evidenced mechanisms.

One of the most fruitful lines of enquiry relates to the role of coping style as a mediator between the stressful impact of an ABI and a range of later emotional outcomes (Lubusko, Moore, Stambrook, & Gill, 1994; Moore, Stambrook & Peters, 1989). The stress-appraisal-coping model based on that proposed by Lazarus & Folkman (1987) has been applied as a general model of post injury adaptation (Godfrey, Knight, & Partridge, 1996; Kendall & Terry, 1996). Moore & Stambrook (1995) suggested that coping style might be the 'final pathway' between BI sequelae and outcomes. Appraisals, coping and self-regulatory processes such as goal discrepancy feature in more recent transdiagnostic models (Brands, Wade, Stapert, & van Heugten, 2012; Gracey, Ford, & Psaila, 2015; Park, 2010). Although there is some variability in findings across studies, in general coping styles that reflect a task or problem-oriented approach appear to be adaptive, whereas coping styles focused on the emotional impact such as avoidance, worry, substance use or wishful thinking appear less adaptive (Anson & Ponsford, 2006; Curran, Ponsford, & Crowe, 2000), echoing Goldstein's (1952) insight that the consequences of ABI arise from three processes: the direct effects of brain damage, the emotional 'catastrophic reaction' to these changes, and loss of abilities due to *avoidance* of the catastrophic reaction.

Another common determinant of psychosocial outcomes is impairment of executive functioning (Douglas, 2010; Spitz, Ponsford, Rudzki, & Maller, 2012). Executive functions incorporate potentially adaptive skills such as inhibitory control, flexibility and problem-solving ability. It has therefore been hypothesized that there may be interactions between level and type of acquired executive functioning deficit, coping style and emotional outcome following ABI (Gracey, Evans, & Malley, 2009; Gracey et al., 2015; Rutterford & Wood, 2006; Spitz, Schönberger, & Ponsford, 2013). This idea has received some attention in non-brain injured groups, with Williams, Suchy, & Rau (2009) proposing individual differences in EF in the general population might underpin variations in vulnerability to stress, with weaker EF being associated with greater stress vulnerability. Compton et al. (2011) employed neuropsychological assessment of cognitive control (Stroop task), behavioural assessment of emotional reactions to stresses over a 14-day period, and ECG data, in a healthy sample, finding that specific aspects of cognitive control were associated with less adaptive coping and greater physiological stress responses. Grech et al. (2015) studying interaction between EF and coping in multiple sclerosis, found EF mediated the relationship between adaptive coping and psychosocial outcome: having better EF meant a greater chance of better emotional outcomes despite less adaptive coping style. Whilst Allott et al.'s (2015) findings were consistent with this for their healthy control group (better working memory buffered against stress), the first episode psychosis (FEP) patient group showed a contrary relationship, suggesting the effects of interaction between cognition and coping on emotional outcomes may vary across healthy and clinical groups.

Krpan, Levine, Stuss, & Dawson (2007) tested whether impaired executive functioning would compromise ability to use problem-oriented coping, leading to increased default use of emotion-focused coping. The results were supportive of study hypotheses within the TBI, but not healthy control group. However, the study employed multiple comparisons and the sample size was small given the regression analyses employed. Furthermore, the impact of the interaction between EF and coping on emotional outcomes was not investigated. Rutterford & Wood (2006) explored prediction of psychosocial outcome in a group of people with ABI 10 or more years post injury, testing the hypothesis that cognitive deficits would have both direct and indirect (via coping style) impacts on outcome, as proposed by Kendall & Terry (1996). The results were only partly supportive of Kendall and Terry's model, with no evidence for indirect effect of cognition on social outcomes via coping. Spitz et al (2013) also tested both direct and indirect effects of cognitive impairment on anxiety and depression, via the mediating role of coping and appraisal, in an ABI sample 6 to 53 months post injury. They found that good performance on an executive verbal fluency test was significantly positively associated with adaptive coping, consistent with Krpan et al (2007). However, they did not find mediation of the effect of cognition on mood by coping style. Only two executive tests were used neither of which directly assessed the solution generation or problem-solving aspects of EF that one might presume are implicated in the relationship between adaptive problem oriented coping and mood.(Wolters Gregório, Ponds, Smeets, Jonker, Pouwels, Verhey, & Heugten (2015) investigated whether executive functioning mediates the relationships between coping style and depression and quality of life in healthy controls and people with ABI with at least 1 neuropsychiatric symptom. Using the Trail Making Test and Stroop test to assess executive functioning, the authors did not find the predicted mediating relationship. However, analysis of self-rated EF using the Frontal Systems Behaviour Scale (FrSBe; Grace & Malloy, 2001) did find support for their predictions: lower levels of self-rated EF were associated with passive coping, and in turn depression and lower QoL; in the high self-rated EF group, problem-focused coping was associated with better QoL. Similarly, Rakers et al. (2017), reporting on coping style and EF amongst moderate-severe TBI participants found greater self-reported EF difficulties associated with more passive and less active coping styles. Of the behavioral measures of EF used, only mental flexibility was associated with coping style.

Therefore, whilst there is a theoretical argument for possible relationships between coping style, EF and mood, findings are mixed and inconclusive. It appears that, in MS and ABI groups, impaired EF may be associated with less problem solving and more emotion-focused coping. In addition, there is some indication that the relationship between coping style and emotional outcomes is stronger for those with lower EF, perhaps because they are less able to implement adaptive coping strategies. However, findings are inconsistent, particularly where cognitive tests, rather than self-rating measures have been employed. The variability in findings may stem from differences in methodologies and in the predicted relationships being tested. Wood and Rutterford's sample were at least 10 years post-injury, whereas Wolters et al included only patients with neuropsychiatric problems. Use of self-rating scales is problematic due to likely shared variance amongst items relating to emotion regulation and problem solving which feature in scales of both coping style and of EF symptoms. The studies of Krpan et al (2011), and Rakers et al (2017) were the only ones to employ a range of EF measures including measures sensitive to problem solving, the most likely candidate for an interaction with adaptive problem-oriented coping and mood. However, neither study measured emotional outcomes. Studies also varied with regard to the specific measures of coping style employed, and as such therefore also varied with regard to what exactly constituted adaptive or maladaptive coping styles.

In the current study we therefore set out to explore the relationship between coping style, EF and three different, common forms of emotional distress. Assuming the relationship between coping style and mood is well established within and outside of the ABI population, our interest is in whether the specific effects of acquired EF impairment alter this relationship. Therefore, we predict that, after accounting for demographic and injury related variables and general intellectual functioning:

1. Specific EF skills associated with goal management (problem solving, plan development and deployment, and maintenance of attention to goals) and coping style will be associated with emotional distress
    a. Consistent with prior studies, more emotion focused, avoidant and less task-oriented coping will be associated with greater emotional distress (depression, tension and aggression)
    b. EF will make a significant contribution to variance in emotional distress (depression, tension and aggression)
2. The interaction between specific EF skills and specific adaptive (task oriented) and maladaptive (avoidant, emotion focused) coping styles will be significantly associated with emotional outcomes (controlling for demographic variables, intellectual functioning and aetiology) such that those with lower EF show a stronger relationship between coping style and emotional outcomes compared to those with better EF

Method

Ethics:

Ethical approval was provided by a UK National Health Service Research Ethics Committee (study reference 08/H0306/45) and the relevant Research and Development Department for each of the health service providers involved in recruitment of participants. All participants provided written informed consent to participate. If consent was withdrawn after anonymization and data entry, data was retained for analysis.

Design:

A cross-sectional, correlational design employing multiple regression was used to develop and explore models to account for different emotional outcomes (Depression; Tension/Anxiety; Aggression/Hostility) from dispositional coping style and EF independently and in interaction.

Participants:

Participants were individuals who had consented to participate in a trial of rehabilitation of everyday executive deficits [author information masked] who had completed baseline measures prior to starting the trial. Inclusion criteria for the trial and therefore for this study were as follows:

- aged 18 or over
- non-progressive brain injury acquired in adulthood
- more than one year post-injury
- clinician, carer or self-reported everyday organization and memory problems

- able to use a mobile phone (required as part of rehabilitation intervention trial)

Exclusion criteria:

- memory impairment of sufficient severity to limit retention of intentions and training information required for the trial (clinical judgment and neuropsychological assessment)
- patient or carer participant with severe and enduring mental health problem, or substance misuse or dependency, as identified by referring clinician
- participation in a rehabilitation intervention with significant overlap with the study intervention

Participants were recruited from a range of community rehabilitation services and also from the Cambridge Cognitive Neuroscience Research Panel (CCNRP; people with ABI who gave prior consent to be approached for relevant research studies). As previously reported [author information masked] 93 individuals were assessed for eligibility for the trial, of whom 83 consented and completed initial assessments and started the baseline phase of the trial. Of these 83, 14 were excluded from the current analysis due to significant levels of missing data. Data from 69 individuals were thus used in the current analysis. Information regarding the sample characteristics is provided in Table 1. In summary, of the 69 included, 43 (62.3%) were male, 34 had experienced a moderate–severe TBI, 22 stroke and 13 other ABI.

Mean age was 47.8 years (s.d. 14), mean years of education were 12.5 (range 10 – 21 years) and mean years post injury was 6.9 years (range 0.9 – 39.8 years).

Table 1 about here

Procedure:

Participants were assessed as part of the larger trial at their homes, at a rehabilitation service or at the research institution.

Measures

1. Coping style:

The Coping Inventory for Stressful Situations (CISS; Endler, Parker, Ridder, & van Heck, 2004) which has been validated for use with ABI (Simblett, Gracey, Ring, & Bateman, 2015), was used to evaluate self-rated dispositional coping style. The Rasch analysis conducted by Simblett et al (2015) using data collected from people with ABI provides guidance on rescoring to provide improved reliability and construct validity of subscales. The CISS data was summarized according to the 3 subscales identified by Simblett et al (2015; Task, Avoidance and Emotion) and in accordance with their rescoring guidance, so as to maximize the fit of data to interval-level measurement.

2. General intellectual functioning:

The National Adult Reading Test (NART Nelson, 1982) was used to derive an estimate of pre-morbid general intelligence. The Wechsler Adult Intelligence Scales-III-UK

(Wechsler, 1999) Matrix Reasoning subtest was used to assess current general intellectual functioning.

3. Neuropsychological measures of EF:

Two measures of executive functioning were included associated with sustaining attention to goal directed behavior, planning and problem solving. The Sustained Attention to Response Test (SART; Manly & Robertson, 2005) is a type of go no-go task in which the target stimuli occur relatively infrequently. The task places demands on ability to sustain attention to the goal of the task, and poor performance in terms of number of commission errors (pressing when one shouldn't) has been shown to be associated with TBI / frontal damage and has been considered a metric of 'goal neglect' (Manly, Hawkins, Evans, Woldt, & Robertson, 2002; Manly & Robertson, 2005) and could be considered a measure of the 'task setting' and 'monitoring' components of executive functioning described by (Stuss, 2011).

The Hotel Test (Manly et al., 2002) is similar to the Modified 6 Elements (Burgess et al., 2000) task in that the person is asked to complete some of each of 5 different tasks, within a set time and according to a set of rules. The test is considered a measure of higher-level EF incorporating multiple skills such as planning, problem solving, prospective memory and goal maintenance, and could be considered a measure of 'metacognitive' skills according to (Stuss, 2011). The Hotel Task is scored in terms of time deviation from the 'ideal' solution of spending equal amounts of time on each subtask.

4. Dependent variables – emotional outcomes:

The Profile of Mood States POMS (McNair, Lorr, & Droppleman, 1992) was used to assess depression, tension-anxiety, and anger-hostility. It is a validated and psychometrically robust measure and has been used to evaluate emotional outcomes in a range of patient groups including pain, surgery, cancer, mental health and brain injury (Hodgson, McDonald, Tate, & Gertler, 2005; Nyenhuis, Yamamoto, Luchetta, Terrien, & Parmentier, 1999).

Analysis:

Analysis was conducted with SPSS version 23 and R Studio. Data were checked and cleaned, and CISS items were rescored as per guidance of Simblett et al. (2015). Despite having a general prediction about the contribution of EF to the coping–mood relationship, given the wide range of possible permutations of interaction terms that could be tested arising from two EF terms and 3 coping terms, we opted to take a model fit approach, rather than significance testing one predetermined model. Specifically, we used the Akaike Information Criterion (corrected for use with smaller samples: AICc, Burnham & Anderson, 2002) which provides a metric against which to select the best fitting models. For the current study, 64 possible models including all main effects and combinations of interaction terms can be derived and compared. We opted to retain only those models within an AIC of 2 of the best-fitting model.

Multiple regression analyses were then conducted for each separate emotional dependent variable: POMS Depression, POMS tension-anxiety and POMS anger-hostility. Each regression equation simultaneously entered estimated premorbid IQ (NART), current intellectual functioning (WAIS-III Matrix Reasoning, MR; to control for any association between general intellectual functioning and EF), aetiology (TBI, stroke, other ABI) and POMS Fatigue, the three modified CISS subscale scores and two EF measures (SART EoC and Hotel TTD). To answer question 1, variables within the full models will be reported. To answer question 2, six interaction terms will be calculated (3 CISS subscales x 2 EF measures) and all possible interaction combinations entered and compared, and the AIC used to select the models with best fit (within 2 AIC). The models retained in this way were then inspected to determine: 1. Whether any of the models retained is a main effects model? If so this would be the most parsimonious model and it would be concluded that there is no meaningful contribution of any interaction terms, and 2. Whether only models including interaction terms are retained? If so, models were inspected to determine which of these is the most parsimonious and also most consistent with the other models retained. Plots to illustrate changes in the contribution of coping style to emotional outcome as level of EF ability changes were produced where the best fitting models included interaction terms.

Results:

Analyses were initially run to check assumptions for parametric analysis, identify outliers, and inspect residual plots. This identified that the residuals for POMS Anger were not normally distributed and square root transformation was conducted. The 3 regression analyses for each emotional outcome were run as previously described, are summarized in Table 2, below. Variance Inflation Factors (VIF) were generated to test for multi-collinearity of covariates – all analyses were found to fall within acceptable limits, with VIF being <2 which is well below the suggested cut off of ten (Hair Jr., Tatham, Anderson, & Black, 1998). The findings are set out for each emotional outcome, addressing each question: 1. Whether addition of the coping * EF interaction terms improves the models of Depression, Tension and Aggression. 2. Which terms are included in the best fitting models.

Insert Table 2 about here

1. **Depression**

Two models were retained both of which included interaction terms. We selected the best fitting and most parsimonious model which included the Hotel TTD * CISS task interaction term (*t* = -2.386; *p* = .020) only, the second model also included Hotel TTD * CISS emotion focused coping interaction. As shown in Table 2, the full model (F (11,57) = 14.24; Adjusted R^2 = .6817; *p* = .000) included significant contributions from aetiology (other ABI versus stroke and TBI), POMS Fatigue, modified CISS task and emotion focused coping, and Hotel TTD, confirming effects of coping and EF as predicted.

Figure 1 provides an illustration of how the association between task oriented coping style and depression changes with increasing Hotel TTD scores (decreasing EF ability). The plot suggests that the predicted negative association between task orient coping and depression (i.e. more task coping, lower depression; less task coping, greater depression) is strongest for those with poorest EF (largest Hotel TTD). The significant effect of Hotel TTD on depression in the base model (β

= .012; *t* = 2.55; *p* = .0154) suggests this interaction is driven by higher depression and lower task-oriented coping, a relationship which increases in strength as EF reduces, as predicted. However, as EF improves (Hotel TTD reduces) self-rated tendency to use task-oriented coping appears to have a progressively weaker association, and below Hotel TTD of approximately 450s the association may be positive so that less task-oriented coping is associated with less depression. This indicates a more complex interaction than that which was predicted.

Insert Figure 1 here

2. POMS Tension – Anxiety

Eleven models were retained all within 2 AIC, indicating multiple similar models of equivalent fit. All models included interaction terms consistent with our hypothesis of the contribution of interaction between coping style and EF to emotional outcomes. Six of the models included Hotel TTD * CISS Task coping, eight models included SART EoC * CISS Emotion focused coping and five included Hotel TTD * CISS Emotion coping interaction terms. The most parsimonious model with the lowest AIC and greatest consistency across all retained models included only Hotel TTD * CISS Task coping and SART EoC * CISS Emotion focused coping interaction terms. However, the interaction terms did not reach significance at the p = .05 level within the model. The full model (F (12, 56) = 13.78; Adjusted R^2 = .6928; p = .000) shows that, as with POMS depression, aetiology (other ABI versus stroke and TBI, POMS Fatigue, modified CISS task and emotion focused coping, and Hotel TTD) were significant in the model for POMS Tension-Anxiety, again confirming effects of coping style and EF as predicted.

Figure 2 illustrates the nature of the interaction for the effects of Hotel TTD on the relationship between task oriented coping style and POMS Tension-Anxiety, and Figure 3 the effect of SART EoC on the relationship between emotion-focused coping and POMS Tension-Anxiety. Figure 2 shows a similar pattern to that for depression, with poorer Hotel performance being associated with a stronger negative relationship between task coping and depression. Figure 3 suggests that as number of SART commission errors increases, the effects of emotion-focused coping on POMS Tension-Anxiety increase, which is consistent with the hypothesized prediction.

Insert Figures 2 and 3 here

3. Anger-hostility

One model was identified with no other models falling within 2 AIC, and this model did not include any interaction terms. The full model (F (10, 58) = 6.827; Adjusted R^2 = .4615; *p* = .000) and terms are set out in Table 2. Aetiology, POMS Fatigue and, consistent with predictions, emotion coping, were significant in the model, however neither EF term made a significant contribution.

Discussion

This study systematically analyzed emotional outcomes in people with ABI recruited on the basis of everyday executive functioning problems, to determine the direct and indirect contributions of coping style and executive functioning. In partial support of study hypotheses, CISS task-oriented coping style and EF as measured by the Hotel task time deviation score were significant in models of depression and tension-anxiety, but not anger-hostility. Also, in partial support of study hypotheses, the best fitting models included coping-EF interaction terms for depression and tension-anxiety, but not anger-hostility. Both models included an interaction between Hotel task performance and task-oriented coping. Estimated pre-morbid and current intellectual functioning variables were included in the models therefore the effects of EF can be considered as independent from any effect of general intellectual functioning.

Coping style and emotional outcomes

The significant contribution of adaptive (task oriented) or maladaptive (emotion-focused) coping styles to different emotional outcomes, including anger-hostility, has been demonstrated, as shown previously in relation to anxiety and depression (Anson & Ponsford, 2006; Curran et al., 2000; Moore et al., 1989). Avoidant coping was not associated with any emotional outcome independently or in interaction with EF despite previous findings (Kegel, Dux, & Macko, 2014; Riley, Dennis, & Powell, 2010) and models that indicate avoidance may be a particularly important maladaptive coping style following brain injury (e.g. Gracey et al, 2015). One possible reason for this is that the CISS Avoidance measure incorporates two further subscales, task-oriented distraction, and person-oriented social diversion, which may show differential effects. For example, previous studies suggest that social connection is adaptive following stroke (Haslam et al., 2008; Jones et al., 2011) and keeping to oneself maladaptive (Curran et al., 2000). Few prior studies have investigated anger or aggression and coping style. Our study indicates a possible role for greater emotion-focused coping in increasing anger-hostility. A number of hypotheses regarding this finding can be generated based on the existing cognitive-behavioral and neuropsychological literature as outlined by Medd & Tate (2000). Firstly, it is possible that a continued focus on angry or hostile thoughts or feelings might serve to maintain or intensify angry feelings and behaviors. Novaco (1976) argued that anger expression might enable people who feel vulnerable or anxious to experience a rewarding sense of (attempted) control. Consistent with this, Gracey et al (2015) argue that aggressive behavior post ABI can often be formulated as a response to 'threat to self' and as such might arise where individuals fail to engage an adaptive regulatory coping strategy (such as arousal reduction, leaving the situation, or addressing the trigger problem). Second, aggressive behavior might arise as a consequence of neuropsychological disinhibition or failure of emotion regulation. Shields et al's (2015) study suggests emotion regulation maybe a transdiagnostic factor underpinning depression, anxiety and stress. The current study tentatively indicates that failed emotion regulation via emotion focused coping, but not reduced EF, might also underpin problems with aggression.

Fatigue and emotional outcomes

POMS Fatigue emerged as associated with all three emotional outcomes. This is consistent with previous research by Johansson & Ronnback (2014) who identified a significant overlap between specific mood and fatigue scale items, including poor concentration, irritability and lack of initiative. Studies have also indicated subjective fatigue to be associated with subjective mental effort to complete tasks (Azouvi et al., 2004), which might arise following brain injury due to neurological, cognitive or psychological changes with increased sense of effort and slowness conceivably influencing depressed feelings, anxiety (through worry about functioning) or aggression (through frustration). Although Grech et al.'s (2015) study of mood, coping and EF in MS did not include a measure of fatigue, they found less effortful coping styles such as acceptance and growth to be associated with better outcomes amongst those with poorer EF. Given the significance of this symptom to people with stroke and TBI, the current study indicates that further research exploring the interplay between cognition, mood, effortful versus effortless (e.g. growth, acceptance) coping, and fatigue might be warranted.

Interaction between coping and EF

We had hypothesized that poorer EF would be associated with a stronger effect of unhelpful coping style on all three emotional outcomes. The inclusion of SART EoC by emotion-focused coping interaction in the model for POMS tension-anxiety is consistent with hypotheses, showing an increasing effect of emotion-focused coping on tension-anxiety as EF reduced. The Hotel task emerged as associated with depression and tension-anxiety on its own (poorer EF, greater anxiety or depression) and in interaction with task-oriented coping. This is partially consistent with previous findings in healthy controls and other neurological populations (Compton et al., 2011; Grech et al., 2015; Williams et al., 2009) in that the previously demonstrated negative association between task coping and mood was stronger for those with poorer EF. The negative association of task-oriented coping with depression was strongest for those with lower functioning EF, consistent with Krpan et al (2011) who concluded that poorer EF was associated with less ability to use task-oriented coping, therefore greater default use of emotion-focused. The current study goes further indicating that this pattern is associated with poorer emotional outcomes for anxiety and depression, but not anger, as predicted, but not confirmed, in previous studies with ABI participants (Rutterford & Wood, 2006; Spitz et al., 2013; Wolters Gregório, Ponds, Smeets, Jonker, Pouwels, Verhey, & van Heugten, 2015). However, contrary to predictions and to prior studies (although Curran et al. (2000) found greater problem-solving coping was not associated with lower depression in people with TBI), those with better EF showed less task-oriented coping to be associated with lower, rather than higher, depression and anxiety.

This pattern of results could be interpreted in a number of ways. First, as suggested by Krpan et al. (2007), the relationship between task-oriented coping, metacognitive EF (rather than more focused cognitive EF skills, as described by Stuss, 2011), and mood could be accounted for by lower EF impairing ability to use problem or task-oriented coping. In addition, impaired monitoring and task setting abilities might interfere with adaptive emotion regulation abilities (impacting anxiety), consistent with cognitive-affective regulation models such as that of Ochsner & Gross (2005) in which verbal down-regulation of negative affect relies on core executive skills. It is also possible that an underlying common variable (as suggested by Williams et al., 2009) or diffuse damage to frontal networks independently gives rise to both impaired self-regulation of affect, on the one hand, and impaired EF (as shown by independent contribution of Hotel task to anxiety and depression) on the other.

These accounts are not mutually exclusive, so it is possible that for a given individual, each of these processes might contribute to emotional outcomes.

Limitations and strengths

The study findings need to be considered in the context of limitations. The correlational design of the current study makes it difficult to interpret the results in terms of direction of effect, therefore it remains open whether higher depression is impacting upon EF, or whether poor EF and subsequent greater emotion-focused and less task-oriented coping are increasing vulnerability to depression, or there is a combination of both effects. In addition, the sample was selected on the basis of self-reported or clinician identified everyday EF problems, rather than assessment using specific standardized assessments of EF, and the aetiologies were mixed resulting in greater heterogeneity of anatomical damage and cognitive profiles. Whilst the sample size was adequate for the multiple regression analysis, confidence in the results would be increased with a larger sample. Finally, the measures of coping style and mood rely on self-report and therefore responses of participants with impaired self-awareness might not be valid indictors of actual coping style or mood. Against that, study strengths include use of behavioral tests of executive functioning that are conceptually better aligned with possible general regulatory or metacognitive skills (Stuss, 2011) requiring integration of multiple lower order processes, as also employed by Krpan et al. (2011) and Rakers et al. (2017). Employing the AIC approach to determine the best fitting models provided a greater degree of flexibility regarding exploration of possibly important interaction effects, in contrast to traditional model testing approaches wherein a greater degree of certainty regarding the tested interactions is required, risking a possible false negative result. Future studies should also employ longitudinal designs, for example as employed by Spitz et al (2013) and include more complex measures of higher-level EF or metacognition. Clinical implications of the study indicate the importance of assessment to formulate an individual's profile of stress and coping efforts in the context of life after brain injury in conjunction with assessment of specific aspects of executive and metacognitive functioning. Attention to fatigue when considering emotional needs is also highlighted. This information should help guide choice, and adaptation, of intervention approach.

Conclusions

This study is one of few with brain injured participants that has found evidence of interaction between metacognitive and core executive functions and self-reported coping style, employing behavioral rather than self-rating measures of EF. Those with lower EF on the Hotel Task appeared to be less able to make use of adaptive task-oriented coping (for depression and anxiety), and worse affected by maladaptive emotion-focused coping styles (for anxiety). Emotion focused coping and fatigue emerged as possible transdiagnostic predictors of all three emotional outcomes. However, whilst the findings indicate further research using more complex, ecologically valid or metacognitive tests may be fruitful, the correlational design limits conclusions regarding causality. Future studies should seek to identify basic processes that might account for links between subjective fatigue, executive and metacognitive functions and emotion regulation, employing longitudinal designs.

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	N	Mean (s.d.)	s.d.
Age (years)	68	47.8 (min. 21 years – max. 72 years)	14
Time since injury (years)	68	6.9 (min. 0.9 years – max. 39.8 years)	7.1
Education (years)	65	12.5 (min. 10 – max. 21 years)	2.8
NART estimated IQ	69	101.4	13.2
WAIS-III Matrix Reasoning standardized score	69	11.9	3.2
D-KEFS Letter Fluency standardized score	69	7.6	3.8
POMS Total Mood Disturbance	69	53.0	43
		n	**%**
Injury type: TBI Stroke Other	69	 34 22 13	 49 31.9 18.8
Gender: Male Female	69	 43 26	 62.3 37.7

Table 1: Demographic and baseline clinical characteristics of the sample. (NART = National Adult Treading Test; WAIS = Weschler Adult Intelligence Scales; D-KEFS – Delis-Kaplan Executive Functioning System; POMS = Profile of Mood States)

	POMS Depression				POMS Tension-Anxiety				POMS Anger-Hostility (sq. root transformed)			
	β	std error	t value	*P*	β	std error	t value	*P*	β	std error	t value	*P*
intercept	-33.010	13.310	-2.480	0.016*	-9.232	8.918	-1.035	0.305	0.127	1.788	0.071	0.944
NART estimated IQ	0.108	0.093	1.172	0.246	0.018	0.061	0.289	0.774	0.002	0.015	0.154	0.878
WAIS Matrix Reasoning	-0.297	0.326	-0.911	0.366	-0.414	0.214	-1.938	0.058	0.007	0.056	0.126	0.900
Aetiology (stroke vs other+TBI)	-1.699	2.444	-0.695	0.490	0.801	1.604	0.499	0.619	**-0.915**	**0.422**	**-2.170**	**0.034***
Aetiology (other vs TBI + stroke)	**-8.353**	**2.875**	**-2.906**	**0.005****	-2.303	1.875	-1.229	0.224	**-1.060**	**0.489**	**-2.167**	**0.034***
POMS Fatigue	**0.798**	**0.134**	**5.965**	**0.000*****	**0.517**	**0.088**	**5.856**	**0.000*****	**0.075**	**0.023**	**3.259**	**0.002****
CISS Task	**0.626**	**0.254**	**2.462**	**0.017***	**0.420**	**0.166**	**2.532**	**0.014***	0.031	0.022	1.387	0.171
CISS Emotion	**1.460**	**0.235**	**6.203**	**0.000*****	**0.616**	**0.220**	**2.796**	**0.007****	**0.155**	**0.041**	**3.816**	**0.000*****
CISS Avoid	0.197	0.281	0.702	0.486	0.022	0.186	0.120	0.905	0.045	0.048	0.938	0.352
Hotel TTD	**0.049**	**0.016**	**3.046**	**0.003****	**0.022**	**0.010**	**2.103**	**0.040***	0.000	0.001	0.413	0.681
SART EoC	-0.001	0.154	-0.006	0.995	-0.044	0.168	-0.261	0.795	-0.032	0.026	-1.212	0.231
Hotel * CISS Task	**-0.001**	**0.001**	**-2.386**	**0.020***	-0.001	0.000	-1.894	0.063				

Hotel * CISS Emo												
Hotel * CISS Avoid												
SART * CISS Emo					0.033	0.018	1.830	0.073				
SART * CISS Avoid												
SART * CISS Task												
Model	F (11, 57) = 14.24 p = 0.000 Adjusted R^2 = .6817				F (12, 56) = 13.78 p = 0.000 Adjusted R^2 = .6928				F (10, 58) = 6.827 p = 0.000 Adjusted R^2 = .4615			

Table 2: Summary models from multiple regression analyses showing main effects and interaction terms for Depression, Tension-Anxiety and Anger-Hostility (NART = National Adult Treading Test; WAIS = Weschler Adult Intelligence Scales; POMS = Profile of Mood States; EF = Executive Functioning; CISS = Coping Inventory for Stressful Situations; Emo = emotion-focused coping; avoid = avoidance coping; task = task-oriented coping; Hotel TTD = Hotel total time deviation score; SART EoC = Sustained Attention to Response Task Errors of Commission score; *** p < .001; ** p < .01; * p < .05).

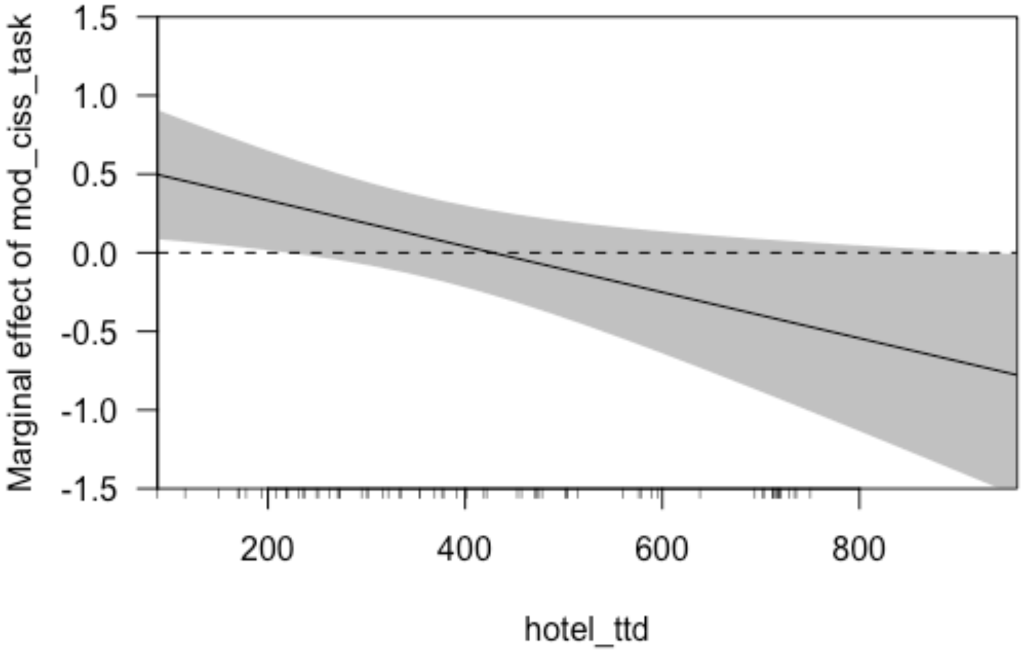

Figure 1: Plot showing how the relationship between POMS depression and CISS task-oriented coping changes as Hotel Test performance deteriorates.

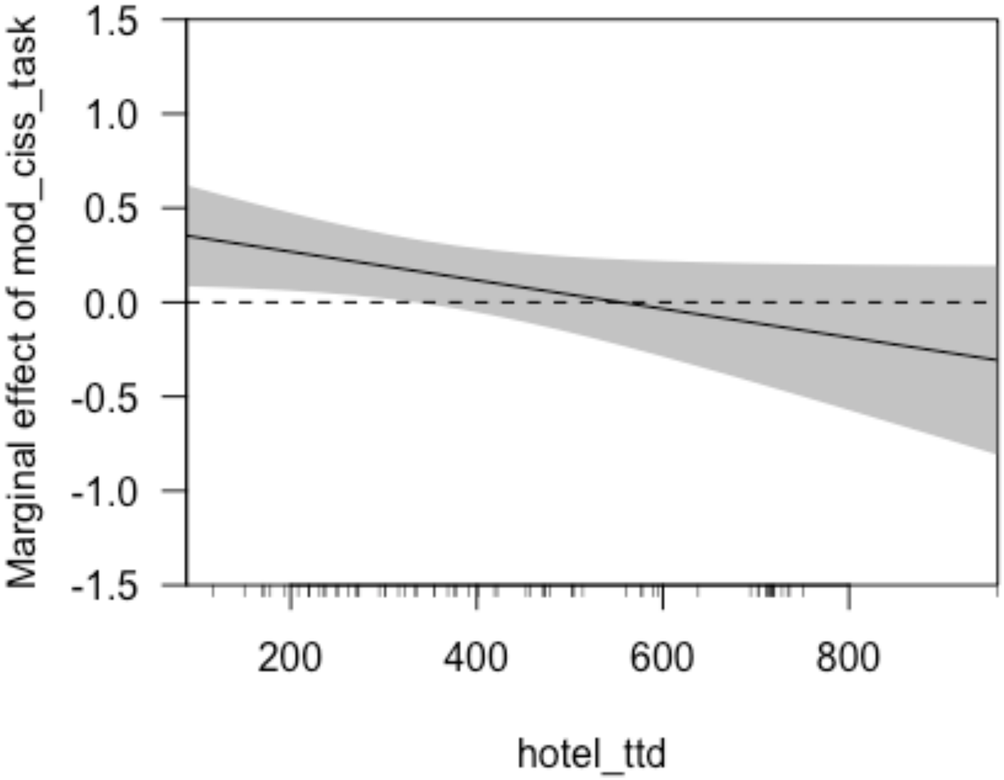

Figure 2: Plot showing the change in relationships between POMS tension-anxiety and CISS task-oriented coping as Hotel Test performance deteriorates.

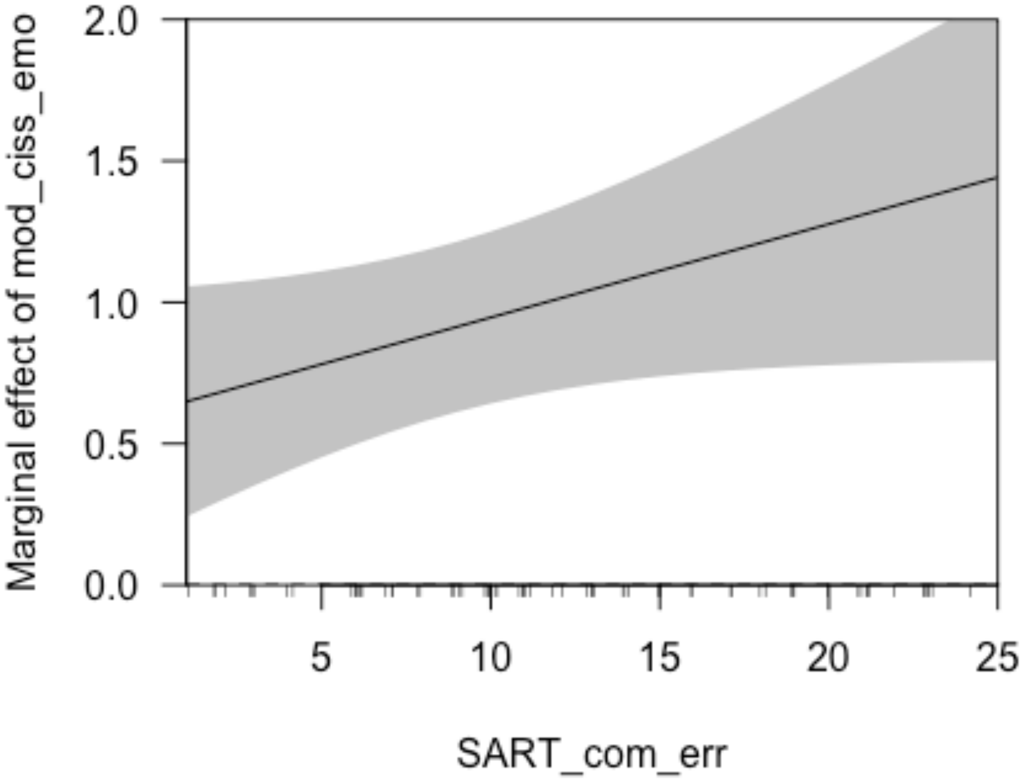

Figure 3: Plot showing the change in relationship between CISS emotion-focused coping and tension-anxiety as SART commission errors increase.

4.2 Summary of findings

The study findings confirmed previous research indicating coping styles are significantly associated with all emotional outcomes. The study also indicated that higher level frontal functions (as measured by the Hotel task) showed an association with anxiety and depression but not anger. Furthermore, depression and anxiety both appeared associated with interactions between Hotel task, and task-oriented coping, and anxiety was also associated with interaction between Sustained Attention to Response Test (SART) and emotion focused coping. As such it can be concluded that 'cold' frontal functions relating to regulation of attention and more complex multitasking and problem-solving skills interact with emotion-focused and task-oriented coping, such that those with better performance are less affected by maladaptive emotion-focused coping, and those with poorer performance are less able to make use of task-oriented coping to reduce depression.

CHAPTER 5

5 Is it possible to enhance goal management skills with automated (SMS text) reminders to improve achievement of everyday tasks?

Gracey, F., Fish, J. E., Greenfield, E., Bateman, A., Malley, D., Hardy, G., Ingham, J., Evans, J. J. and Manly, T. (2016). A randomized controlled trial of Assisted Intention Monitoring (AIM) for the rehabilitation of executive impairments following acquired brain injury (ABI). *Neurorehabilitation and Neural Repair,* 31(4), 323-333.

5.1 Introduction to the paper

The previous 3 studies addressed issues of the contribution of, and interaction between 'hot' and 'cold' frontal functions in relation to social and emotional outcomes to improve understanding of variability between clinical assessments and specific aspects or predictors of everyday functioning. It is increasingly recognised that those presenting for rehabilitation of EF's present a 'double whammy': the difficulties they have are the very abilities required to aid successful adaptation of new behaviours and transfer into everyday life. Prior to this study, whilst there had been some indication of benefit of goal-management type interventions, questions remained about potential application of 'content free cuing' for achievement of everyday intentions, and how to enhance far transfer of goal-management type interventions into everyday life. Theoretically, models such as the Supervisory Attentional System model predict that periodic 'mental review' (stopping and reflecting on one's ongoing goals and intentions) might support maintenance of goals and action schema in working memory. The 'Assisted Intention Monitoring' trial sought to test the potential benefit of mobile phone SMS text reminders delivered at random intervals during the trial active intervention phases on completion of participants' everyday plans and intentions.

Original Research Article

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A Randomized Controlled Trial of Assisted Intention Monitoring for the Rehabilitation of Executive Impairments Following Acquired Brain Injury

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Abstract

Background. Acquired brain injury (ABI) can impair executive function, impeding planning and attainment of intentions. Research shows promise for some goal-management rehabilitation interventions. However, evidence that alerts assist monitoring and completion of day-to-day intentions is limited. *Objective.* To examine the efficacy of brief goal-directed rehabilitation paired with periodic SMS text messages designed to enhance executive monitoring of intentions (assisted intention monitoring [AIM]). *Methods.* A randomized, double-blind, controlled trial was conducted. Following a baseline phase, 74 people with ABI and executive problems were randomized to receive AIM or control (information and games) for 3 weeks (phase 1) before crossing over to either AIM or no intervention (phase 2). The primary outcome was change in composite score of proportion of daily intentions achieved. A total of 59 people (71% male; 46% traumatic brain injury) completed all study phases. *Results.* Per protocol crossover analysis found a significant benefit of AIM for all intentions [$F(1, 56) = 4.28$; $P = .04$; $f = 0.28$; 3.7% mean difference; 95% CI = 0.1%-7.4%] and all intentions excluding a proxy prospective memory task [$F(1, 55) = 4.79$; $P = .033$; $f = 0.28$, medium effect size; 3% mean difference; 95% CI = 0.3%-5.6%] in the absence of significant changes on tests of executive functioning. Intention-to-treat analyses, comparing AIM against control at the end of phase 1 revealed no statistically significant differences in the attainment of intentions. *Conclusion.* Combining brief executive rehabilitation with alerts may be effective for some in improving achievement of daily intentions, but further evaluation of clinical effectiveness and mechanisms is required.

Keywords

brain injuries, rehabilitation, executive function

Introduction

Impairments in executive functioning are common following acquired brain injury (ABI) involving the prefrontal cortex[1,2] and are associated with poorer functional and social outcomes.[3,4] Executive processes include breaking down a complex goal into a series of ordered subgoals that determine behavior, holding the steps and overarching goal in mind, constraining attention and behavior to the main goal, and weighing its priority against competing demands that may arise.[1,5-7] When a goal cannot be executed immediately it becomes a prospective memory (PM)[8] that does not remain at the forefront of consciousness but remains latent, to be recalled at the appropriate time (time-based PM), when the appropriate opportunity arises (event-based PM), or at some future stage (step PM[9]). PM failure can result from memory difficulties (forgetting the plan) and executive difficulties[8] (failure to act despite memory of one's intention, also known as goal neglect[10]). Rehabilitation of executive functioning is, therefore, inherently challenging because the capacities that maximize adaptive change, including ability to transfer rehabilitation from clinic to everyday life, are compromised, resulting in reduced effectiveness of rehabilitation[11-13] and poorer emotional outcomes.[14]

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Interventions for executive deficits such as Goal Management Training (GMT[5,15]) emphasize effective implementation of intentions to varying degrees. Typically run in groups over 8 or more sessions, GMT includes education to develop awareness and structured practice of goal setting, self-monitoring, and managing competing distractions.[16] Reviews of intervention studies favor metacognitive strategy training (incorporating self-monitoring and self-regulation)[17] and approaches combining GMT with other strategies such as supports for transfer into daily life[16] over stand-alone goal management. The latter review concluded that proof of principle was demonstrated for studies of content-free cues provided at random intervals for improving goal-directed behavior during brief (10-15 minute), complex office-based tasks.[18,19] However, whereas the international INCOG guideline for rehabilitation of executive impairment supports the use of metacognitive strategy training,[20] the INCOG guideline for rehabilitation of attention deficits[11] states that evidence for periodic content-free cues is conflicting and further clinical outcome studies are required. A functional imaging study failed to find beneficial effects of periodic alerts on the Sustained Attention to Response Test (SART) but did show reduced right dorsolateral prefrontal activation during provision of alerts. This was interpreted by the authors as indicating that cues assisted the maintenance of intentions by reducing reliance on specific endogenous control processes underpinned by the right frontoparietal control and attention networks involved in sustaining attention to task goals.[21] A recent trial[22] found that GMT incorporating text message reminders resulted in gains on self-report and neuropsychological measures, although the independent contribution of cueing was not evaluated. Previous trials have used questionnaires or neuropsychological tests rather than real-world behavioral measures to evaluate outcome. In one exception to this, Fish et al[23] evaluated transfer of training on a naturalistic task of remembering to make phone calls at set times each day over a 2-week period. Participants with ABI learned specified times to call the study's answerphone, then received very brief (30-minute) GMT in which the process of pausing current activity to mentally review one's intentions was linked with a cue phrase ("STOP"; Stop, Think, Organize, Plan). STOP cues were provided on randomly selected days at random intervals. Cued days were associated with significantly more, and more accurately timed, calls than noncued days. Although promising for potential application in rehabilitation, the effectiveness in terms of participants' own everyday intentions and potential effect on emotional outcomes were not evaluated. Further evaluation of the effect of combined brief GMT and cueing on everyday goals is, therefore, required.

Here we report a trial examining the efficacy of assisted intention monitoring (AIM) comprising brief GMT followed by randomly timed SMS text messages for improving achievement of everyday intentions. The broad aim was to extend prior research using GMT plus periodic alerts to evaluate potential efficacy in improving achievement of everyday intentions. The primary outcome was a composite score of proportion of all intentions achieved, made up of different types of intention and an objectively scored proxy task (the phone task). The primary study hypotheses were the following:

1. the proportion of all intentions achieved will be significantly greater during AIM than control phases, and
2. the proportion of all intentions achieved excluding the phone call task will be significantly greater during AIM than control phases.

A subsidiary hypothesis was that increased goal attainment would be associated with improved self-rated mood. Exploratory analyses were planned to identify factors that might influence response to intervention, a necessary process in the development of complex health care interventions.[24]

Method

Ethics

Ethical approval to conduct the study was provided by a National Health Service Research Ethics Committee (study reference 08/H0306/45) and the relevant Research and Development Department for each of the health services involved in recruitment of participants. All participants provided written informed consent to participate.

Trial Design

The study used a randomized controlled, parallel group crossover design with 3 phases (baseline phase, intervention phase 1, intervention phase 2), each of which lasted 3 weeks, with a 1-week break between phases for completion of measures (phases shown in Figure 1). Assessments and primary analyses were conducted blind to group allocation. Following consent, participants completed initial assessment questionnaires and neuropsychological tests and were supported in identifying daily intentions to be monitored for the study duration. They were then randomized to either AIM or control for intervention phase 1 (equal numbers in each), after which they crossed over to phase 2, during which AIM-first participants received no intervention or usual care and control-first participants received AIM. A conceptually symmetrical crossover was not possible for the AIM-first group because messages from the study had already been associated with reviewing intentions. The cessation of messages to

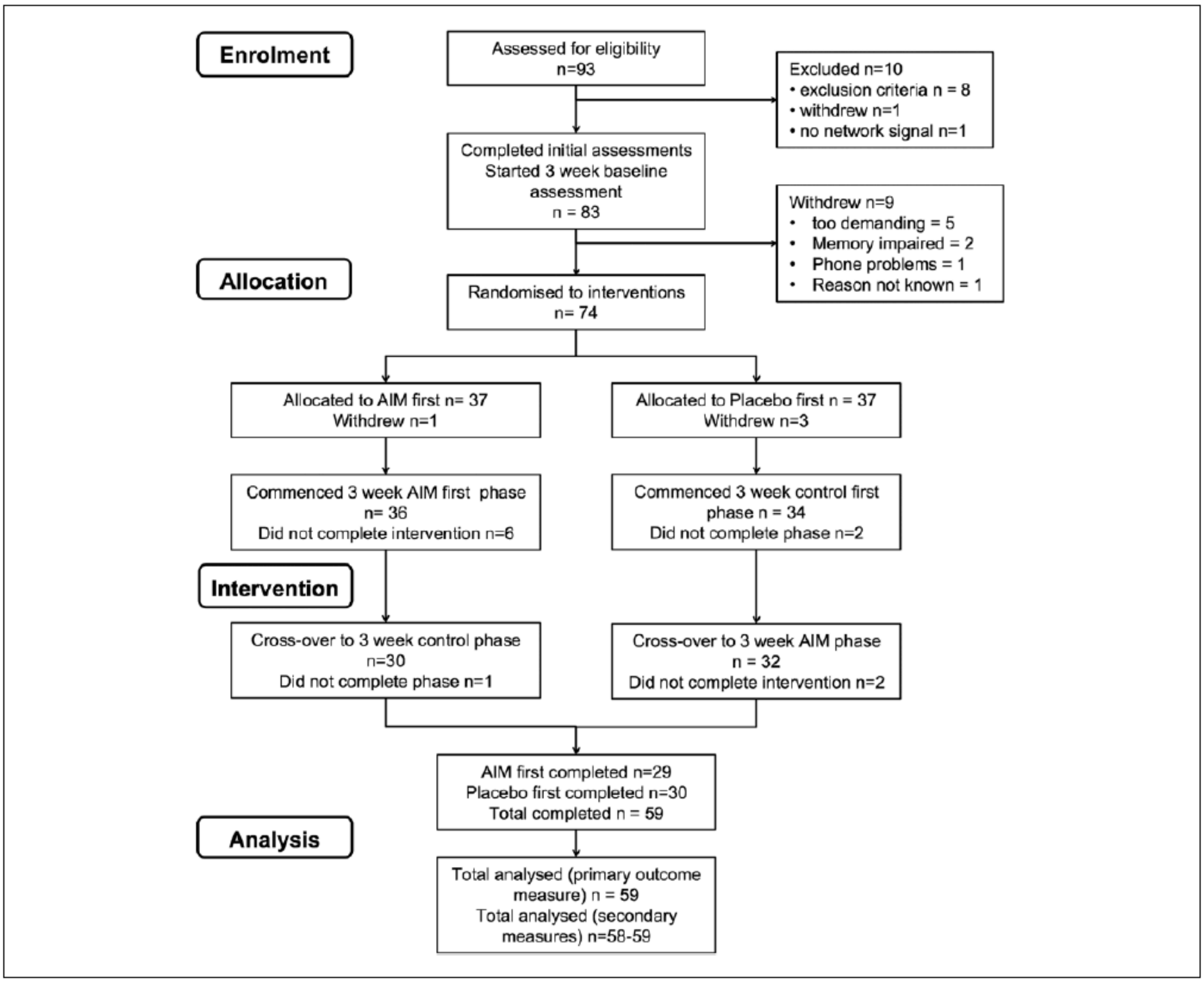

Figure 1. Trial flow chart showing numbers of participants referred, excluded, randomized to intervention, completed, and analyzed.
Abbreviation: AIM, assisted intention monitoring.

the AIM-first group in phase 2, therefore, allowed examination of whether their receipt was relevant to efficacy of goal management. This design also ensured that all participants had access to an intervention hypothesized to be useful, minimized the possible confounding effect of group differences on treatment effects, provided increased power to detect effects, and allowed examination of the maintenance of any gains in the AIM-first group.

A Steering Group comprising researchers, the local NHS Research and Development manager, and a person who had sustained a brain injury oversaw study management. The trial was conducted in accordance with National Institute for Health Research (NIHR) Good Clinical Practice in research guidelines, was adopted by the United Kingdom Clinical Research Network (UKCRN), and registered onto their research portfolio (Study ID: 5368).

Participants

Participants were recruited from UK community services in the East Anglia region, the Cambridge Cognitive Neuroscience Research Panel (CCNRP; a group of people with ABI who have agreed to be approached for relevant research studies) between February 2009 and August 2011. Health care professionals working with ABI patients were asked to provide potential participants with information about the study and seek their consent to be contacted by the research team. Members of the CCNRP were contacted directly by the researcher.

Inclusion criteria were as follows:

- age ≥18 years;
- nonprogressive brain injury, acquired in adulthood;

- more than 1 year postinjury;
- clinician, carer, or self-reported everyday organization and memory problems; and
- able to use a mobile phone.

Exclusion criteria were as follows:

- memory impairment of sufficient severity to limit retention of intentions and training information (clinical judgment and neuropsychological assessment);
- patient or carer participant with severe and enduring mental health problem, or substance misuse or dependence, as identified by referring clinician; and
- participation in a rehabilitation intervention with significant overlap with the study intervention.

Interventions

Interventions were delivered by a member of the research team (EG), a qualified occupational therapist with significant experience in providing cognitive rehabilitation interventions in both clinical and research settings with people with stroke and ABI. TM, a coauthor of the GMT materials, provided supervision.

Assisted Intention Monitoring. A brief GMT was provided by EG in participants' homes or a community setting on a one-to-one basis over 2 sessions, not more than 5 days apart, each lasting between 90 and 120 minutes. Training materials were selected from the full GMT program (as described by Levine and colleagues[5,6,15]) and presented on a laptop as a PowerPoint presentation with an accompanying workbook. The slides selected covered the following topics supported with discussion of examples drawn from the workbook or provided by the participant:

- utility of setting goals and breaking goals into steps (module 1)—for example, breaking a large goal or problem such as planning a trip away into doable steps;
- absentmindedness and slip-ups (module 2)—for example, walking into a room and forgetting what you went there for and discussion of factors that can increase slips, such as fatigue;
- using the "mental blackboard" to take note of goals and steps (module 5)—for example, rehearsing the mental visualization of written or pictorial checklist of steps on a "blackboard"; and
- checking the status of one's intentions (module 9), which was linked with the acronym "STOP"—for example, discussing how periodically stopping and thinking about our intentions can help us stay on track.

The training was provided to the point where the trainer was confident that the participant understood the material and the STOP acronym, so the training period varied depending on the knowledge and abilities of the participant. Participants were told that after training they would receive 8 "STOP" texts each day, designed to increase the frequency of such reviews. These occurred at random points between 08:00 and 18:00 hours on each working day. They did not occur within 30 minutes of each other or a set phone call time (see below). Messaging was provided via a reminding service[25] with the capacity to send SMS text messages.

Control Intervention. This involved one-to-one sessions (also provided by EG) of the same duration as AIM consisting of brain injury information[26] (excluding reference to executive functioning) presented using Powerpoint and a computerized visuospatial game involving increasingly speeded mental rotation (Tetris) plausibly linked to improving cognitive skills but not hypothesized to improve PM. Participants in the control phase also received 8 daily SMS text messages reading: "AIM research study. Please ignore."

Measures

Assessment and Screening Measures. Standardized neuropsychological assessments were completed and demographic and injury-related data collected. The National Adult Reading Test[27] was used to derive an estimate of premorbid general intelligence. The Speed and Capacity of Language Processing[28] was used to assess speed of processing. Nonverbal reasoning abilities were assessed with the Matrix Reasoning subtest of the Wechsler Adult Intelligence Scale, Third Edition.[29] Immediate and delayed verbal recall was assessed using the Logical Memory subtest of the Wechsler Memory Scales.[30] Executive functioning and attention were assessed using the Letter Fluency part of the Verbal Fluency subtest (Delis-Kaplan Executive Functioning System),[31] the SART,[32,33] and the multipart Hotel Test[18] (similar to the 6 elements[34]). The Coping Inventory for Stressful Situations,[35] which has been validated for use with ABI,[36,37] was included to identify possible moderators of treatment response.

Primary Outcome. The primary outcome was the mean daily proportion of intentions achieved by a participant averaged over the final 2 weeks of each 3-week study phase (consistent with previous studies,[23,25] data from the first week were excluded because of novelty effects). The primary outcome measure was a composite of participants' own, ongoing "set" intentions, established at initial assessment with the researcher and set for the study duration; participants' ad hoc intentions, one-off tasks that might arise during the course of the study; 7 fixed intentions set to ensure compliance with study procedures (eg, make sure mobile phone is with you, charged, and switched on); and the phone task[23] described below. With the exception of the phone task, participants recorded success or otherwise in a structured diary and relayed this information to the research team in a daily phone call initiated by the researcher (according to preference, this could be via less-frequent phone calls, no fewer than 3 per week, or via email). This was also used to determine if goals were irrelevant (eg, "remembering keys and wallet when going out" would be irrelevant on a day intentionally spent indoors).

At initial assessment, participants were asked to nominate 3 times of the day when it would be convenient to make a brief call to the study's answerphone. These had to be at least 30 minutes from a previous phone call and not set to coincide with a memorable time of day. Participants were asked to make their calls as close to the set time as possible over the 9 weeks of the study phases (ie, time-based PM) in addition to 1 further phone call at an unscheduled time each day (ie, step PM). Participants were simply asked to state their name on connection. Attainment and timing accuracy were scored from answerphone records. Scheduled calls made within 5 minutes (±) of the target time scored 6. This decreased by 1 for each additional 5-minute discrepancy down to 1 (±25 out) and 0 (call missed completely). Unscheduled calls gained 1 point if they were made at all, a further point if they were more than 30 minutes from another call, and a final point if they were made at a different time to the unscheduled call on previous days of the study. Not all calls were possible on all days because of phone malfunction, poor signal, or clash with important activity, and accordingly, the score was based on the proportion of the score achieved out of the total score attainable that day.

For each day, the total number of relevant intentions for each participant in each intention type (set, ad hoc, fixed, and phone calls) was summed and the daily proportion attained calculated. These values were then averaged across each 2-week assessment period.

Secondary Outcome. Given expectations that the phone call task would benefit from AIM, our second planned comparison considered attainment of all goals excluding the phone call task.

Subsidiary Measures. Subsidiary measures were administered after each baseline and intervention phase. The Profile of Mood States[38] total mood disturbance (POMS MD) score was used to evaluate the impact of AIM on overall emotional functioning. The Hotel Task and Verbal Fluency were used to evaluate effect of AIM on executive functioning in the absence of cues.

Randomization

The randomization procedure was administered by the academic department of one of the authors (JJE) at a site remote from the main research site. Blocked sequences (6 and 4, via http://www.randomization.com) enabled equal numbers of participants to be allocated to each group. Only 1 investigator (JJE) was able to access the sequence and allocation, which remained concealed until the researcher delivering the interventions (EG) requested the next participant allocation code, which was provided via email. Allocations were not revealed to any other member of the study team, clinical staff in recruitment sites, or participants.

Analysis and Sample Size Calculation

Hypotheses 1 and 2 were tested with crossover analyses conducted on the complete data set on a per protocol (PP) basis using repeated-measures ANOVA, the within-subject factor being study phase (postintervention 1 vs postintervention 2) and between-subject factor being group (control-first vs AIM-first), with baseline scores as a covariate. Significant Group × Phase interaction effects were taken as indicating relative efficacy of the AIM intervention. A power calculation for this design carried out using G Power,[39] with $\alpha = .05$, 80% power, 2 groups, and 1 covariate based on detection of a medium-large effect size (as previously found[24] and to identify potentially clinically meaningful response), indicated that a sample size between 52 ($f = 0.40$) and 67 ($f = 0.35$) would be required. We, therefore, sought to recruit 60 participants. The same analysis was conducted on Hotel and Verbal Fluency test data to explore the effect of AIM on executive functioning. Group comparisons postintervention phase 1 between AIM-first and control-first groups, on both intention to treat (ITT; including data from all participants analyzed according to their initial group assignment regardless of whether or not they withdrew) and PP (analyzing data only from participants who completed intervention in accordance with protocol) bases, were also conducted. Significant correlates of response to intervention ($P \leq .015$, α corrected for multiple comparisons) were identified for inclusion in a multiple regression.

Results

Participant Characteristics

Enrolment and allocation information is provided in Figure 1. Eligibility screening was carried out for 93 people, 74 proceeded to randomization, and 60 participants completed the study, with 58 participants completing the trial and all outcome measures; 1 further person completed only the daily intention diary, and another completed only the

POMS. In the PP group, cause of injury was predominantly traumatic brain injury (TBI; 27, 46%) or stroke (21, 35%). Severity of injury was obtainable for 15 (55%) TBI participants (severe: 11, 41%; moderate: 2, 7%; mild: 2, 7%). Notable differences (PP and ITT) were found in preinjury employment and time since injury and (ITT only) work hours (see Table 1).

Hypotheses 1 and 2: Crossover Analyses

Hypotheses 1 and 2 were tested with repeated-measures ANOVA to identify the presence of Group × Time interaction effects in favor of AIM, as planned. Mauchly's test of sphericity for equality of variances was not significant, and missing data were excluded. Figure 2 shows changes in performance for AIM-first and control-first groups across all phases, for all intentions, and also all intentions excluding the phone call task. For hypothesis 1, the repeated-measures ANOVA yielded a statistically significant Group × Time interaction [$F(1, 56) = 4.28$, $P = .04$; $f = 0.28$, medium effect size; 3.7% mean difference; 95% CI = 0.1%-7.4%]; participants achieved a greater proportion of intentions during the AIM intervention relative to control. For hypothesis 2, the ANOVA was repeated without the phone call data and again indicated greater goal attainment with AIM [$F(1, 55) = 4.79$; $P = .033$; $f = 0.28$, medium effect size; 3% mean difference; 95% CI = 0.3%-5.6%]. Analysis of phone task data replicated the previously reported advantage of cueing on this task [$F(1, 56) = 9.904$; $P = .003$; $f = 0.41$, large effect size; 7% mean difference; 95% CI = 2%-11.8%].

In terms of subsidiary analyses, no significant Group × Time interaction effect was found for the POMS MD score [$F(1, 55) = 0.091$; $P = .76$; $f = 0.04$, negligible effect] or measures of executive functioning [Hotel Test: $F(1, 52) = 0.080$; $P = .78$; $f = 0.03$, no effect; Verbal fluency: $F(1, 51) = 0.719$; $P = .4$; $f = 0.12$, small effect).

Group Differences Postintervention Phase 1

Data summarizing group differences postintervention phase 1 are provided in Table 2. For analysis, missing data were excluded, and Levene's test for equality of variances was not significant. No significant differences on all intentions were identified with ITT ($P = .87$; 1% mean difference; 95% CI = −9% to 11%) or PP analyses ($P = .688$; 1.4% mean difference; 95% CI = −5.6% to 8.8%; $d = 0.11$, negligible effect; 7% observed power). A significant difference in favor of AIM was found on the phone task with PP [$t(57) = 2.031$; $P = .047$; 9% mean difference; 95% CI = 0% to 18%; $d = 0.53$, medium effect size; 51% observed power] but not ITT analysis ($P = .43$; 5% mean difference; 95% CI = −8% to 18%).

Exploratory Analyses

To examine factors that may have influenced response to treatment, simple correlations between possible predictor variables (age, time since injury, avoidant coping style, POMS MD) and change (difference between AIM and Control for all intentions and the phone task) were evaluated. The only near-significant correlation (at corrected $P \leq .015$) was between POMS MD at baseline and change in achievement of all intentions ($r = 0.28$; $P = .032$); multiple regression was, therefore, not conducted. Differences between injury etiology groups' (TBI, n = 27; stroke, n = 21; other ABI, n = 11) response to intervention were explored with repeated-measures ANCOVA (Group × Injury type × Phase; covariates were baseline performance and time since injury). Significant interactions were detected between study phase, injury type, and group [$F(2, 51) = 5.62$; $P = .006$] for the phone task. Tukey's post hoc pairwise comparisons revealed significant differences between the TBI and "other ABI" groups (mean difference = 0.20; $P = .014$), with the TBI group showing the hypothesized response to intervention on the phone task, the stroke group appearing to drop with removal of AIM more than benefitting from AIM, and the other ABI group appearing to do worse with AIM. Given that a previous study found a drop in performance after removal of reminders for stroke, but not TBI, participants,[40] a 1-way ANOVA comparing the 3 injury type groups was conducted. No significant group differences in preintervention executive functioning were found [Hotel Task: $F(2, 54) = 0.169$, $P > .05$; Verbal Fluency: $F(2, 53) = 0.014$, $P > .05$].

Discussion

Interpretation

This study examined whether AIM intervention was associated with enhanced attainment of daily intentions for people with self- or clinician-reported everyday organizational problems and objective executive impairment following ABI. The results show that participants achieved their everyday intentions at a significantly higher frequency during the AIM phases of the study than the control conditions. The findings build on the body of work that shows that randomly occurring periodic cues to prompt "mental review" of intentions may contribute to improved performance on tasks requiring attentive control of goal-directed behavior.[18,19,23] The results suggest that any benefit of the training offered in AIM was only detectable when participants were receiving cues. Although this comparison has a confound of the extra time since training, it forms some indication that generalization from training is likely to be enhanced when participants are reminded about it in everyday life. There were no training effects on executive neuropsychological tests (during which

Table 1. Demographic Information and Neuropsychological Test Performance at Initial Assessment for Intention To Treat (ITT) and Per Protocol (PP) Groups.

	Intention to Treat		Per Protocol	
	Control First (n = 34)	AIM First (n = 36)	Control First (n = 30)	AIM First (n = 29)
Sex				
Male	23	23	21	21
Female	11	13	9	8
Etiology				
CVA	12	11	11	10
Infection	1	2	1	2
TBI	16	17	14	13
Tumor	4	6	4	4
Missing	1	0	0	0
Vocational situation				
Paid work	10	7	9	6
Retired	4	8	4	8
Voluntary	8	3	7	2
Unemployed	11	18	10	13
Missing	0	0	0	0
Work hours				
Full-time	7**	4	6	3
Part-time	11	4	9	3
Unemployed	16	28	15	23
Missing	0	0	0	0
Preinjury employment				
Professional	21**	12	19**	10
Elementary/Service	10	23	10	19
Unemployed	1	0	1	0
Missing	2	1	0	0
Mean age (SD), years	50.18 (12.76)	46.36 (14.88)	49.76 (12.94)	47.79 (14.72)
Mean years of education (SD)	12.47 (2.65)	12.69 (2.92)	12.43 (2.67)	12.79 (3.01)
Mean time since injury, years (SD)	8.62** (8.60)	4.89 (5.02)	9.15** (8.70)	5.00 (5.03)
D-KEFS letter fluency[a]	7.94 (3.65)	7.97 (4.01)	7.67 (3.58)	7.86 (4.02)
WMS-III LM I[a]	9.12 (3.44)	9.11 (3.56)	8.97 (3.61)	8.83 (3.52)
WMS-III LM II[a]	9.24 (3.57)	8.94 (3.87)	9.07 (3.63)	8.55 (3.71)
NART[a]	103.94 (14.42)	101.00 (12.89)	102.73 (14.83)	102.00 (11.55)
SCOLP speed of comprehension[a]	8.85 (3.54)	8.36 (3.25)	8.81 (3.67)	8.45 (3.29)
SCOLP spot the word[a]	10.82 (3.33)	9.88 (2.91)	10.63 (3.47)	10.03 (2.91)
WAIS-III matrix reasoning[a]	11.79 (3.03)	12.31 (3.25)	11.73 (2.97)	12.93 (2.96)

Abbreviations: AIM, assisted intention monitoring; D-KEFS, Delis-Kaplan Executive Functioning System; NART, National Adult Reading Test; SCOLP, Speed and Capacity of Language Processing; TBI, traumatic brain unjury; WAIS-III, Wechsler Adult Intelligence Scale, Third Edition; WMS-III LM, Wechsler Memory Scales 3rd Edition Logical Memory; CVA, cerebro-vascular accident.
[a]Mean of standardised score (standard deviation).
**Control first and AIM first groups significantly different at $P \leq 0.05$.
ITT (Intention to Treat) group differences: time since injury $t(67) = 2.1$; $P = 0.038$); previous employment (= 8.5; $P = 0.02$) and work hours (=7.3; $P = 0.03$).
PP (Per Protocol) group differences: time since injury $t(57) = 2.3$, $P = 0.025$); pre-injury employment (= 6.57, $P = 0.04$).

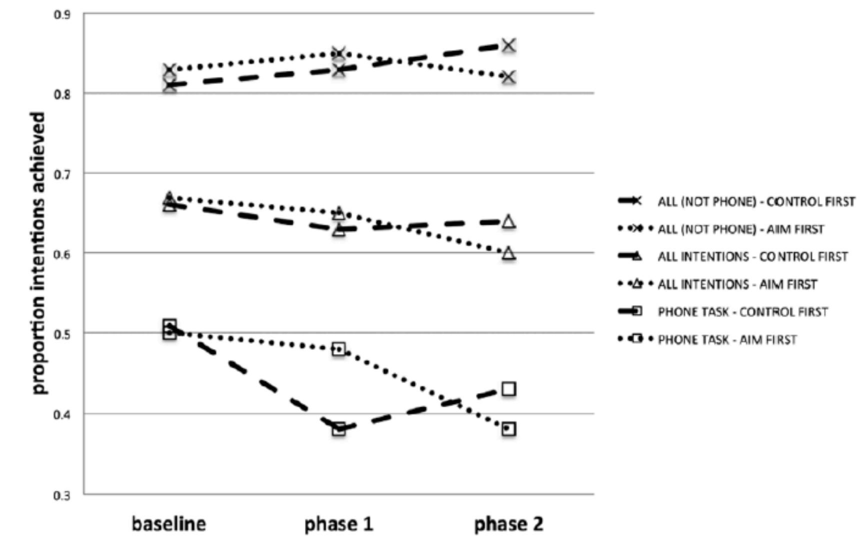

Figure 2. The proportion of intentions achieved for AIM-first and control-first groups at baseline, end of intervention phase 1, and end of intervention phase 2 for all intentions, all intentions minus phone task, and phone task.
Abbreviation: AIM, assisted intention monitoring.

Table 2. Comparison of Changes in Primary and Secondary Outcome Measures for Placebo First and AIM First Groups Between Baseline and Intervention Phase One on Intention to Treat and Per Protocol Basis.

Intention to Treat	Control First (n = 34), Mean (SD)	AIM First (n = 36), Mean (SD)	Mean Difference (95% CI), *P*
Primary outcome			
Overall intention attainment	0.63 (0.21)	0.64 (0.17)	0.01 (−0.09 to 0.11), *P* = .87
Missing values frequency (%)	3 (9%)	4 (11%)	
Secondary outcome			
Mean daily proportion of nonphone intentions achieved	0.83 (0.17)	0.85 (0.13)	0.05 (−0.06 to 0.10), *P* = .62
Missing values-frequency (%)	3 (9%)	4 (11%)	
Mean daily proportion phone score	0.42 (0.28)	0.47 (0.24)	0.05 (−0.08 to 0.18), *P* = .43
Missing values-frequency (%)	4 (12%)	4 (11%)	
POMS MD	47.3 (37.9)	47.2 (40.6)	−0.02 (−19.37 to 19.34), *P* = 1.00
Missing values-frequency (%)	2 (6%)	2 (6%)	
Per Protocol	**Control First (n = 30), Mean (SD)**	**AIM First (n = 29), Mean (SD)**	**Mean Difference (95% CI), *P* Value**
Primary outcome			
Overall intention attainment	0.63 (0.21)	0.65 (0.18)	0.014 (−0.056 to 0.084), *P* = .35
Missing values-frequency (%)	0	0	
Secondary outcome			
Mean daily proportion of nonphone intentions achieved[x]	0.83 (0.18)	0.85 (0.13)	−0.011 (−0.065 to 0.042), *P* = .68
Missing values-frequency (%)	1 (3%)	0	
Mean daily proportion of phone calls	0.38 (0.27)	0.48 (0.24)	3.38 (0.001 to 0.179), *P* = .047
Missing values-frequency (%)	0	0	
POMS MD	2.83 (20.3)	−0.55 (25.6)	3.38 (−8.78 to 15.54), *P* = .58
Missing values-frequency (%)	1 (3%)	0	

Abbreviations: AIM, assisted intention monitoring; POMS MD, Profile of Mood States total mood disturbance. *x* both groups *n*=29.

cues were not present), suggesting that treatment effects are a result of compensatory management of, rather than improvement in, executive difficulties. A recent trial[22] found that combined group GMT and reminders resulted in improvements to neuropsychological functioning sustained at the 6-month follow-up, suggesting potential benefits of increased intervention time. Fish et al[40] reported independent maintenance of routines after prolonged experience of timed specific reminders, which was evident for TBI participants but not those with stroke; this was attributed to better executive functions in the former group. In the current study, we did not find such group differences in executive functioning, although it is important to note the smaller group sizes, participant selection on the basis of poor organizational skills rather than memory, and the use of cues that occurred at random rather than fixed times each day. Further investigation of the treatment duration and intensity required for internalization of metacognitive or mnemonic cues over time is, thus, warranted.

Comparing groups postintervention phase 1, there was no evidence of significant benefit of the AIM intervention versus placebo on achievement of intentions or mood (ITT and PP analyses) or performance on the phone task (ITT analyses only), although PP analysis found a benefit of AIM for the phone task. At the most conservative level, this result indicates rejection of the study hypotheses. However, the study was not designed with this analysis in mind, and hence, these comparisons were underpowered to detect anything other than large effects. The PP analysis of the effect of training on the phone task at the end of phase 1 did yield favorable results, as did the adequately powered primary crossover analysis. We have, therefore, cautiously rejected the null hypothesis, bearing in mind the study limitations and, in particular, threats to the comparability of groups after crossover.

There were no significant effects of AIM on POMS mood disturbance scores, suggesting that a simple model of enhanced attainment of intentions leading to improved mood may be wrong.

Limitations

At 20%, drop-out rates were high, contributing to selection bias and limiting generalizability of results. It is likely that this attrition is attributable to aspects of the protocol (daily goal-attainment recording, daily phone calls, and long assessment sessions), not the intervention itself. The crossover design was justified to provide an opportunity for both groups to receive the AIM intervention, for the AIM-first group to have a meaningful control phase, for withdrawal of alerts to be monitored in 1 arm, and to provide increased power to detect effects of undergoing the intervention. However, this design combined data from the different control phases, compromising the comparability of arms after the point of crossover. Furthermore, it was not possible to examine efficacy of the intervention at follow-up.

Randomization produced groups that were well matched on primary and secondary outcome measures, neuropsychological functioning, or other demographic variables but that differed on time postinjury and employment. Although any effect is less problematic for the within-subjects crossover analysis, it may have influenced postintervention phase 1 analyses. Regarding precision of measurement, the evaluation of real-world impact of the intervention relied on participants' own ratings in contrast to the phone task, which provided an objective metric of attainment and, therefore, may have been a more sensitive measure. Although the study was appropriately powered for the analysis of the crossover data, the subsidiary and exploratory analyses should be interpreted with caution. Finally, a number of statistical analyses were used to address main and subsidiary hypotheses and exploratory analyses. To reduce the likelihood of false-positive results, we limited the number of analyses used to test the primary hypotheses and specified the directions of predicted relationships. The exploratory findings are reported as tentative.

Generalizability

The current study included elements of evaluation of effectiveness, such as referral on the basis of clinician, carer, or self-identified problems; intervention deliverable within health services; and evaluation of "real-world" outcomes. However, the delivery of intervention was not tailored to each individual on the basis of specific needs or ongoing response to intervention, and a placebo control condition was included, limiting clinical generalization. Many participants had difficulty with identifying and articulating intentions in precise terms, and results suggested differences in effects depending on etiology. Therefore, careful thought is needed in clinical application. The relatively brief 2-session GMT adopted here (in comparison with the 14 or more hours of face-to-face GMT training typically reported[16]) might be considered insufficient for many with ABI. Future evaluation of clinical effectiveness should consider a more extended and tailored period of strategy and self-regulation training[16,17,22] and inclusion in the intervention of additional components that enhance likelihood of transfer of strategies.[16,22,41-43]

Conclusions

The results of this trial show some support for the efficacy of combining a brief goal management intervention and cueing. Findings are consistent with previous proof-of-principle studies and have been extended to show some improvement in subjective reports of goal attainment in everyday life. However, when only the initial training period was considered and when ITT was taken into account, effect sizes were small or negligible and not supportive of the efficacy of AIM. The challenge of identifying intentions that are both easy to measure and meaningful to participants may have made detection of effects more difficult. Given the potential effectiveness of AIM, the costliness of neuropsychological rehabilitation interventions, and difficulty transferring skills from rehabilitation to everyday life, further investigation of periodic cues to enhance realization of intentions in everyday life following rehabilitation is warranted.

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Declaration of Conflicting Interests

The authors declared the following potential conflicts of interest with respect to the research, authorship, and/or publication of this article: The authors declare that there are no conflicts of interest. TM is a contributing author to Goal Management Training but receives no income from its commercialization. AB is the manager of the NeuroPage reminding service but receives no personal income from the service.

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5.2 Summary of key findings

The findings indicated a possible benefit of the combined brief goal management and text reminders, with the effect appearing stronger for the proxy task (making phone calls at set times) compared to participants' own everyday intentions. Tentative exploration of predictors of outcome also suggest that those with TBI rather than stroke or another ABI may be more likely to benefit. Although effects were only present in per-protocol analyses including data from the cross-over phase, it was concluded that provision of text reminders to prompt 'mental review' might aid carry-over from brief goal management training into everyday life. Despite the suggestion that 'cold' goal management skills might have a role in better emotional outcomes, this study did not find an effect of the rehabilitation intervention on emotional outcomes, although negative emotional state was associated with reduced response to intervention, as found in previous rehabilitation research.

CHAPTER 6

6 Discussion, critical review and theoretical integration

The four studies included in this thesis explored different aspects of ‘hot’ and ‘cold’ frontal functions in relation to one another and everyday life. As such, the studies allow reconsideration of the schematic presented in Chapter 1 (Figure 1). This model suggests an overarching structure for differentiating frontal functions, including ‘hot’ and ‘cold’ executive functions. In short, the studies suggest a role of ‘cold’ cognitive processes (working memory, planning, problem solving, goal maintenance) in social outcomes (peer relationships of children with ABI) and emotional outcomes (depression and anxiety, but not anger, in main effect and interaction with coping styles). However, ‘cold’ working memory functioning did not emerge as associated with a novel emotion-based decision-making task, where patterns of performance appeared more aligned to individual differences in approaching the task. In the randomised controlled trial (RCT) in Chapter 5 some evidence of improvement in everyday intentions was shown, in the absence of improved ‘cold’ EFs or emotional outcomes. The tested relationships are summarised in Figures 2a (significant associations found) and 2b (non-significant associations).

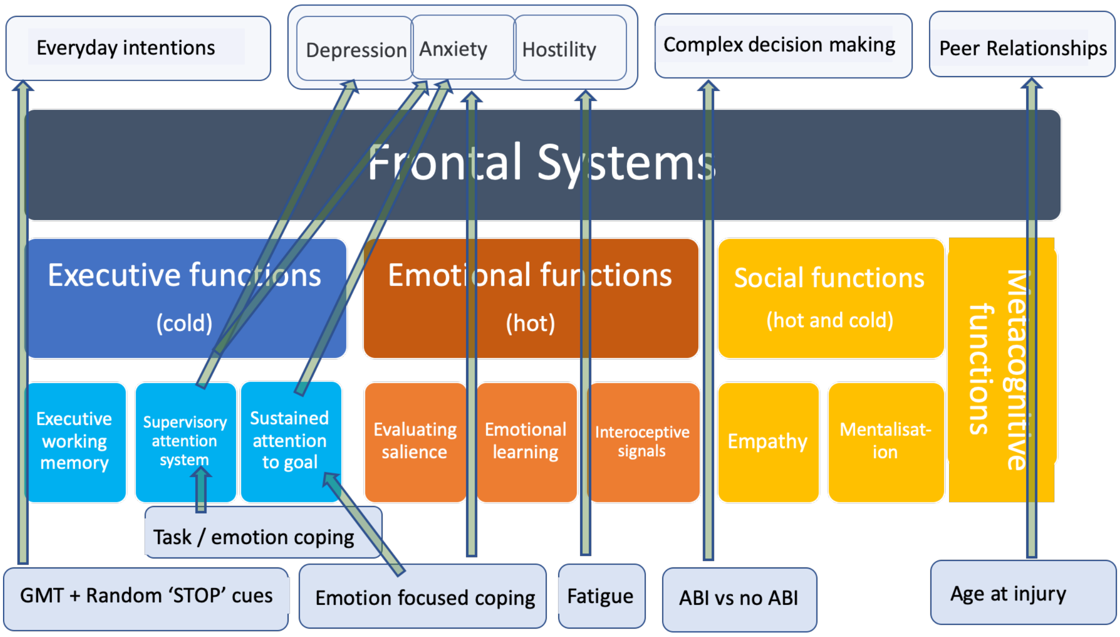

Figure 2a:

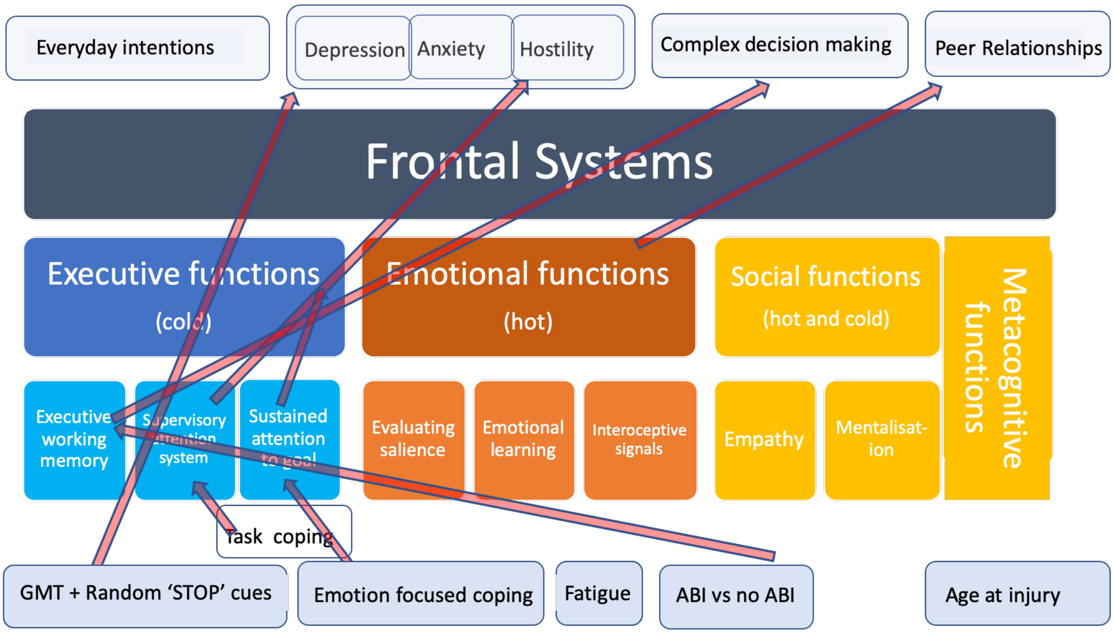

Figure 2b

FIGURES 2a and 2b: Key associations tested across the four studies showing significant associations (2a) and non-significant (2b). Arrow heads denote the dependent variable, not direction of causality.

With study methodological limitations in mind, these findings present a challenge to interpretation, as no clear single conclusion about the role of specific 'cold' processes in regulating 'hot' processes, or about the ways in which activation of 'hot' processes might impact upon cold cognitive processes, can be reached. As such, whilst we can conclude a potential interplay between 'cold' and 'hot' processes from the four studies, a simple hypothesis that inclusion of 'hot' processes improves models of the relationship between 'cold' frontal functions and everyday functioning cannot be fully supported. Relationships between 'hot' and 'cold' functions and everyday outcomes therefore likely vary according to the specific processes being measured and the conditions under which they are being elicited. For example, patterns of interaction between behaviour, and specific hot and cold frontal functions may well differ between decontextualized lab or clinic-based tasks and meaningful (therefore potentially 'hot') everyday situations.

In order to develop and integrate conclusions from the four studies, attention will now be paid to reviewing findings considering wider and more recent literature in relation to 'hot' and 'cold' functions across the conceptual areas covered in Chapter 1 (social processes and outcomes, emotional outcomes, participation and rehabilitation engagement).

6.1 Can specific neuropsychological 'hot' or 'cold' frontal functions be identified that are associated with social, emotional and participatory indicators of everyday functioning?

6.1.1 Social processes and social relationship outcomes

The two papers addressing this question are Gracey et al. (2014) which looked at peer relationships and EFs in childhood ABI, and Adlam, Adams, Turnbull, Yeates, & Gracey

(2017), which looked at cognition and emotion-based decision-making in adult TBI and non-injured controls.

Our study of peer relationships following child ABI seemed to support a role of 'cold' metacognitive executive processes, which on the BRIEF includes working memory. This is partially consistent with findings from two studies by Levan and colleagues (Levan et al., 2016; Levan, Baxter, Kirwan, Black, & Gale, 2015). These authors found a range of executive tests (letter fluency, trail making test) were not associated with social problems, but they did find an association between 'cognitive proficiency' (combined working memory and speed of processing abilities) and social competence (Levan et al., 2015), and also Continuous Performance Test errors and social problems (Levan et al., 2016). The variable relationships between social outcomes, social processes and specific 'cold' executive functioning tests might arise due to the heterogenous effects of brain injury or skills which are networked rather than focally localised. Focusing on the contribution of the 'social brain network' (SBN) and associated social processing skills (Theory of Mind, ToM), Ryan et al. (2016) found injury severity directly impacted SBN morphology. More severely injured children showed greater social difficulties at 24 months post injury on the Child Behavior Checklist (CBCL) compared with typically developing children, this being mediated by composite ToM scores at 6 months post-injury. As such, studies of children whose brain injuries involve both SBN and areas associated with 'cold' executive functions might give rise to correlations in performance that do not reflect actual functional interactions between these brain areas. Alternatively, ToM tasks often involve an element of 'cold' verbal working memory. Our study finding, mediation of age at injury and peer relationship outcomes by 'metacognitive' functions ('cold' EFs including working memory), could therefore be in part attributable to impact of working memory on 'cold' aspects of specific social abilities such as ToM. Studies focused on a wider range of predictors of outcome show that social outcomes can also vary greatly depending on family, parent and socioeconomic factors (Taylor et al., 1995; Wade et al., 2003; Wade et al., 2011), suggesting a more complex set of social determinants than a focal domain of either hot or cold cognitive functioning.

Our study of performance of people with TBI on the BGT was also concerned with 'cold' cognitive correlates of a 'hot' social processing task considered a potentially ecologically valid measure of social performance. Age, gender, estimated IQ, presence of TBI and, as found in other studies of social processes, speed of processing contributed significantly to 'gamble' choices. However, unlike the child brain injury and peer relationships study, and research into the contribution of working memory to IGT performance (Cui et al., 2015) we did not find associations between executive working memory and social (in this case BGT) functions. This would appear to confirm the reduced working memory demand of this simpler gamble task as intended by (Bowman & Turnbull, 2004). Although the TBI group appeared to perform worse overall than the non-injured control group, group differences in gender and age make it difficult to draw firm conclusions.

Bechara (1994) postulated that somatic markers guide allocation of resources in working memory. It is possible that the BGT reduces the working memory load on the task, leaving performance to be more strongly related to other processes such as implicit learning (Cui et al., 2015), sensation seeking or risk aversion. Icenogle et al.'s (2017) study explored the hypothesis that IGT performance of adolescents and young adults is associated with relative contributions of sensation seeking versus impulse control. They found that pubertal maturation predicted approach (via sensation seeking), whereas age predicted cost-avoidance (via impulse control). The reference to two interacting brain systems in the development of decision making under uncertain circumstances is helpful for considering patterns of performance in adults with and without brain injury. The data from our study illustrate what appears to be individual differences that could be made sense of in terms of sensation seeking (propensity to avoid gambling altogether or to generally opt to 'gamble' as in clusters 1 and 3 respectively) and balance between sensation seeking and inhibitory control (giving rise to learning over time, cluster 2). Performance on this type of task could therefore be determined by individual differences in: 1. Tendency to approach or avoid risk, 2. Inhibitory control, and 3. Integration of these systems. Sustaining a TBI could potentially impact brain systems associated with any of these processes individually or in combination.

An alternative explanation of the mechanism behind IGT performance related to task demands has been proposed by Steingroever, Wetzels, Horstmann, Neumann, & Wagenmakers (2013). They found substantial evidence that healthy participant performance is less based on 'learning' which decks are advantageous (pay out better) in the long term, and more on responding to the frequency of losses. They also note 'impaired' performance is commonly reported amongst healthy controls. The variation in healthy control performance was also identified within our BGT study, with about two thirds performing below the cut off proposed by Bowman & Turnbull (2004) for impaired performance.

Although the BGT appears to show a similar overall block-to-block pattern of performance as the IGT in our own and previous studies (Bowman & Turnbull, 2004) and might simplify certain potentially ‘cold cognitive’ aspects of the IGT, the task would not allow separation of scores on the basis of loss frequency as a determinant of choice over time. Therefore, the modified IGT and analysis employed by Icenogle et al (2017) might be preferable in future studies to further unpack neuroanatomical and cognitive correlates, particularly in a developmental clinical context. Performance seems to arise from a complex mix of individual tendency towards sensation seeking, inhibitory control, working memory (if required to allow encoding of trial outcomes, hold and decide between multiple options), and ability to integrate information across these systems to optimise outcome.

A further challenge with interpretation of results in this area is that whilst a specific domain of hot or cold functioning might appear to be associated with social relationship outcomes (when rated by others, as in our child ABI study), this does not preclude the influence of social contextual factors, not necessarily related to social skills. For example, as argued by Yeates et al. (2016) in relation to workplace abilities, judgements made by people about the social desirability of someone with ABI might stem from judgements of a range of abilities not just social skills, in addition to social stigma associated with disability (Jones et al., 2011).

The varied pattern of results across studies concerning relationship of hot (social) and cold (social and executive) functions, as measured on behavioural tests and ratings of social outcomes, may be indicative of the difficulties of evaluating simple predictions of association in the context of interactions between multiple component functions. Variability in findings concerned with prediction of everyday social behaviour could similarly arise from the interactions of multiple cognitive and social processes, sub-served by complex underlying brain systems.

6.1.2 Emotional outcomes

Paper 3 (Gracey et al, under review) explored correlates of emotional outcomes post ABI including coping style and executive functions independently and in interaction, finding some evidence for contribution of specific EFs to anxiety and depression but not anger and confirming the contribution of emotion and task-oriented coping. The general trend appeared to be that as EF reduces, the strength of relationship between coping and emotional outcome increases. The findings are at least partially consistent with predictions of processes proposed in the models of Hofmann et al. (2010), Ochsner & Gross (2005) and Salas, Gross and Turnbull (2019) and extend those of Krpan, Levine, Stuss, & Dawson (2007), in that presence of EF difficulties (in this case, with regulation of attention to goals, developing and implementing a plan) might contribute to emotional vulnerability via effects on coping.

It may be that people with reduced EF are less able to make use of adaptive coping strategies to reduce the impact of emotional responses or are more vulnerable to focusing on the emotional experience. However, a recent paper by Ownsworth et al.

(2018) proposed two routes by which emotional outcomes might be impacted by neurocognitive functioning: 1. Elevated emotional symptoms might arise from increased negative self-focused attention (rumination) where cognitive impairment impairs ability to regulate through reappraisal for example, or 2. Generally lowered cognitive resources might reduce tendency for negative self-focused attention leading to less emotional symptoms. Their findings appeared to indicate a protective effect of impaired memory, working memory and EF, consistent with the second pathway to emotional outcomes. Within a mixed group of participants with ABI, both processes might be represented resulting in null or contradictory findings depending on the profiles of participant clusters.

Paper 4 (Gracey et al, 2016) explored the potential impact of the AIM intervention on mood arising from 1. Feeling better about being more successful in achieving everyday intentions, or 2. Periodic interruption of ongoing routine behaviour to perform a 'mental review' disrupting unhelpful patterns of emotional response. However, we did not find an effect on emotional outcomes, unlike full GMT interventions (Stamenova & Levine, 2018). This could be accounted for by issues in study design - participants were selected based on executive difficulties in everyday life rather than depression. It may also be that depending on individual variation in mood or coping style, cues to reflect or stop and think might be disregarded (Richardson, McKay, & Ponsford, 2015), or, in keeping with Ownsworth et al. (2018) might cue negative self-focused attention, rather than disrupting it – as one client undergoing rehabilitation put it: "stopping and thinking is the last thing I want to do". As such, intermittent cues to self-monitor or skills to problem solve and manage goals might well be helpful but capacity to benefit will depend upon emotional reactions to everyday challenges and use of strategies, and the extent to which rehabilitation also addresses such processes.

Vagal tone (as measured by heart rate variability, HRV) also provides a possible basis for exploring co-variations in executive task performance and emotion regulation. Vagal regulation is thought to be triggered under situations of novelty, threat or challenge with potential impacts across cognitive, emotional and social processes (Porges, 2009; Thayer & Lane, 2000). It is therefore possible that apparent associations between executive loaded tasks and emotional outcomes are dependent on individual variation in vagal functioning, which would be linked to the affective aspect of certain executive tasks. Prime candidates for this effect would be tasks with time pressure or tasks where slips (e.g. go no-go tasks), negative feedback (e.g. card sort tests) or interference effects (e.g. Stroop task, Brown-Peterson task) are likely to be frequent. This suggests a possible further dimension along which relationships between hot and cold frontal functions might interact, in part accounting for issues in translation of assessment and rehabilitation from clinic to everyday life. We are currently undertaking work to investigate these processes.

6.1.3 Everyday goals and intentions, and transfer of rehabilitation to everyday life in adults with ABI

Investigations of cognitive rehabilitation targeting working memory through training packages and targeting higher-level metacognitive functions through goal management approaches (such as Goal Management Training, GMT), have seen an increase over the last 15 years, with the latter appearing to have the strongest emerging evidence base (Krasny-Pacini, Chevignard, & Evans, 2014; Stamenova & Levine, 2018). There is now some indication of effectiveness for specific interventions for deficits in self-awareness, also a major barrier in rehabilitation and in transfer from clinic to everyday life (Schmidt, Fleming, Ownsworth, & Lannin, 2013; Schmidt, Lannin, Fleming, & Ownsworth, 2011). Working memory emerges in several studies as a key determinant of social, emotional and practical everyday outcomes. However, efforts to improve everyday outcomes through working memory training have yielded at best mixed results, despite evidence for 'close transfer' of skills to tasks similar to those used in training across age and patient groups (Bahar-Fuchs, Clare, & Woods, 2013; Dunning, Holmes, & Gathercole, 2013; von Bastian & Oberauer, 2014). In contrast, Poulin, Korner-Bitensky, Dawson, & Bherer (2012) suggest there may be some potential for training-type approaches to executive rehabilitation post-stroke.

Approaches such as GMT typically include a mix of skills development, self-awareness, social support and compensatory strategy use. The meta-analysis of goal management training programmes conducted by Stamenova & Levine (2018) indicated beneficial effects of GMT across a range of outcome measures immediately after training and at follow up. Improvements in executive functioning tests, other cognitive processes, activities of daily living and mental health were found post training. Meta-regression indicated that number of hours of intervention was strongly positively associated with improvements in EF and in activities of daily living. Full GMT packages appeared to yield medium effect sizes for improvement in EF test performance, where we found no such effect. It is possible therefore that increased intervention hours lead to improvements in EF skills, which, with repeated practice of between-session homework could transfer into everyday life. Nevertheless, results from our trial, comprising only about 2 hours of intervention rather than 20, yielded a medium effect size for the proxy (phone call) task (compared to a strong general effect size for ADLs for full GMT in the meta-analysis). Whilst this supports the notion that number of hours of intervention is an important consideration, the fact that a medium effect size was yielded from our study suggests that nature and type of between-session cues might also be a significant factor in treatment and transfer effects. Inclusion of alerts might further facilitate this transfer without reliance on improvement in underlying executive skills. Whilst the meta-analysis showed largely positive effects across patient groups, in our study the effects of cues were strongest for people with TBI rather than stroke or other ABI, suggesting other factors relating to nature of underlying deficits might also be a factor in treatment response, for example emotional state or emotional reaction to cues, as discussed in the previous section. Alternatively, people post-stroke might be more likely to have focal lesions impacting upon everyday executive difficulties, compared with TBI where wider diffuse or network damage might be more likely. It may therefore be possible that the focus of the AIM intervention on maintenance of goals in working memory might not address underlying difficulties in core frontal functions.

Therefore, in terms of challenges to everyday participation such as work, education or social relationships, GMT in an adapted form, incorporating cues to ‘stop and think’ to help maintain activation of goals and plans in working memory, in addition to social or psychological supports to aid acceptance, might be most beneficial. Greater intervention intensity is also warranted, but further research is needed to explore how to extend findings of near-transfer from working memory training packages into everyday life.

6.1.4 Summary and consideration of findings in relation to the schematic model of frontal systems and processes

Several factors or processes can, therefore, be considered as influencing findings in studies attempting to relate 'hot' and 'cold' frontal systems to domains of everyday life and wellbeing:

- Differences between subgroups of participants based on aetiology of injury and age at injury and related neurocognitive differences
- Variation between individual participants in terms of:
    - Profiles of strengths and difficulties in hot and cold frontal functions (with potential for either positive or negative effects on emotion)
    - Enduring heuristics or disposition for guiding decisions in specific types of task (e.g. gambling / risk taking)
    - Biological predispositions to respond to situational demands including risk, conflict or novelty in particular ways
    - Subjective fatigue and related underlying brain metabolic processes
    - General intellectual functioning
  - The meaningfulness of cognitively demanding everyday situations to people with ABI in comparison to lab or clinic-based situations. Effectively, everyday life or ecologically valid tasks are more likely to implicate 'hot' systems

associated with processes such as risk/reward sensitivity, social connection, meaning and threat/challenge.

- Difficulty isolating and testing simple causal effects when there are multiple interacting processes across biological, psychological and social domains
- Issues of statistical power given the large number of variables that may need to be included in models

6.2 Synthesis of findings: A complex adaptive systems approach to the issue of ecological validity and far transfer between frontal systems and everyday life

The work presented and discussed in this thesis builds on our prior work on improving understanding of everyday social and emotional outcomes following brain injury (Brindley, Bateman, & Gracey, 2011; Gracey et al., 2008, 2009; Gracey, Longworth, et al., 2016; Gracey & Ownsworth, 2012; Longworth, Deakins, Rose, & Gracey, 2018; Yeates et al., 2008). Here we focused on the specific ways in which problems arising from disruption to frontal systems interact with everyday challenges and outcomes. The studies we conducted, and the literature here reviewed indicate likely interactions between 'hot' and 'cold' processes and metacognitive functions. Performance in everyday situations is likely influenced by subtle moment-to-moment shifts in meaning or emotional salience which can impact upon several distinct but interacting systems including physiological systems, component executive and social processes and metacognitive processes. Such contextual sensitivity is consistent with a view of frontal systems as having evolved to maximise our adaptability to changing or challenging contexts.

Whilst advances are being made in understanding the immediate or proximal cognitive and behavioural effects of underlying brain changes, if considered in isolation these advances may not help us understand, predict and address longer-term participation challenges in the everyday life of people post-ABI. Therefore, a further basis for issues of poor ecological validity in assessment of frontal functions, and also of poor transfer from rehabilitation clinic to everyday life, is that the features of these interrelated processes might be better conceived of as a complex adaptive system which is highly sensitive to context, rather than in terms of (multiple) linear causal relationships that can be understood outside of the context in which they are applied.

6.2.1 A frontal-contextual systems model

We propose that relationships between frontal systems dysfunction and everyday behaviour are best understood as arising from a set of nested complex adaptive systems each of which have the capacity to interact across system boundaries, as illustrated in Figure 2. A complex adaptive system is a system in which a connected network of components, governed by simple rules, act and react depending on the actions of neighbouring components. There is no component responsible for top down regulation or control, rather control emerges as a property of interactions across the system, and in response to the context in which the system sits. A complex system might itself sit within a wider complex system, such that it effects and adapts to that wider system. As such, it may be difficult to clearly delineate boundaries around complex systems which themselves are nested within wider systems (Health Foundation, 2010; Kim, 1999).

The model includes a social contextual system (located in the external, everyday world), a frontal system (located in the internal neuroanatomical realm), and a frontal-contextual system which emerges through interaction between multiple frontal component functions (including specific cognitive and psychophysiological processes), and social systems components (including the nature of task demands). Given the evidence that ‘cold’ cognitive systems can operate on a range of ‘hot’ or ‘cold’ content, these specific component processes have been relabelled as ‘neutral’. For simplicity, the developmental and contextual influences on cognitive content acquired by an individual, and lower-level cognitive processes are omitted. Many component processes could be included within the boundary of frontal systems. However, based on the work presented here, we have included attention orientation and control, working memory and SAS functions, particularly self-monitoring and schema generation and implementation (problem solving and flexible plan implementation). ‘Hot’ processes are labelled as such to denote the embodied subjective affective experience associated with these social and emotional processes. These include level of parasympathetic nervous system activation (specifically vagal tone), sensitivity to losses or rewards, and social processes such as empathy. Energisation, as described by Stuss is omitted for simplicity but would be included as a hot process acting across other systems in a similar manner to vagal tone.

Metacognitive functions are also included as to represent the specific processes involved when integration of component processes is required. The evidence we have reviewed appears to support a stand-alone metacognitive or integrative system associated with frontal polar or medial frontal networks. However, viewing frontal systems in everyday situations as a complex adaptive system implies that metacognitive functions could be understood as emergent system properties that arise under certain processing configurations, rather than discrete top-down control modules.

If considering assessment of a specific function, such as working memory, a particular frontal-contextual system configuration will arise in order to adapt to and address the task demands (as indicated by the upper green arrow) specific to that situation. Similarly, a decontextualized evaluation of a specific social or emotional task may give rise to specific hot or 'neutral' processing configurations (the lower red arrow). However, the demands of everyday life after brain injury are located within specific social and cultural contexts, where family functioning, peer relationships and social connection, attitudes about disability, social welfare and educational support systems, and the availability of information or discourses with which to deal with life after injury will all vary.

In the context of everyday life, prior to a specific situation or demand, an individual's cognitive executive skills and parasympathetic self-regulatory capacity might vary depending on developmental history, as well as any damage sustained and the age and developmental stage the damage was acquired at. The individual will carry with them expectations about performance, anticipated outcome (reward / risk), and about the perspectives of others, based on learning from prior experience, which will underpin appraisals made of performance and choice of coping strategy. In everyday life, novel or challenging situations will therefore necessarily comprise a blend of cognitive, social and emotional demands (the vertical green-red arrow on the right), dynamically interacting with component frontal functions (the vertical green-red arrow on the left). Component frontal functions themselves (such as maintenance of a goal in working memory) might dynamically interact and change as emotional and/or social processes are activated, or conscious, metacognitive regulatory strategies applied. Goals associated with, for example, immediate rewards or avoidance of anticipated negative experiences might compete in working memory with goals associated with longer-term gains. If able to reflect upon and communicate these dilemmas emotional activation might be reduced. Communication with others might further engage helpful supports. These changes could further impact task performance, which might in turn further trigger emotional responses. Over time a (perceived) tendency to ‘fail’ or ’succeed’ might influence decision making or behaviour in future similar scenarios. Equally, repeated contextualised application of strategies might enable a specific type of demand to become more routine exerting less demand on frontal systems. Individual differences in tendency to persevere, avoid losses or sensation-seek might further dynamically alter behaviour. Individual variability in HRV, reduced HRV secondary to frontal systems damage, or increased HRV related to positive psychological states and social connection could further contribute to these dynamic moment-to-moment interactions between processing demands and context. The pattern of behaviour that arises in a given novel, challenging or non-routine situation can therefore be made sense of as an emergent property of a frontal-contextual system, itself arising from the interplay between underpinning frontal brain systems and social systems, and therefore dynamically changing over time.

6.2.2 Implications for future research

If the interplay between component parts of the system conform to characteristics of a complex adaptive system (e.g. interdependence, sensitivity to feedback, lack of overarching organising component), research focusing on simpler causal relationships for predicting everyday outcomes may inadequately inform improvements in assessment and rehabilitation in patient groups with diffuse or system-wide damage as in ABI. A shift away from the study of simple linear relationships to the study of component interactions and system properties is required. Methodologies and analytic approaches for the study of complex adaptive systems are relatively well-developed in contemporary neuroscience research (for example Bullmore & Sporns, 2009). These approaches map the activity of low-level neuroanatomical nodes and their connections and provide a novel way of understanding functional connectivity and behaviour, but this has not yet significantly influenced clinical assessment or neuropsychological research concerning assessment and rehabilitation of everyday

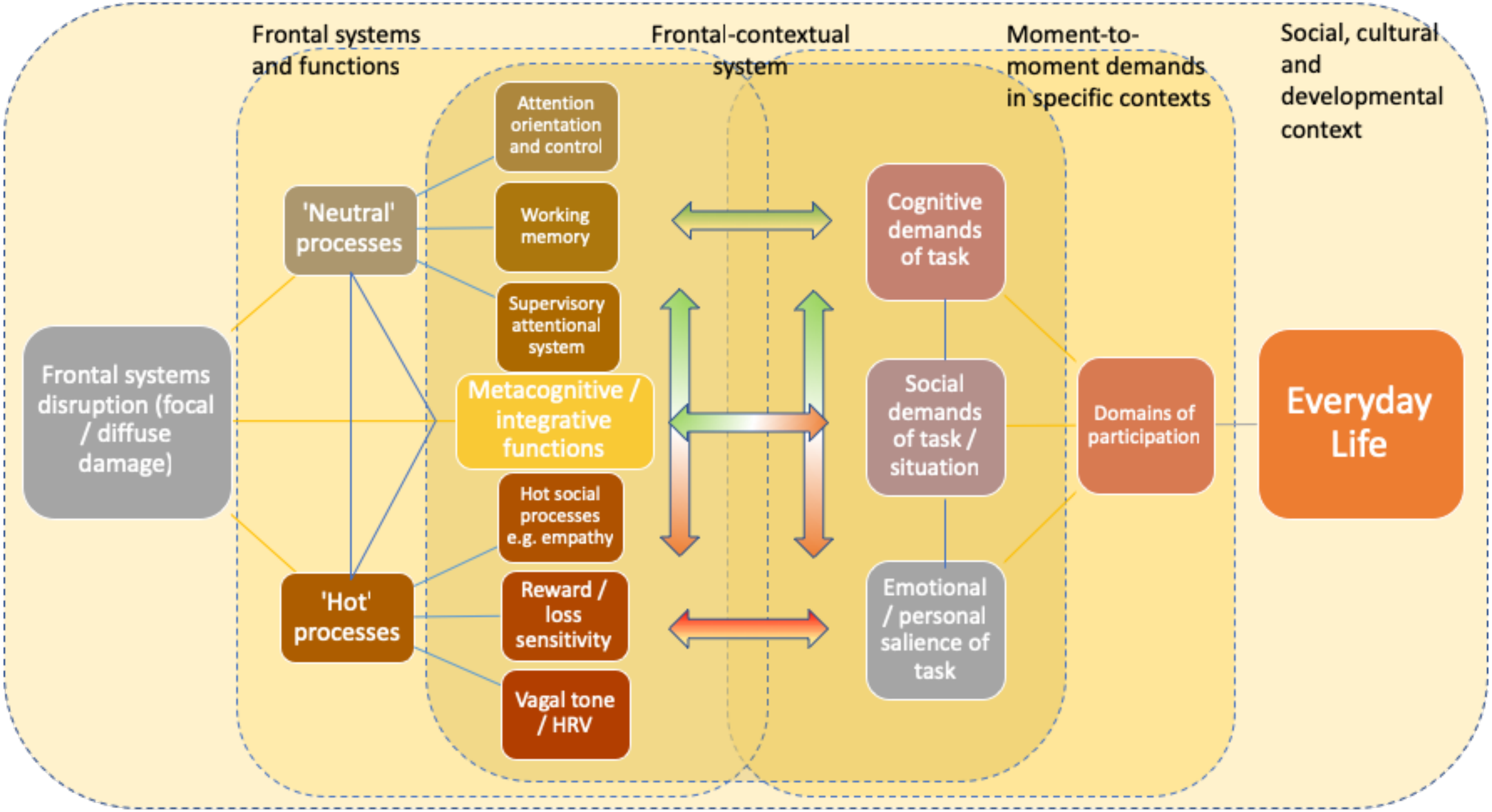

Figure 2: A framework for understanding frontal systems and everyday behaviour as a complex adaptive system: the frontal-contextual systems model.

difficulties. Understanding brain networks involved across multiple types of task, and identification of processes associated with highly interconnected 'hubs', might assist in identifying domains of assessment that are more likely implicated in everyday tasks with matching demands. Such an argument has been presented by Duncan (2010) in his review of the 'multiple demand network' of fronto-parietal control process and Zaki & Ochsner (2012) in their review of empathy research. Along similar lines, there is also a move towards understanding inter-individual variability in mapping cognitive processes to brain systems and behavioural tasks. Inter-individual variation is viewed as noise in traditional statistical techniques, however, approaches that seek to measure variability between individuals have been developed and show promise in accounting for brain-behaviour relationships (Kanai & Rees, 2011).

Future research could also attempt to map dynamic shifts in the interactions between component processes under changing cognitive, emotional or social task demands in such a way as to improve modelling of complex everyday scenarios. Time series classification approaches to neuroimaging data are increasingly used to understand brain activity associated with cognitive processes unfolding over time (Grootswagers, Wardle, & Carlson, 2017). These types of analyses may provide further opportunities for understanding the complex systems arising between everyday task demands and frontal systems. A novel approach that provides opportunities for understanding the interplay between brain and behaviour in everyday life is functional near-infrared spectroscopy (fNIRS), a highly portable and non-invasive approach which lends itself to applications outside the laboratory. Pinti et al. (2018) suggest that the next frontier for cognitive neuroscience is to move away from study of basic processes to the understanding of how people cope with everyday challenges. They suggest that, whilst big data approaches (such as the Human Connectome Project) might go some way to moving this endeavour forward using aggregated data, development of novel, portable techniques such as fNIRS breathe new life into traditional clinical neuropsychological approaches concerned with understanding a unique individual's processing ability under varying real-world demands. Pinti et al. (2018) summarise some recent applications to understanding patterns of prefrontal cortex activation and associated cognitive functioning in real-world experiments, providing some validation of models such as the SAS model. Increasing interest in wearable technology capable of gathering and integrating multiple markers of contextual sensitivity (such as portable EEG, ECG and skin conductance) might also support research along these lines.

6.2.3 Implications for interventions to support people with frontal systems deficits

Understanding the impact of frontal systems impairments on everyday life as occurring within a complex adaptive system has specific implications for intervention with people with ABI. A focus on targeted training of specific processes (such as adaptive training of working memory), even if successful in near transfer of gains to non-trained tasks, might not be sufficient to improve performance in everyday life, and may result in unintended consequences. For example, the 30 minutes per day of working memory training required to generate improvements in near-transfer tasks might take away from a child's time spent practicing maths skills or may increase stress in the family home with implications for behaviour and development.

Complex multicomponent interventions such as GMT incorporating interventions addressing component interactions would be more likely to result in gains in everyday behaviour as the evidence indicates. This approach involves repeated practice in monitoring attentional slips, thinking about and setting goals and plans, conducting periodic 'mental reviews' and managing factors such as fatigue and environment, and implementation of strategies in everyday life. Such approaches are sometimes dismissed in the context of healthcare intervention research as unsystematic 'shotgun' approaches. However, within a systems approach, the multiple components of the intervention would not be thought of as addressing individual components in a listwise manner. Rather, the co-occurrence of intervention components addressing interactions would be more likely to result in system adaptation, and behaviour change. For example provision of new information about how we are all vulnerable to attention slips and how we might manage these better, facilitation of self-monitoring in everyday life, non-judgemental attitude of facilitators to patient problems, mindful control of attention under stress, social connection between group members and supporting positive affect, might together facilitate psychological acceptance, engagement in behaviour change and adaptive skills practice, echoing the socio-cultural acquisition of skills through contextualised practice (Ardila, 2008; Vygotsky, 1978). Change in complex adaptive systems is achieved through changing the contextual conditions to facilitate the system in developing its own new self-regulatory processes rather than imposition of 'top-down' control. Therefore, interventions should also attend to the process of delivery and qualities of the therapist or social context in which rehabilitation is provided. In this vein, comprehensive day programme approaches (e.g. Wilson, Gracey, Malley, Bateman, &

Evans, 2009) also seek to present structured and supported opportunities to address real-world challenges that explicitly address experiences of threat to self in the context of a therapeutic milieu comprising staff, clients and family members involved in the programme. In support of the importance of contextual processes, quality of the therapeutic relationship in such programmes has been found to be associated with improvements in self-awareness (Schönberger, Humle, Zeeman, & Teasdale, 2006). A number of approaches to rehabilitation that incorporate multiple components to varying extents have been proposed, including context-sensitive behavioural supports and identity-oriented approaches (Ylvisaker, 2003), contextual-metacognitive therapy (Ownsworth, Fleming, Desbois, Strong, & Kuipers, 2006) and the Y-Shaped model of rehabilitation (Gracey et al., 2009) focused on contextualised integration of identity, strategy use and self-regulation. Diamond & Ling (2016) also provide a framework for consideration of self-regulatory interventions including combined physical and cognitive practise (as required in yoga or Tai Chi).

In addition to these multi-component interventions, the model presented also allows identification of specific processes that might facilitate gains through general effects across the system, such as energisation or enhancing parasympathetic adaptation to novelty or challenge by increasing HRV through stimulation (Clancy et al., 2014). Enhancing the vagal response to challenge or threat could be achieved through a variety of therapeutic techniques such as HRV biofeedback (Francis, Fisher, Rushby, & McDonald, 2016b; S. Kim et al., 2013; O'Neill & Findlay, 2014) or mindfulness-based approaches (Krygier et al., 2013) or other approaches associated with increasing wellbeing. Alteration of the regulatory potential of the whole system might enhance regulation of negative affect in the face of real-world challenges or demands, thus facilitating potential application of strategies developed in rehabilitation. Issues with the potential aversiveness of periodic reminders to ‘stop and think’, for example, might be reduced by enhancing an individual’s ability to accept and implement this strategy.

Although not addressed in the studies reported in this thesis, we are currently undertaking a single case experimental design trial for the feasibility and effects of transcutaneous vagal nerve stimulation (tVNS) on aggression in people with ABI, intellectual disability or autistic spectrum disorders. The trial is ongoing, but data from an initial case shows potential benefits, echoing findings of implanted vagal nerve stimulation in people with Prader-Willi syndrome (Manning et al., 2016). We are also about to undertake a study of the short-term effects of tVNS on executive (‘flanker’ task) and non-executive (lexical decision) cognitive task performance in healthy controls to further explore these mechanisms. Alongside this, we have recently researched an intervention that emphasise positive effects of social connection and creativity in an Arts and Health group intervention (Ellis-Hill et al., 2015), and are developing projects with the objective of further elucidating the interplay between affective, cognitive and social self-regulatory processes, social context and well-being following stroke and brain injury.

The next step in developing application of the frontal contextual systems model in clinical research would be to develop a programme to evaluate the additional benefits (in terms of improved carry-over into everyday life domains and reduced variability or unpredictability of responses) to routine rehabilitation (including training) that might be achieved through specific interventions that address ‘hot’ processes. Extending our prior research in this area, key interventions to evaluate include:

1. Enhancing rehabilitation of higher cognitive functions through interventions that improve capacity to manage the affective component of novelty, challenge or complex challenges in everyday life. For example, including mindfulness-based interventions, HRV biofeedback, or vagal nerve stimulation, and addressing self-efficacy in managing cognitive issues. Management of fatigue would also be necessary.
2. Identifying aspects of identity (self-goals or values, for example) associated with challenges in everyday life that might serve to increase the individual’s capacity to remain connected to pre-injury sense of self in the moment, thereby reducing negative affect / threat response, enabling a more adaptive approach to coping and increasing attention to potential future reward.
3. Identifying activities and social contexts that can enhance experience of safety and connectedness with self and others. Interventions here might include peer support, resource facilitation, arts and health interventions and community projects or groups.
4. Linking interventions in these domains to automated alert or reminding systems delivered via smart phone app to further enhance carry-over into everyday life.

Interventions would thus need to be chosen and combined per individual to address a balance of core skills that might need improving, along with interventions that address connection in social context, including family context and development in the case of childhood brain injury.

6.3 Conclusion

Traditionally, ecologically valid assessment of ‘executive functions’ and transfer of rehabilitation gains to everyday life has been challenging. By taking a broader approach to frontal systems functioning than solely focusing on ‘cold’ cognitive EFs, it has been possible to identify a range of ‘hot’ affective, and ‘hot’ and ‘neutral’ social and cognitive functions, that together and in interaction adapt to the demands of everyday situations. We have proposed a frontal-contextual system as a way of understanding the high level of contextual sensitivity of frontal systems, and view performance characteristics as emergent properties of the interplay between frontal brain systems and social contextual systems. It is concluded that methodologies developed for cognitive neurosciences research in to brain networks be extended to incorporate multiple data sources bridging brain and everyday behaviour. Implications for intervention support the application of complex multi-component interventions that create conditions under which the frontal-contextual system can adapt to maximise adaptive behaviour. Where emotional salience or meaning causes dynamic reduction in cognitive, social or emotional self-regulation, the potential to benefit from rehabilitation might be facilitated through approaches that maximise system adaptability. These might include interventions that aim to increase HRV (directly or indirectly), facilitate social relationships or connections, as well as providing opportunities to practice and develop skills. This shift towards social connection and wellbeing as intrinsic components of the rehabilitation of frontal dysfunction represents a radical departure from decontextualized cognitive training approaches and carries implications for rehabilitation that extend beyond healthcare into the wider community and social policy.

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APPENDICES

Appendix 1: List of Publications

Appendix 2: Statement regarding published material submitted in this thesis and nature of contribution

Appendix 3: Copies of correspondence from co-authors confirming contribution

Appendix 4: Correspondence relating to the publication of Paper 4

Appendix 1: List of publications (* submitted for PhD by Publication)

- ***Gracey, F.,** Fish, J. E., Wagner, A. P., Simblett, S. K., Bateman, A., Malley, D., Evans, J. J. & Manly, T. (submitted / under review). The influence of executive functioning on the relationship between coping style and emotional outcomes in the chronic phase following acquired brain injury. *Revisions requested, Neuropsychology, December 2018.*
- Simblett, S., Wagner, A., Watson, P., **Gracey, F**., Ring, H., & Bateman, A. (2017). A feasibility study piloting a randomised controlled trial of computerised Cognitive Behavioural Therapy to treat emotional distress after stroke. *Journal of Medical Internet Research* 4 (2), e16. doi:10.2196/mental.6022
- ***Gracey, F.,** Fish, J. E., Greenfield, E., Bateman, A., Malley, D., Hardy, G., ... Manly, T. (2016). A randomized controlled trial of Assisted Intention Monitoring (AIM) for the rehabilitation of executive impairments following acquired brain injury (ABI). *Neurorehabilitation and Neural Repair,* 31(4), 323-333. DOI: https://doi.org/10.1177/1545968316680484
- *Adlam, A.-L. R., Adams, M., Turnbull, O., Yeates, G., & **Gracey, F.** (2017). The Bangor Gambling Task: Characterising the Performance of Survivors of Traumatic Brain Injury. *Brain Impairment*, *18*(1), 62–73. doi:10.1017/BrImp.2016.30
- **Gracey, F.** (2016). The place of inner language in feeling: a commentary on "Revisiting the Left Convexity Hypothesis: Changes in the Mental Apparatus after Left Dorso-medial Prefrontal Damage" by Salas & Yuen. *Neuropsychoanalysis.* 10.1080/15294145.2016.1237307
- Longworth, C., Deakins, J., Rose, D., & **Gracey, F**. (2016). The nature of self-esteem and its relationship to anxiety and depression in adult acquired brain injury. *Neuropsychological Rehabilitation*, 1–17. doi:10.1080/09602011.2016.1226185
- Ford, C. E., Malley, D., Bateman, A., Clare, I. C., Wagner, A. P., & **Gracey, F.** (2016). Selection and visualisation of outcome measures for complex post-acute acquired brain injury rehabilitation interventions. *Neurorehabilitation*, *39*(1), 65–79. doi:10.3233/NRE-161339

- Cuberos-Urbano, G., Caracuel, A., Valls-Serrano, C., Garcia-Mochon, L., **Gracey, F**. & Verdejo-Garcia, A. (2016). A pilot investigation of the potential for incorporating lifelog technology into executive function rehabilitation for enhanced transfer of self-regulation skills to everyday life. *Neuropsychological Rehabilitation*. doi:10.1080/09602011.2016.1187630
- **Gracey, F**., Longworth, C. & Psaila, K. (2015) A provisional transdiagnostic cognitive behavioural model of post brain injury emotional adjustment. *Neuro-Disability and Psychotherapy.* 3(2), 154-185.
- Ellis-Hill, C., **Gracey, F**., Thomas, S., Lamont-Robinson, C., Thomas, P. W., Marques, E. M., ... & Jenkinson, D. F. (2015). 'HeART of Stroke (HoS)', a community-based Arts for Health group intervention to support self-confidence and psychological well-being following a stroke: protocol for a randomised controlled feasibility study. *BMJ Open*, *5*(8), e008888.
- Simblett, S, **Gracey, F**, Ring, H and Bateman, A. (2015) "Improving measurement of coping style following acquired brain injury using Rasch analysis." *British Journal of Clinical Psychology.* 54(3), 249-265. doi: 10.1111/bjc.12070.
- ***Gracey, F**, Watson, S, McHugh, M, Swan, A, Humphrey, A and Adlam, A. (2014) Age at injury, emotional problems and executive functioning in understanding disrupted social relationships following childhood acquired brain injury. *Social Care and Neurodisability* 5(3), 160-170.
- Malley, D., Wheatcroft, J. & **Gracey, F.** (2014) Fatigue after Acquired Brain Injury: a model to guide clinical management. *Advances in Clinical Neuroscience and Rehabilitation*, 14(2), 17-19.
- **Gracey, F.,** Malley, D., Wagner, A.P. and Clare, I.C.H. (2014) Characterising neuropsychological rehabilitation service users for service design. *Social Care and Neurodisability,* 5(1), pp 1-13.
- Ashworth F, **Gracey F** and Gilbert P. (2011) Compassion Focused Therapy After Traumatic Brain Injury: Theoretical Foundations and a Case Illustration. *Brain Impairment,* 12(2), pp 128-139.

- **Brindley R, Bateman A, Gracey F (2011)** Exploration of use of SenseCam to support autobiographical memory retrieval within a cognitive-behavioural therapeutic intervention following acquired brain injury. *Memory* 19(7):745-757**.**
- **Gracey F, Evans JJ, Malley D (2009),** Capturing process and outcome in complex rehabilitation interventions: A "Y-shaped" model. *Neuropsychological Rehabilitation.* 19(6):867-890.
- **Gracey F, Palmer S, Rous B, Psaila K, Shaw K, O'Dell J, Cope J, Mohamed S (2008),** "Feeling part of things": personal construction of self after brain injury. *Neuropsychological Rehabilitation.* 18(5-6):627-650.
- **Yeates GN, Gracey F, McGrath JC (2008),** A biopsychosocial deconstruction of "personality change" following acquired brain injury. *Neuropsychological Rehabilitation.* 18(5-6):566-589.
- **Gracey F, Ownsworth T (2008),** Editorial: Special issue on the self and identity in rehabilitation. *Neuropsychological Rehabilitation.* 18(5-6):522-526.
- **Dewar BK, Gracey F (2007),** "Am not was": cognitive-behavioural therapy for adjustment and identity change following herpes simplex encephalitis. *Neuropsychological Rehabilitation.* 17(4-5):602-620.
- **Yeates G, Henwood K, Gracey F, Evans J (2007),** Awareness of disability after acquired brain injury and the family context. *Neuropsychological Rehabilitation.* 17(2):151-173.
- **Gracey F, Oldham P, Kritzinger R (2007),** Finding out if "The 'me' will shut down": successful cognitive-behavioural therapy of seizure-related panic symptoms following subarachnoid haemorrhage: a single case report. *Neuropsychological Rehabilitation.* 17(1):106-119.
- **Dawkins N, Cloherty ME, Gracey F, Evans JJ (2006),** The factor structure of the Hospital Anxiety and Depression Scale in acquired brain injury. *Brain Injury.* 20(12):1235-1239.
- **Wilson BA, Berry E, Gracey F, Harrison C, Stow I, Macniven J, Weatherley J, Young AW (2005),** Egocentric disorientation following bilateral parietal lobe damage. *Cortex.* 41(4):547-454.

- **Macniven JA, Poz R, Bainbridge K, Gracey F, Wilson BA (2003),** Emotional adjustment following cognitive recovery from 'persistent vegetative state': psychological and personal perspectives. *Brain Injury.* 17(6):525-533.
- **Wilson BA, Gracey F, Bainbridge K (2001),** Cognitive recovery from "persistent vegetative state": psychological and personal perspectives. *Brain Injury.* 15(12):1083-1092.

Appendix 2: Statement regarding published material submitted in this thesis and nature of contribution

I confirm no part of the material presented in this PhD by Publication has previously been submitted by me for a degree in this or any other University.

The papers 1-4 presented in the thesis represent work undertaken by me in collaboration with co-authors. Confirmation of my contribution is provided by co-authors for each paper in Appendix 3.

Paper 1:

Gracey, F, Watson, S, McHugh, M, Swan, A, Humphrey, A and Adlam, A. (2014) “Age at injury, emotional problems and executive functioning in understanding disrupted social relationships following childhood acquired brain injury.” *Social Care and Neurodisability* 5(3), 160-170.

I established the system of routine in-service assessment and outcome data, developed the research question and methods, acquired relevant institutional approvals, supervised collation of the dataset, and descriptive analyses, conducted the multiple regression and collaborated with Anna Adlam and a statistician on the mediation analysis. I led on the write up and manuscript revisions.

Paper 2:

Adlam, A.-L. R., Adams, M., Turnbull, O., Yeates, G., & **Gracey, F.** (2017). The Bangor Gambling Task: Characterising the Performance of Survivors of Traumatic Brain Injury. *Brain Impairment*, *18*(1), 62–73. doi:10.1017/BrImp.2016.30

I devised the study in collaboration with G Yeates and in consultation with O Turnbull, and set up the in-service data collection, and healthy control data collection via the MRC-CBU participant panel. I contributed to data collection, and supervised others collecting data, oversaw collation of the dataset. The initial analysis was conducted by Anna Adlam under supervision of myself and M Adams, and this analysis and report was submitted by Anna Adlam as part fulfilment of a Clin Psy D at UEA. This paper was submitted for publication and rejected. I worked with Anna Adlam on the revisions, conducted new analysis (cluster analysis) and contributed to the revised manuscript. I then contributed significantly to revising the manuscript that was accepted.

Paper 3:

Gracey, F., Fish, J. E., Wagner, A. P., Simblett, S. K., Bateman, A., Malley, D., Evans, J. J. & Manly, T. (submitted / under review). The influence of executive functioning on the relationship between coping style and emotional outcomes in the chronic phase following acquired brain injury. *Accepted (with revisions) to Neuropsychology, November 2018*

I led on the writing of the grant application which funded the main study through which this data was collected and acted as chief investigator for the trial. In the development of the main study I proposed inclusion of measures of coping and the plan to analyse this data to explore interactions of EF, mood and coping. I wrote the paper, with co-authors providing suggestions and revisions. I conducted the analyses under supervision of A Wagner and led on addressing the requested revisions. The manuscript submitted here was accepted with revisions (see Appendix 4) in December 2018.

Paper 4:

Gracey, F., Fish, J. E., Greenfield, E., Bateman, A., Malley, D., Hardy, G., ... Manly, T. (2016). A randomized controlled trial of Assisted Intention Monitoring (AIM) for the rehabilitation of executive impairments following acquired brain injury (ABI). *Neurorehabilitation and Neural Repair,* 31(4), 323-333.

I led on the writing of the grant application which funded the trial and I acted as chief investigator for the trial, chairing research team meetings, reporting on progress and outcomes to funders, and co-supervising members of the research team with T Manly. I contributed to the analyses with T Manly and a statistician. I led on writing the paper, with co-authors providing suggestions and revisions, and led on addressing the requested revisions.

Appendix 3: Copies of correspondence from co-authors confirming contribution.

Paper 1:

Gracey, F, Watson, S, McHugh, M, Swan, A, Humphrey, A and Adlam, A. (2014) “Age at injury, emotional problems and executive functioning in understanding disrupted social relationships following childhood acquired brain injury.” *Social Care and Neurodisability* 5(3), 160-170.

Wednesday, December 19, 2018 at 3:03:31 PM Greenwich Mean Time

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Subject: Re: <no subject>
Date: Friday, 23 February 2018 at 05:29:10 Greenwich Mean Time
From: Adlam, Anna
To: Fergus Gracey (MED - Visitor), Suzanna WATSON, ah290@cam.ac.uk

Dear Fergus

Thank you for your email.

I am happy for this paper to be submitted in part completion of your PhD by publication and confirm that you made a significant contribution including: developing the original study idea, collating and analyzing data, and writing up the manuscript.

With best wishes, Anna

Dr Anna Adlam
Senior Lecturer/Clinical Psychologist
University of Exeter
01392 72 2209
http://cedar.exeter.ac.uk/staff/index.php?web_id=Anna_Adlam
Child and Adolescent Neuropsychology, Washington Singer Building, University of Exeter, Perry Road, Exeter, Devon, EX4 4QQ
Follow me on twitter: @CANexeter
Follow our Teen Online Problem Solving feasibility trial on twitter: @uk-tops
Get involved in our research: http://psychology.exeter.ac.uk/research/centres/ccnr/getinvolved/

Paper 2:

Adlam, A.-L. R., Adams, M., Turnbull, O., Yeates, G., & **Gracey, F.** (2017). The Bangor Gambling Task: Characterising the Performance of Survivors of Traumatic Brain Injury. *Brain Impairment*, *18*(1), 62–73. doi:10.1017/BrImp.2016.30

Wednesday, December 19, 2018 at 3:08:15 PM Greenwich Mean Time

Subject: Re: BGT paper question
Date: Friday, 23 February 2018 at 05:25:15 Greenwich Mean Time
From: Adlam, Anna
To: Fergus Gracey (MED - Visitor), Yeates Giles (Bucks Healthcare), o. turnbull

Dear Fergus,

Thank you for your email.

I confirm that you made a significant contribution to the paper in terms of: developing the original idea for the study, data collection, data analysis, and write up of the manuscript.

You also supervised my contribution to data collection and analysis which was completed as a Service Research Project in part completion of a Doctorate in Clinical Psychology (2006 - 2009).

With best wishes, Anna

Dr Anna Adlam
Senior Lecturer/Clinical Psychologist
University of Exeter
01392 72 2209
http://cedar.exeter.ac.uk/staff/index.php?web_id=Anna_Adlam
Child and Adolescent Neuropsychology, Washington Singer Building, University of Exeter, Perry Road, Exeter, Devon, EX4 4QQ
Follow me on twitter: @CANexeter
Follow our Teen Online Problem Solving feasibility trial on twitter: @uk-tops
Get involved in our research: http://psychology.exeter.ac.uk/research/centres/ccnr/getinvolved/

Wednesday, December 19, 2018 at 3:10:05 PM Greenwich Mean Time

Subject: RE: BGT paper question
Date: Friday, 23 February 2018 at 12:47:34 Greenwich Mean Time
From: Yeates Giles (Bucks Healthcare)
To: Fergus Gracey (MED - Visitor)

I also confirm you made a significant contribution to this paper

Giles

Dr Giles Yeates
Principal Clinical Neuropsychologist
Community Head Injury Service
Buckinghamshire Healthcare NHS Trust
Jansel Square
Aylesbury
Bucks. HP21 7ET.

Giles.Yeates@buckshealthcare.nhs.uk
tel: 01296 337760
fax: 01296 337743

Please note I work Mondays, Tuesdays, Thursdays & Fridays 9am-5pm and access emails intermittantly during these hours, unless otherwise stated. I do not work on Wednesdays.

I may not be able to respond to an email on the same day so for urgent matters do call the service on the number above.

Paper 3:

Gracey, F., Fish, J. E., Wagner, A. P., Simblett, S. K., Bateman, A., Malley, D., Evans, J. J. & Manly, T. (submitted / under review). The influence of executive functioning on the relationship between coping style and emotional outcomes in the chronic phase following acquired brain injury. *Accepted (with revisions) to Neuropsychology, November 2018*

Wednesday, December 19, 2018 at 4:25:26 PM Greenwich Mean Time

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Subject: Re: AIM coping paper - urgent request ...
Date: Wednesday, 19 December 2018 at 16:12:06 Greenwich Mean Time
From: Tom Manly
To: Fergus Gracey (MED - Staff)

To Whom It May Concern:

I am pleased to confirm that Fergus Gracey was centrally and principally involved in the design, conduct, analysis and write up of the work reported in "The influence of executive functioning on the relationship between coping style and emotional outcomes in the chronic phase following acquired brain injury" published in *Neuropsychology*

Tom Manly (Programme Leader University of Cambridge MRC Cognition and Brain Sciences Unit).

Wednesday, December 19, 2018 at 4:25:58 PM Greenwich Mean Time

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Subject: RE: AIM coping paper - urgent request ...
Date: Wednesday, 19 December 2018 at 15:40:44 Greenwich Mean Time
From: Simblett, Sara
To: Fergus Gracey (MED - Staff)

Hi Fergus,

Happy to confirm your involvement in the design, conduct, analysis and write up of the AIM coping paper. Looking forward to the final draft.

All the best with submitting your thesis,

Sara

Subject: Contribution to paper for publication
Date: Wednesday, 19 December 2018 at 15:31:48 Greenwich Mean Time
From: Jon Evans
To: Fergus Gracey (MED - Staff)

Dear Fergus
I am writing to confirm that you took the lead role in relation to the design, conduct, analysis and write up of the work entitled:

The influence of executive functioning on the relationship between coping style and emotional outcomes in the chronic phase following acquired brain injury

I understand that this paper is being submitted as part of your PhD by publication submission.

With kind regards
Jon

Professor Jonathan Evans PhD, FBPsS
Professor of Applied Neuropsychology

Tel: +44 (0) 141 211 0694
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Email: jonathan.evans@glasgow.ac.uk
Institute of Health and Wellbeing
College of Medical, Veterinary and Life Sciences

Mental Health and Wellbeing
University of Glasgow
The Academic Centre
Gartnavel Royal Hospital
1055 Great Western Road
Glasgow
G12 0XH
Calls from within Glasgow University 780 7000 33978
https://emea01.safelinks.protection.outlook.com/?
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518a070865e5d8f090%7C0%7C0%7C636808303102118434&sdata=jKXMs85Oyp0m%2B5cEensFCC9gtUVG
UQT3W8I5b7Q3590%3D&reserved=0
The University of Glasgow, charity number SC004401

Subject: Re: AIM coping paper - urgent request ...
Date: Wednesday, 19 December 2018 at 15:24:49 Greenwich Mean Time
From: BATEMAN, Andrew (CAMBRIDGESHIRE COMMUNITY SERVICES NHS TRUST)
To: Fergus Gracey (MED - Staff)

excellent news, of course I am happy to fully endorse and confirm from my point of view, you had the pivotal role in this work, at all stages

best wishes
Andrew

ps
Here is a film of a graduation ceremony at University of St Andrew's Jump to 1hr02minutes in for a solo by my son which I think will this year be my best way of sending seasonal good wishes to you!
https://youtu.be/-ufPfNXl-84

Andrew Bateman PhD MCSP
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Oliver Zangwill Centre for Neuropsychological Rehabilitation
Cambridgeshire Community Services NHS Trust
Princess of Wales Hospital
Ely CB6 1DN
www.ozc.nhs.uk
reception 01353652165
direct line 01353 652169
FAX 01353 652164

Subject: Re: AIM coping paper - urgent request ...
Date: Wednesday, 19 December 2018 at 16:32:30 Greenwich Mean Time
From: Fish, Jessica
To: Tom Manly
CC: Fergus Gracey (MED - Staff), Jon Evans, Andrew BATEMAN, Adam Wagner (MED - Staff), Simblett, Sara

Hi Fergus,

Yes, I'm happy for this email to serve as confirmation of your primary involvement in the design, conduct, analysis and write up of the work. Or to sign anything else as necessary!

Best wishes and merry Christmas!

Jess

Sent from my iPhone

Paper 4:

Gracey, F., Fish, J. E., Greenfield, E., Bateman, A., Malley, D., Hardy, G., ... Manly, T. (2016). A randomized controlled trial of Assisted Intention Monitoring (AIM) for the rehabilitation of executive impairments following acquired brain injury (ABI). *Neurorehabilitation and Neural Repair*, 31(4), 323-333.

Wednesday, December 19, 2018 at 3:10:42 PM Greenwich Mean Time

Subject: Re: AIM study paper question
Date: Friday, 23 February 2018 at 13:57:56 Greenwich Mean Time
From: Tom Manly
To: Fergus Gracey (MED - Visitor), Jon Evans, Jessica FISH
CC: Andrew BATEMAN

Hi Fergus

I am very happy with it.

I hope by this email I can confirm that you were centrally involved in all stages of the project from initial design, management, analysis and write up.

Thanks

Tom

Wednesday, December 19, 2018 at 3:06:00 PM Greenwich Mean Time

Subject: Re: AIM study paper question
Date: Friday, 23 February 2018 at 09:31:49 Greenwich Mean Time
From: FISH, Jessica (CAMBRIDGESHIRE COMMUNITY SERVICES NHS TRUST)
To: Fergus Gracey (MED - Visitor)

Hi Fergus,

That sounds like a great plan. Will the following do?

I am writing to confirm that Dr Fergus Gracey made major contributions to all components of the AIM trial as reported in the paper on which he is first-named author.

Best wishes,
Jess

Dr Jessica Fish
Clinical Psychologist
Oliver Zangwill Centre for Neuropsychological Rehabilitation, Princess of Wales Hospital, Lynn Road, Ely, Cambridgeshire CB6 1DN
Tel: +44 1353 652165 (reception), +44 1353 656682 (direct) Email: mailto:jessicafish@nhs.net

Subject: RE: AIM study paper question
Date: Friday, 23 February 2018 at 08:41:13 Greenwich Mean Time
From: Jon Evans
To: Fergus Gracey (MED - Visitor)

Dear Fergus
I have no objection to you submitting the AIM trial paper as part of your PhD portfolio. As lead author and PI on the study it is clear that you made a major contribution to planning, running and reporting the study.
Best wishes
Jon

Appendix 4: Correspondence relating to the publication of Paper 4

Subject: Your Submission NEU-2018-2579 - [EMID:d48e32c76a6349d6]
Date: Sunday, 18 November 2018 at 18:50:31 Greenwich Mean Time
From: em.nps.17228.5f57e4.6b16d050@editorialmanager.com on behalf of Gregory G. Brown
To: Fergus Gracey (MED - Staff)

CC: gbrown@ucsd.edu

NEU-2018-2579
The influence of executive functioning on the relationship between coping style and emotional outcomes in the chronic phase following acquired brain injury
Neuropsychology

Dear Dr Gracey,

Thank you very much for submitting your manuscript "The influence of executive functioning on the relationship between coping style and emotional outcomes in the chronic phase following acquired brain injury" for review and consideration for publication in *Neuropsychology*. I sincerely appreciate the opportunity to review the manuscript. I have now received the reviews of your manuscript and am able to make an editorial decision at this time.

Reviewers have now commented on your paper. You will see that they are advising that you revise your manuscript. For your guidance, reviewers' comments are appended below. Please note that some very specific recommendations have been outlined that will need to be addressed, including issues with how the results are presented and interpreted.

If you decide to revise the work, please submit a list of changes or a rebuttal against each point which is being raised when you submit the revised manuscript.

To submit a revision, go to https://nps.editorialmanager.com/ and log in as an Author. You will see a menu item called Submission Needing Revision. You will find your submission record there.

Sincerely,
Erin D Bigler, Ph.D.
Associate Editor
Neuropsychology

Reviewers' comments:

Reviewer #1: Overall, I find this an interesting and clinically relevant topic, the article is well written and the statistical methods are scholarly applied and interpreted. However, I have some serious comments that need to be addressed.
The introduction could be more concise, some details can be left out or moved to the discussion. At the same time the research questions are too global. The authors propose that executive functioning (EF) deficits may, in interaction with coping styles, influence emotional outcomes of patients with brain injury. Although this does make some sense, this hypothesis is not clearly worked out and lacks specificity. The authors introduce three general constructs (EF, coping style, emotional outcome) which are all rather broad. Therefore, it does not suffice to just provide general predictions about their mutual relations. As far as I know, interactions of deficient problem solving and an avoiding coping style with anger may be very different from interactions of planning deficits and a passive coping style with depression. Just investigating all possible interactions and relations without clear assumptions may create the impression of fishing for significance. It gets even more complicated when suddenly in the method and results sections other constructs turn up such as fatigue, without (again) a clear rationale why this might be relevant. In addition the two aspects of EF that the authors included seem to be chosen somewhat haphazardly; the necessity of choice for exactly these tasks is not clear but should be provided. The paper would benefit from a reduction in constructs (two copingstyles (active vs passive), two types of emotional outcomes (depression vs anxiety) with clarity about the expected interactions.
Specific comments:

Introduction:
- Prediction one states that a. coping styles will be associated with emotional distress b. EF will contribute to variation in emotional distress. I would expect these questions to be addressed by means of with correlational analyses showing the strength of associations between the different constructs, but these were not provided. I strongly recommend to do so.
Method:
- Why was having EF impairments (determined with tests) not an inclusion criterion given their central importance in the research question?
- The description of the participants is not very informative. In particular relevant details are lacking regarding the injury characteristics of the patients: TBI: how was moderate-severity determined, were GCS scores or MRI findings available? Stroke: what types of stroke? Severity, laterality, minor strokes? ABI: what types of ABI exactly?
- The range of time since injury seems very broad, with subacute to very chronic patients. Could this have been of influence on the results? I suspect that emotional outcome will vary over time (there is some evidence for this), not allowing comparison of patients in different stages of recovery.
- For age no range is provided but I find this very relevant. Please do so.
- Table 1 shows clinical characteristics of the sample. The test results in this table are not informative. It would be relevant to know whether the patients were on average impaired on the D-KEFS letterfluency task, a measure of EF. Why was this measure not used to select patients on EF deficiencies, as it was available. Why was this measure not included in the analyses since previous studies (both Spitz, 2013, Krpan, 2007) had shown its relevance?
Results:
Scores of the patients on the different EF tests, as well as on the different questionnaires are lacking. Where these in the impaired range? Were there any differences between the subgroups of patients? Please provide these results.
Correlations between the EF tests with the emotional outcomes and of Coping styles with the emotional outcomes are also lacking. I recommend to provide these data as well.
Discussion:
Discussion of the findings should be done in the light of the previously set hypothesis. Fatigue seems to be a relevant construct but there was no mention of its possible relevance in the introduction.

Reviewer #2: Review of The influence of executive functioning on the relationship between coping style and emotional outcomes in the chronic phase following acquired brain injury

NEU-2018-2579

Introduction

The authors present a causal model for the relationships between EF and emotional status. The study only presents what are essentially correlations between these.

The sentence "Progress is being made..." is too broken by reference citations. This could be rewritten to minimize this.

What are "transdiagnostic models"?

The abilities underlying EF in relationship to coping and psychological adjustment are not defined. How would findings measured by a typical Stroop task relate to problem solving, or any other construct that predicts psychological and emotional adjustment? The Introduction itemizes study descriptions without presenting a model explaining why EF should be related to adjustment and mood disorder.

The authors make a good observation concerning the overlap of self-report measures. However, it may be that self-report measures directly asses psychological coping where a Stroop test measures nothing related to this. It is hard to argue that tests such as Trailmaking measure anything like the constructs involved in psychological problem solving related to depression. There is even an argument to be made that high levels of EF impairment are related to psychological indifference and concrete reasoning (Goldstein, 1952).

Methods

The sample size is too small for regression studies.

The inclusion criterion beginning "clinician, carer ... " is not clear. Does this refer to a brain-injured subject?

The role of memory disorder in the inclusion and exclusion criteria is unclear. How memory impaired were the subjects?

The degree and type of mental health problem in the subject and their caregivers is unclear. Were subjects chosen because caregivers had mental health problems or substance use disorder? What was the mental health status of the brain injured subjects?

Results & Discussion

Tables are not APA style. Authors need to get Presenting Your Findings by APA.

Table 1 does not preset the means and SDs for the major study variables. What were the means for the SART, Hotel Test and POMS major scores?

Table 2 is an incomprehensible mass of numbers. What is meant by the grayscale sections in Table 2? What was the criterion for making some of the numbers bold type?

The figures are unecessary. The grayscale is poor form. The marginal titles use variable names from the program used to make the graphs.

The analyses represent statistical overkill for what is essentially a simple correlation study. What were the simple correlations between the two EF tests and the psychological outcome measures?

There is a suggestion based on the Matrix Reasoning Test that this is a very high functioning sample of patients with ABI. Since the EF tests means were not presented, it is impossible to estimate this. If true, it suggests that variance in EF and adjustment measures was constrained. This makes for small correlations and can explain the negative findings on the Aggression Scale of the POMS.

The major problems with this study involves the conceptualization of EF and psychological adjustment. How much will arcane measures of sustained attention or simple reasoning ever predict psychological adjustment? Would it not be better to examine a specific measure of EF in the domain of psychological adjustment rather than try to predict it from something like the Hotel or SART tests? A good place to start might be the measures of adjustment used by the practitioners of problem oriented therapy (e.g. D'zurilla & Nezu, Problem Solving Therapy, 2007). Numerous theories of EF suggest that each cognitive ability that needs to be controlled has its own EF control network and psychological/emotional adjustment EF is independent from the system controlling functions such as sustained attention. The self report measures suggest that this specificity prevails and each system of EF control requires its own method of assessment.
