Assessing and Addressing Unmet Needs in People Affected by Brain Tumours

by

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Declaration

I declare that this work is submitted in partial fulfilment of the requirements for the degree of Doctor of Philosophy.
I confirm that the work has not been submitted for any other degree or professional qualification.

I confirm that this thesis is the result of my own independent work under the supervision of my director of studies and supervisors.

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Abstract

Introduction

Guidelines recommend that brain tumour patients have their unmet needs assessed by a Holistic Needs Assessment (HNA), followed by appropriate support and care to maximise self-management. The aim of this study was to determine the role of HNA amongst brain tumour patients and their families.

Methods

A sequential mixed methods approach with three distinct phases was used. The first phase undertook a systematic review of available tools for HNA in brain tumours to determine if there was a psychometrically valid tool. This was followed by qualitative interviews and focus groups that explored patients' and carers' experiences and perceptions of unmet needs as well as strategies and support to assess and address these. The next phase was a realist synthesis of the HNA programmes in brain tumour patients which explored how HNA programmes could work. The thesis was underpinned by realist methods which were used to draw each of the phases of the thesis together.

Results

It was determined that there was no psychometrically valid tool that could be recommended for HNA in this group. Furthermore, there are significant challenges in using a patient completed HNA due to cognitive difficulties. Patients with brain tumours have diverse needs, but also unique compared to most other cancers due to the neurocognitive impairments. These also impact the carers as neurocognitive changes can be distressing but also cause a higher carer burden, therefore carers need specific support in their role. While the HNA as a process is useful in addressing unmet needs and enabling more self-management; the existing programme for HNA in UK is not suitable.

Conclusions

This thesis supported that a modified HNA programme may be beneficial in meeting the needs of brain tumour patients. Due to neuro-cognitive impairments, an assessment of the patient's ability to self-manage, combined with inclusion and support of carers in HNA programmes for brain tumour patients, is needed

to maximise the potential for self-management. The assessment and management of distress as well as increased access to the neuro-oncology team is also recommended. This research has proposed an alternative, novel practice-based model integrating the findings of this research.

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Abbreviation key

CNS – central nervous system

GBM – Glioblastoma Multiforme

HCP – health care professional

HNA - Holistic Needs Assessment

HRQOL – health related quality of life

MDASI - MD Anderson Symptom Inventory – Brain Tumor Module

NFbrSI-24 - National Comprehensive Cancer Network/Functional Assessment of Cancer Therapy-Brain Symptom Index

NHS – National Health Service

PCI – Patient Concern Inventory

PBT – primary brain tumour

QOL – quality of life

RCT – randomised controlled trial

SCNS34-BS - Supportive Care Needs Survey 34 plus brain subscale

UK – United Kingdom

1 Chapter One - Introduction

1.1 Introduction

A diagnosis of brain tumour affects nearly 10,000 adults a year in the United Kingdom and has a devastating impact on individuals and their significant others (Cancer Research UK, 2015b). Gliomas are the most common type of primary intracerebral cancer and constitute approximately 75% of cases (Taphoorn et al., 2010). Of these > 60% are high-grade glioblastoma multiform (GBM) that has a median survival of 12 months (Hayat, 2011; National Institute for Health and Care Excellence, 2018). Although 40-50% of brain cancers occur in the > 60-years old age range, compared to most other cancers the incidence is more widely distributed across all age groups. There is a significant impact on younger adults due to the poor survival rate and the age at which brain cancers occur (Cancer Research UK, 2015a). Consequently, brain cancer is the most common cause of death from solid cancers in the 16-24 years old age range and the second most common cancer death in adults in the 25-49 years old range in the United Kingdom (Cancer Research UK, 2015a).

Brain tumour differs significantly from other cancers due to the unique neurocognitive impact and higher symptom burden (Ford et al., 2012). Upon diagnosis, patients must manage the implications of a life-limiting illness whilst dealing with the symptoms, which can be severe and progressive (Cavers et al., 2012). Approximately 50% of patients present with headaches and other common symptoms include confusion, hemiparesis, gait imbalance, language difficulties personality changes, mood disturbance (for example, anxiety, fatigue, depression), decreases in mental capacity and problems with concentration (Butowski & Chang, 2007; Catt et al., 2008; Janda et al., 2008; Omuro & DeAngelis, 2013). Seizures are common, with rates of up to 60-75% for individuals diagnosed with low-grade gliomas and 25-60% for those with high-grade gliomas. This may have an effect on employment, social interactions and independence (Englot et al., 2016). Patients have self-reported behavioural changes such as disinhibition (19%), anger (27%), inappropriate behaviour (27%)

or apathy (41%) (Simpson et al., 2015). This presents significant challenges for the patients and their carers.

Treatment options in brain cancer can cause multiple local and general side effects. Patients undergoing brain radiotherapy will typically have a number of side effects including hair loss, nausea and fatigue as well as cognitive symptoms related to the area of the brain being treated due to injury and swelling (Raghavapudi et al., 2021). Similarly, the common side effects of chemotherapy include haematological toxicity, fatigue, nausea and vomiting, rash, and impaired liver function (Omuro & DeAngelis, 2013). Other treatments such as steroids, used to reduce brain swelling, can result in myopathy, hyperglycaemia, personality change, weight gain and insomnia (Dietrich et al., 2011). There are also numerous common side effects from antiepileptic drugs, such as fatigue, nausea, dizziness or visual disturbances and these may be more pronounced and common in patients with brain tumours (Perucca, 2013). Identifying the problems that affect patients and their families is an important first step in providing supportive care.

Numerous studies and reviews have demonstrated high levels of unmet needs in cancer patients, such as lack of support in managing anxiety, depression and fatigue, or lack of information (Barg et al., 2007; Harrison et al., 2009; Hwang et al., 2004; Janda et al., 2008; McDowell et al., 2010; Moore et al., 2013; Pigott et al., 2009; Puts et al., 2012; Sanders et al., 2010). However, patients often do not communicate psychological concerns to their clinicians and there is evidence that health care professionals (HCPs) may not detect emotional distress (Mitchell et al., 2011). The lack of detection of needs for all cancers is problematic, but possibly more significant in patients with brain tumours, as they have one of the highest rates of depression (Hartung et al., 2017). This, in combination with their significant and unique needs, necessitates targeted mechanisms to deliver appropriate supportive care.

The holistic needs assessment (HNA) has been widely used to identify concerns and unmet needs (National Cancer Action Team, 2012). HNA typically includes a review of physical, psychological or emotional, practical, social, environmental and spiritual or other domains of needs to provide a systemic assessment which can aid in planning appropriate supportive care or referrals (Johnston et al., 2019). There are numerous tools that are commonly used for HNA across all cancer types, such as the Sheffield Profile for Assessment and Referral to Care (SPARC) (Ahmed et al., 2014), National Comprehensive Cancer Network (NCCN) distress thermometer and concerns checklist (National Comprehensive Cancer Network, 2013) or the Macmillan electronic HNA (Ipsos MORI Social Research Institute, 2013). However, it has been suggested that these are not suitable for brain tumours as they lack specificity and sensitivity in assessing the unique needs of this patient group, such as neurocognitive symptoms, which are often the most significant problems (Armstrong et al., 2005; Dirven et al., 2018; Janda et al., 2006; Rooney et al., 2014). Cognitive impairments may present additional challenges for this population in completing patient reported outcome measures (PROMs). Therefore, it is important that the development of tools includes individuals with brain tumours to assess user comprehension and acceptability (for example, length and format) and the specific needs of individuals with brain tumours.

This research set out to investigate the role and value of the HNA followed by appropriate support and care and how it could improve brain tumour patients' outcomes. The evidence above highlights the unique and complex nature of this illness and the lack of available research. Consequently, several questions remain relating to what assessment tools might be considered suitable for this group, their experiences and responses to unmet needs, and what interventions or responses could help improve outcomes.

Brain tumour patients in the United Kingdom (UK) report poorer patient experiences than most other cancer patient groups (NHS England and NHS Improvement, 2018; Scottish Government, 2016). Currently, there is little evidence to suggest why this is. However, cancer charities have suggested that an absence of care plans focused on patient needs may be linked to this poor experience (Brainstrust, 2019). The National Institute for Health and Care Excellence (2018), in their guidance for managing brain tumours, confirms the existence of many unmet needs for brain tumour patients, a lack of knowledge relating to what interventions may help in meeting these needs, and even how we should go about appraising the value of such assessment tools with this group. Additionally, referral to support services such as palliative care, that may have an impact on quality of life (QOL) for those with higher grade tumours, is insufficient.

1.2 Complexity of the diagnosis and treatment pathways for brain tumour patients

Cancer causes one in four deaths in the United Kingdom, with individuals born in 1960 having a 50% chance of being diagnosed with cancer at some point in their lifetime (Cancer Research UK, 2015b). Brain, central nervous system (CNS) and other intracranial tumours account for 3% of all new cancer diagnoses in the UK and are the 9th most common cancers (Cancer Research UK, 2015a). Incidence rates continue to rise, with an increase in brain tumour rates of 8% in males and 22% in females in the last decade (Cancer Research UK, 2015a). In contrast to most other cancers, brain tumours occur relatively frequently across all age groups (rather than focused on an older population) (Cancer Research UK, 2015a).

Brain tumours can originate in the brain tissue or the covering of the brain called the meninges (National Institute for Health and Care Excellence, 2018). They can be benign (low grade) or malignant, with most malignant tumours originating in the brain tissue (95%) (Cancer Research UK, 2015a). In the UK, benign brain tumours are treated by neuro-oncology teams and are considered in the same care guidance documents as malignant tumours by organisations such as the National Institute for Health and Care Excellence (2018) due to many similarities in treatments and symptoms. Additionally, statistics are often reported in conjunction or combined with cancer statistics in organisations such as Information Services Division, Scotland (Information Services Division Scotland, 2019) and Cancer Research UK (2019). Therefore, brain tumours encompassing both benign and malignant types will be included in this research.

There are more than 130 types of tumour which can occur in the brain, CNS or intracranial area, but the majority are astrocytomas (34% in England) or meningiomas (21% in England) (Cancer Research UK, 2015a). Within these types, astrocytomas are generally in the brain tissue and 80% of these are classed as high-grade gliomas (IV) which have a median survival of 12 months (Hayat, 2011; National Institute for Health and Care Excellence, 2018). In contrast, most meningiomas (92%) are considered grade 1 (benign). However, despite being classified as benign tumours, they are associated with significant long term neurological morbidity and are similar to cancerous brain tumours in relation to recurrence and impact (National Institute for Health and Care Excellence, 2018). The use of the terminology 'Brain tumour patient' is used throughout this research to mean someone who has received a diagnosis of having had a brain tumour at some point (not time limited). The term 'brain tumour patient' will be used this work for convenience and in keeping with medical literature. However, it is recognised that other terms may be used such as survivor or person affected by a brain tumour, or people living with a brain tumour may be a preferred or more patient centred terminology.

One area of exception is mortality and this is relevant only for those tumours classed as malignant. In 2017, the incidence in Scotland of brain and central nervous system (CNS) cancer was 466 diagnoses with 394 registered deaths (Information Services Division Scotland, 2019). For all malignant brain cancers in adults, the overall survival rate at five years (based on England and Wales) is 18.5% (Cancer Research UK, 2015a) and the five-year survival of a patient with a high-grade glioblastoma (WHO classification IV) is 5% (Omuro & DeAngelis, 2013). This poor survival rate, and the age at which brain cancer occurs, results in it being the most common cause of cancer deaths in 15-24 year olds, the second most common cancer death in adult males, and fourth for females in the 25-49 years old age range in the UK (Cancer Research UK, 2019). However, older patients have an even poorer prognosis, with patient age the single most important prognostic factor among glioblastoma patients (Arvold & Reardon, 2014). The contributing factors to this poor prognosis could include less favourable tumour biology, less aggressive treatment, treatment toxicity, and other comorbidities.

Although the age and grade of tumour vary, there is a significant the impact for most patients and carers with requirement for effective supportive care to help mitigate the significant needs and symptoms. These needs often are apparent at diagnosis with neuro-cognitive symptoms the first sign of the tumour.

1.3 Diagnosis

Patients with brain tumours may often receive a diagnosis after a sudden event, such as an epileptic seizure, caused by irritation of the brain cortex (Goffaux et al., 2012). Up to 58% present at accident and emergency (Ozawa et al., 2018). If presenting through their GP, the most common presentations are focal neurology including stroke (33.2%), then episodic attacks, ‘fits, faints or falls’ (20.8%), and headache (20.8%), with 30% of cases having three or more consultations prior to referral to cancer services (Ozawa et al., 2018). The location of the brain tumour links to functional status and symptoms that the individual may experience may be focal, such as speech difficulties or visual field disturbance (Omuro & DeAngelis, 2013). The figure below illustrates the impact on function that the location of the tumour may have.

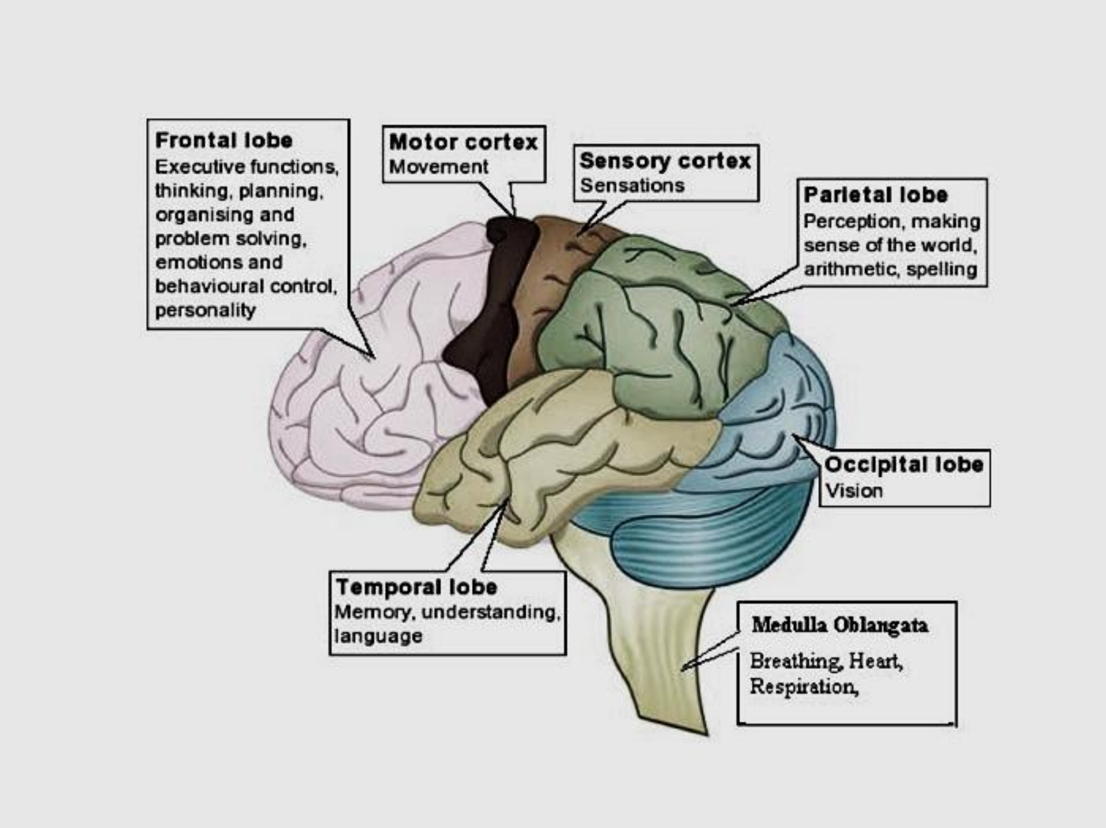


Figure 1 - Functional Areas of the Brain

(The James – Ohio State University, 2021)

Patients in the UK may present to their general practitioner, however, in 60% of cases due to the emergence of acute symptoms they often present at accident and emergency and investigations are subsequently undertaken (National Institute for Health and Care Excellence, 2018). These should include a magnetic resonance imaging scan (MRI). If a tumour is suspected the patient is referred to a specialist neuro-oncology multi-disciplinary team (Natalwala et al., 2011). At this point there may be a decision to proceed to a stereotactic biopsy and/or immediate debulking of the tumour by the neurosurgeon. Based on these investigations and the histopathology results, the diagnosis and treatment options are discussed at a multidisciplinary meeting (Natalwala et al., 2011).

1.3.1 Classifications of brain tumours

Brain and CNS tumours are diagnosed based on the 2016 World Health Organization Classification of Tumours of the Central Nervous System (Louis, Ohgaki et al., 2016). This was updated from the 2007 version, taking account of molecular parameters in addition to histology to define the many tumour entities, as this plays a significant role in treatment decisions and prognostic information.

WHO grades of select CNS tumours	
Diffuse astrocytic and oligodendroglial tumours	
Diffuse astrocytoma, IDH-mutant	II
Anaplastic astrocytoma, IDH-mutant	III
Glioblastoma, IDH-wildtype	IV
Glioblastoma, IDH-mutant	IV
Diffuse midline glioma, H3 K27M–mutant	IV
Oligodendroglioma, IDH-mutant and 1p/19q-codeleted	II
Anaplastic oligodendroglioma, IDH-mutant and 1p/19q-codeleted	III
Other astrocytic tumours	
Pilocytic astrocytoma	I
Subependymal giant cell astrocytoma	I
Pleomorphic xanthoastrocytoma	II
Anaplastic pleomorphic xanthoastrocytoma	III
Ependymal tumours	
Subependymoma	I
Myxopapillary ependymoma	I
Ependymoma	II
Ependymoma, *RELA* fusion–positive	II or III
Anaplastic ependymoma	III
Other gliomas	
Angiocentric glioma	I
Chordoid glioma of third ventricle	II
Choroid plexus tumours	
Choroid plexus papilloma	I
Atypical choroid plexus papilloma	II
Choroid plexus carcinoma	III
Neuronal and mixed neuronal-glial tumours	
Dysembryoplastic neuroepithelial tumour	I
Gangliocytoma	I
Ganglioglioma	I
Anaplastic ganglioglioma	III
Dysplastic gangliocytoma of cerebellum (Lhermitte–Duclos)	I
Desmoplastic infantile astrocytoma and ganglioglioma	I
Papillary glioneuronal tumour	I
Rosette-forming glioneuronal tumour	I
Central neurocytoma	II
Extraventricular neurocytoma	II
Cerebellar liponeurocytoma	II
Tumours of the pineal region	
Pineocytoma	I
Pineal parenchymal tumour of intermediate differentiation	II or III
Pineoblastoma	IV
Papillary tumour of the pineal region	II or III
Embryonal tumours	
Medulloblastoma (all subtypes)	IV
Embryonal tumour with multilayered rosettes, C19MC-altered	IV
Medulloepithelioma	IV
CNS embryonal tumour, NOS	IV
Atypical teratoid/rhabdoid tumour	IV
CNS embryonal tumour with rhabdoid features	IV
Tumours of the cranial and paraspinal nerves	
Schwannoma	I
Neurofibroma	I
Perineurioma	I
Malignant peripheral nerve sheath tumour (MPNST)	II, III or IV
Meningiomas	
Meningioma	I
Atypical meningioma	II
Anaplastic (malignant) meningioma	III
Mesenchymal, non-meningothelial tumours	
Solitary fibrous tumour / haemangiopericytoma	I, II or III
Haemangioblastoma	I
Tumours of the sellar region	
Craniopharyngioma	I
Granular cell tumour	I
Pituicytoma	I
Spindle cell oncocytoma	I

Table 1 - WHO grades of Central Nervous System Tumours (2016)

(from Louis et al., 2016)

1.3.2 Diagnosis of gliomas

Gliomas are recognised as the most frequent brain tumours, so they will be used as an exemplar to detail the diagnostic pathway. Traditionally, these have encompassed a number of different classifications based on the cell origin and grade. Based on the cell type, they are classified into ependymomas, astrocytomas, oligodendrogliomas, and mixed gliomas (Hayat, 2011). Tumours are then classified according to a grading system based on the microscopic appearance. Low-grade tumours (grade 1 and 2) are well-differentiated (non-anaplastic) and high-grade tumours (Grade 3 and 4) are undifferentiated (anaplastic) and have a poorer prognosis (Hayat, 2011). While this provides relevant information, the WHO 2016 classifications include other factors relevant to treatment and prognosis.

The gene or molecular characteristics of glioma are now a routine part of diagnosis to aid treatment decisions. O[6]-methylguanine-DNA methyltransferase (MGMT) promoter methylation and TERT promoter mutations in IDH-wildtype glioma are recommended to be standardised to guide treatment decisions (National Institute for Health and Care Excellence, 2018). In particular, O-6-methylguanine-DNA methyltransferase or MGMT promoter hyper methylation is also associated with enhanced survival in GBMs and can help to determine the optimal treatment regime (Mansouri et al., 2019; Weller et al., 2012).

1.4 Treatment options

Standard treatments based on the diagnostic findings include surgery, radiotherapy, chemotherapy and novel therapies, such as molecular targeted therapies alone or in combination (National Institute for Health and Care Excellence, 2018). For example, optimal treatment for high-grade gliomas involves maximal resection followed by radiotherapy and concomitant and adjuvant temozolomide (Omuro & DeAngelis, 2013). However, not all patients are candidates for this treatment. Factors such as the location of the tumour (not all tumours are resectable), age and performance status need to be considered.

Treatment options are less clear in the elderly. A systematic review of treatment for older patients with glioma highlights that there is a lack of consensus in relation to a number of aspects of optimal treatment for those over 65 (Arvold & Reardon, 2014). However, there appears to be some benefits to concomitant chemotherapy and radiotherapy in those who have good performance status in ages 65 to 70, but not in those over 70 (Rampling & Erridge, 2014). In elderly patients with poor performance status, options include best supportive care, chemotherapy alone, hypo-fractionated radiotherapy alone, or whole brain radiotherapy if needed urgently for symptomatic patients (Omuro & DeAngelis, 2013). For many patients, clinical trials or other research to improve survival or quality of life, such as novel targeted agents, may also be a treatment option.

Medications used to control or alleviate symptoms are common and these often include anti-epileptics for seizures and steroids to control pressure on the brain (Omuro & DeAngelis, 2013)

1.4.1 Surgery

Surgery is used to provide a pathological diagnosis, debulking to relieve distressing symptoms to improve survival or eliminate the tumour (Omuro & DeAngelis, 2013; Wang & Jiang, 2013). The decision to undertake surgery is related to the age of the patient, performance status, location and size of the tumour, and the patient's wishes. Occasionally medical devices such as chemotherapy wafers, which can administer treatments locally to the tumour, are implanted.

The benefits of any surgery must be balanced against the potential morbidity and mortality associated with this invasive procedure. While extensive resection could more fully excise a tumour, and enhance therapy and/or survival, impairment of neurological function is a significant issue (Wang & Jiang, 2013). Postoperative functional deterioration can occur which may be irreversible. Therefore, in some patients who are likely to be incurable, the iatrogenic insult should be minimised. Surgery is infrequently offered alone in high-grade tumours as it is rarely curative due to the glioma cells' permeation into the surrounding brain (Wang & Jiang, 2013). The value of surgery must be considered along with the fitness for other therapy on completion. However, in other tumours such as meningiomas, surgery alone can be the only treatment if the tumour is fully excised and of low grade (National Institute for Health and Care Excellence, 2018). However, radiotherapy often needs to be considered for higher-grade tumours or for an incomplete excision.

1.4.2 Radiotherapy

After surgery, adjuvant radiotherapy combined with chemotherapy should be considered in all high-grade glioma patients (National Institute for Health and Care Excellence, 2018). The typical radiotherapy dose is 60 Gy divided in 30 fractions although variations based on fitness, or access to intensity-modulated radiotherapy, may influence the treatment schedule or mode of delivery (Omuro & DeAngelis, 2013). Decisions to treat brain tumours with radiotherapy need to take in patient preferences, histological features and performance status (National Institute for Health and Care Excellence, 2018). Patients undergoing brain tumour radiotherapy will typically have a number of side effects, which include hair loss, dermatitis, nausea and fatigue. Cognitive symptoms may appear later (Hansen & Roach, 2018). These side effects are related to the area of the brain being treated due to injury and swelling and later effects may appear up to six years after the radiotherapy.

1.4.3 Chemotherapy

Until 2004, only modest survival was demonstrated with the addition of chemotherapy. However, in 2004 the results of large Phase 3 trials were reported, which demonstrated a significant improvement in two-year overall survival, from 10.4% with postoperative radiotherapy alone to 26.5% with post-operative radiotherapy plus temozolomide with high-grade gliomas (Stupp et al., 2005). A recent overview of the current treatment of glioma patients suggests that survival remains similar for patients on this treatment regime outwith trials and this has now become routine practice (Koshy et al., 2011; National Institute for Health and Care Excellence, 2018). In addition, carmustine implants (inserted during surgery) may be an option in this group when > 90% of the tumour is excised (National Institute for Health and Care Excellence, 2018). The role of chemotherapy in low-grade gliomas may be considered in those over 40 who have a 1p/19q co-deleted, IDH-mutated low-grade glioma (oligodendroglioma) (National Institute for Health and Care Excellence, 2018). Other chemotherapy options may be considered, such as PCV (a combination of procarbazine, lomustine and vincristine) in certain circumstances. The common side effects of chemotherapy include haematological toxicity, fatigue, nausea and vomiting, rash, and impaired liver function, with many other side effects often apparent (Omuro & DeAngelis, 2013)

If a large proportion of the tumour is removed (>90%) or surgery is refused, implantable wafers which are impregnated with carmustine may have some additional survival benefit. For this group of patients, this additional treatment can be considered alongside the 'gold standard' of radiotherapy plus temozolomide (National Institute for Health and Care Excellence, 2018a). With locally implanted chemotherapy, side effects such as nausea and vomiting may be fewer, but local effects, such as wound healing delay, brain oedema, seizures or cyst formation can occur (Kuramitsu et al., 2015).

1.4.4 Other treatments

Medications to control symptoms can be numerous with many side effects. These often include anti-epileptic drugs (AEDs) to control seizures and steroids to control pressure on the brain. While steroids are very effective in reducing swelling in the brain, they can also have significant side effects such as myopathy, hyperglycaemia, personality change, weight gain and insomnia – therefore, these should be tapered as soon as possible (Armstrong et al., 2015). There are also numerous common side effects from AEDs, such as fatigue, nausea, dizziness or visual disturbances. These may be more pronounced and common in patients with brain tumours (Perucca, 2013).

1.4.5 Management of recurrence

The vast majority of all high-grade tumours will recur within a year, despite intense multimodality therapies (Wang & Jiang, 2013). However, the management of relapsed gliomas is challenging, with no widely agreed standard of care (Omuro & DeAngelis, 2013). Current treatment options include surgery with or without carmustine wafers, re-irradiation, second-line chemotherapy and anti-angiogenic therapy or treatments within a clinical trial (Omuro & DeAngelis, 2013; Wang & Jiang, 2013). However, the need for specialist palliative and supportive care is a priority at this stage of the disease.

1.5 Symptoms management and supportive care of brain tumour patients

The previous section has given an overview of some of the symptoms, treatments and side effects that patients and their carers or significant others must cope with. Brain tumours differ significantly from other cancers due to their unique symptom profile and a number of studies have demonstrated that there is both an increased symptom burden and a psychological impact of this disease.

1.5.1 What are the experiences of patients and carers?

Patient experience surveys highlight that, in some respects, brain tumour patients may have a poorer experience than those with other cancers, some of which is relevant to the support and care they receive. The Scottish Cancer Patient Experience Survey highlights a number of findings that may help identify areas where there are gaps in the care that patients receive (Scottish Government, 2016). Only 66% of brain tumour patients receive an understandable explanation of side effects (with only sarcoma lower) and only 49% receive practical advice and support for side effects (the lowest score). Other areas, such as confidence in the ward nurses and administration of care, were again the lowest of any tumour group. This survey involved 4,835 cancer patients but only 39 had brain tumours, so the results could have been influenced significantly by one care provider (for example, there are only five clinical nurse specialists in Scotland). In contrast, the English National Cancer Patient Experience Survey from 2017 involved 69,072 cancer patients, 389 with brain tumours. Based on their cross-tumour comparisons on nine core questions, brain tumour patients had the lowest scores on four of the nine questions, including the overall quality of care question.

1.5.2 Supportive care needs of brain tumour patients

The symptoms of brain tumours can have a devastating impact on QOL for patients and caregivers (Ford et al., 2012). Upon diagnosis, patients must deal with the implications of a life-limiting illness whilst dealing with the symptoms from the tumour. The consequent side effects of treatment can also be severe and progressive (Ford et al., 2012). Seizures, headaches, drowsiness and neurological deficits are common symptoms and these affect the QOL and performance status of the patient (Janda et al., 2008; Langbecker & Yates, 2016). Up to 80% of patients present with neurocognitive deficits and these can be accompanied by other symptoms such as motor weakness, aphasia or impaired visual functioning (Day, Gillespie et al., 2016). Physical and neurological functioning can also be strongly affected by the side effects of treatment regimens. Postsurgical morbidity, acute, sub-acute, and late radiation effects on the normal brain, chemotherapy-induced toxicity, high-dose corticosteroids and anticonvulsants can all negatively affect QOL (Langbecker & Yates, 2016).

1.5.3 Neurological symptoms and impact

Patients often experience cognitive dysfunction associated with the tumour and its treatment. Patients who survive more than two years after focused whole brain irradiation have a continually increasing risk of developing dementia over time (Kehayov et al., 2012). Seizures may be presented in 30-50% of patients and these are treated with an AED. However, there is evidence that brain tumour patients are more susceptible to the adverse effects of these drugs, such as impaired concentration (Perucca, 2013). Seizures can also other impact on other areas, such as the loss of a driving licence affecting independence. Chemotherapy is also known to cause cognitive deficits but the mechanisms for this are not clearly defined (Zucchella et al., 2013).

Cognitive dysfunction is seen more frequently at diagnosis in high-grade glioblastomas than in low-grade gliomas (Kehayov et al., 2012). Day, Gillespie et al. (2016) reported that more than 80% of brain cancer patients described neurocognitive deficits during radiotherapy and added that 75% of inpatients will have three or more neurological symptoms, while 39% will have five or more. Another study evaluated the prevalence of neurocognitive deficits in a non-selective cohort of 147 neuro-oncology patients, in which 90% were glioma patients, using a number of validated neurocognitive tests (Zucchella et al., 2013). This study found that 80 (54.4%) showed cognitive impairment, 43 (53.75%) presented a multi-domain impairment, while 37 (46.25%) patients revealed cognitive deficits limited respectively to language (n=13, 16.25%), memory (n=11, 13.75%), attention (n=7, 8.75%), logical-executive functions (n=5, 6.25%), and visual-spatial abilities (n=1, 1.25%).

Personality changes, mood disturbance (anxiety, fatigue, depression) and a decrease in mental capacity and concentration are common (Day, Gillespie et al., 2016). Symptom severity fluctuates during the course of the disease, with many patients who respond to treatment experiencing temporary improvement, and some treatments may cause a reversible deterioration such as radiotherapy due to swelling.

There is evidence that increasing cognitive difficulties can cause social isolation and reduce support networks (Madsen & Poulsen, 2011). While there are many different contributing factors that may cause neurological difficulties, the impact on QOL and interference with normal function can be severe for the patient and their caregiver.

1.5.4 Psychological symptoms and impact

Distress is very common in cancer, with data from 7,000 patients reporting rates of 40% with significant distress (Carlson, et al, 2012). This information was obtained by pooling data from a number of studies which used the Brief Symptom Inventory – 18 (BSI - 18) – which has been extensively validated for use as a screening tool for psychological distress in cancer patients (Zabora,et al., 2001; Zabora, et al., 1990). There is some suggestion that this is even higher in glioma patients, with some studies reporting up to 47% using the same tool (Ford et al., 2012). Another study examined rates of anxiety and depression between surgery and radiotherapy. Based on analysis of the Hospital Anxiety and Depression scale (HADs) scores, it indicated that anxiety was only present in 13-22% of patients (Kilbride et al., 2007). However, in this study, content analysis of the telephone and unstructured interview transcripts indicated that 75% of people were anxious about aspects of their radiotherapy and 58% were anxious about their tumour growing. This disparity may help to explain some of the discrepancies that exist within the literature regarding the prevalence of anxiety after a malignant diagnosis.

It does need to be noted that these are screening instruments – not diagnostic – and it may be advisable that these should only be used as part of an integrative approach (Mitchell et al., 2015). A study which undertook a more robust approach to diagnosis in the form of a structured clinical interview was made utilising the

Diagnostic and Statistical Manual of Mental Disorders (Version 4). Out of 81 patients, major depressive disorders in 20% of brain tumour patients were found in the first eight months after diagnosis (Rooney et al., 2011).

Regardless of the exact incidence, anxiety and depression is quite common in this patient group and the impact of psychological distress can have a significant effect, as this is highly correlated with having a lower QOL (Ford et al., 2012; Hickmann et al., 2017). However, research has not determined if a reduced QOL causes distress or if the distress causes a reduced quality of life. In addition, psychological distress may be linked with poorer perceived cognitive function in brain tumour patients (Nicol et al., 2019).

1.5.5 Are supportive care needs of brain tumour patients different to other cancers?

As previously discussed, patients with brain tumours differ from other cancers in terms of neurological problems, such as cognitive impairments, memory loss, or uncontrolled seizures (Janda et al., 2008). Compared to control groups of patients with non-CNS cancers, and healthy patients, even low-grade glioma patients specifically report more fatigue, cognitive dysfunction and altered mood states (Liu et al., 2009). Fifty-three primary brain tumour (PBT) patients completed a brain tumour-specific, patient concern inventory (PCI) and the top concerns were fatigue (62%), memory (57%), fear of recurrence (57%), concentration (55%) and depression (45%) (Rooney et al., 2014). This contrasts with a prospective study of 45 advanced cancer patients which examined the perspectives of patients, physicians and caregivers. The five most common symptoms were reported to be pain (95%), dyspnoea (88%), tiredness (80%), lack of energy (65%) and dry mouth (65%) (Oechsle et al., 2013). Neurocognitive deficits are infrequent or absent as a symptom in other cancers and the presence of these symptoms in brain tumour patients requires significantly different care and support. This difference is also recognised by those caring for them.

A study in the US investigated what nurses' and doctors' rate as the most significant concerns and symptoms of advanced cancer patients (Cella et al., 2003). They cited headaches and seizures – but these were not ranked for any other cancer. In another study which examined supportive care needs in

Australia, it was found that the symptoms noted by brain tumour patients are unique and exceed those found in other cancer patients (Janda et al., 2008).

1.5.6 Implications for assessing supportive care needs of patients who have experienced a brain tumour

Supportive care is a priority which should be based on the needs of the patient. Those assessing needs should have full awareness of the options for provision of this support (National Institute for Health and Care Excellence, 2018). However, a key element is providing an opportunity which facilitates open communication of needs from the patient's perspective to plan supportive care. Increasingly generic cancer needs assessments, using patient reported outcomes (PROs) – particularly needs assessment questionnaires – have been developed and are widely used in combination with a clinical discussion. It has been suggested that tools developed for assessment of needs, such as HNAs can be used in all cancers to aid in this process (National Cancer Action Team, 2012). However, these may not be appropriate for brain cancer patients due to the lack of focus on neurological symptoms. The complexity of the questionnaire for individuals with cognitive deficits could also affect the ability to complete them (Armstrong et al., 2006; Lai, Jensen et al., 2014; Rooney et al., 2014; Taphoorn et al., 2010).

1.6 Needs assessment in cancer

Needs assessment is a systematic assessment of the unmet needs of an individual to identify concerns and problems with the underlying purpose of addressing these needs (Macmillan Cancer Support, 2015; National Cancer Action Team, 2012). HNA typically includes a review of physical, psychological or emotional, spiritual, social, practical and other domains of needs to provide a systemic assessment which can aid in planning appropriate supportive care or referrals (Johnston et al., 2019). There are numerous tools commonly used for HNA across all cancer types, such as the Sheffield Profile for Assessment and Referral to Care (SPARC) (Ahmed et al., 2014), National Comprehensive Cancer Network (NCCN) distress thermometer and concerns checklist (National Comprehensive Cancer Network, 2013) or the Macmillan electronic HNA (Ipsos MORI Social Research Institute, 2013). In the UK, HNA is part of the recovery package which also includes care planning, a treatment summary and cancer care review (Macmillan Cancer Support, 2018). HNA is the first step of this process but cannot be viewed in isolation as other steps are completed in responses to the HNA findings, so this should be viewed as a package. An important component of HNA is that it gives control back to cancer patients in order to manage their condition (Scottish Government, 2016b) or support self-management (NHS Improvement, 2016). Figure 2 below sets out the recovery package and HNA as it is envisioned within this pathway as part of the assessment and care planning component, which then guides other interventions and supports self-management.

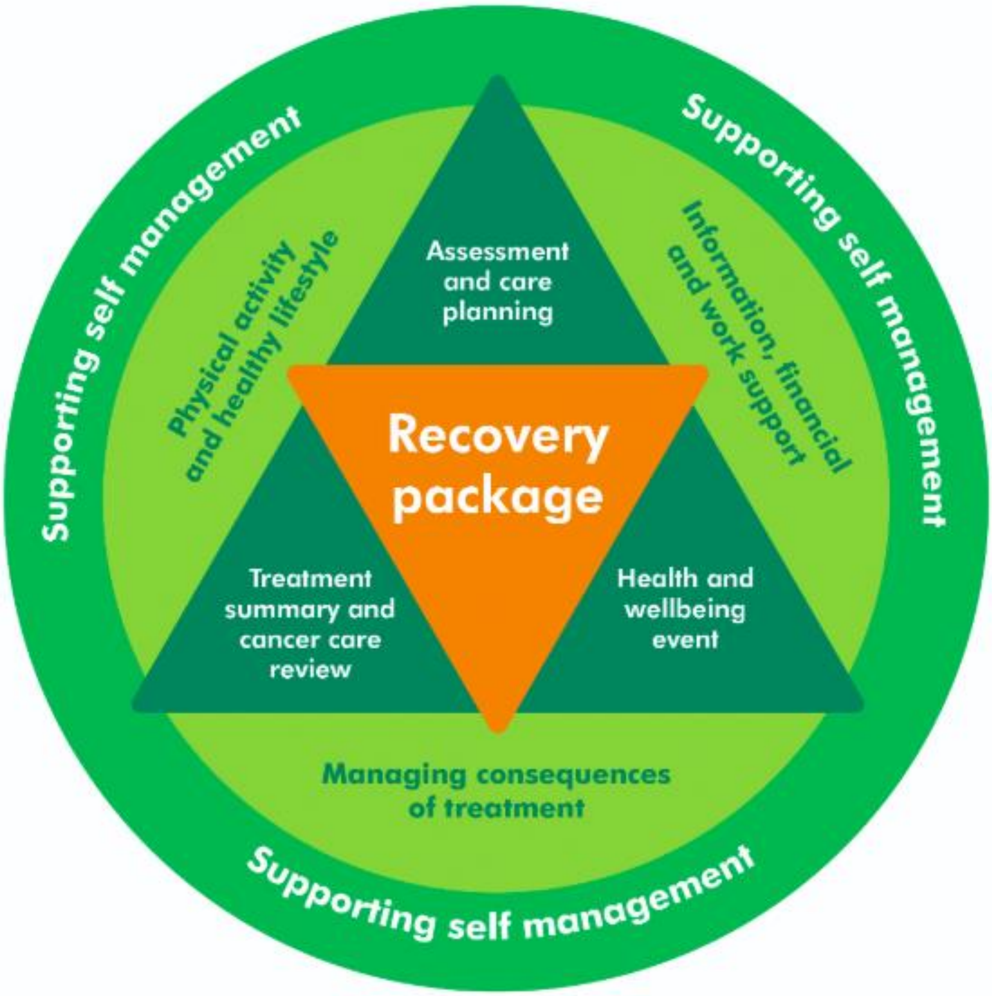


Figure 2 - The UK Recovery Package Model

The rationale for usage of an HNA is not the same in countries. For example, in Canada and the United States where the completion of an HNA has been advocated as an essential process in the detection and assessment of distress in cancer patients (Canadian Partnership Against Cancer, 2012; National Comprehensive Cancer Network, 2013). Routine screening for unmet supportive care needs of oncology patients has been suggested as a necessary component of comprehensive cancer care, as unaddressed needs can lead to inadequate care and long-term problems (Jiao et al., 2015).

Regardless of the focus, early identification of risk for psychological morbidity or other unmet needs could allow targeted interventions if clinically effective screening tools are used (Jiao et al., 2018). Patient-centred care can be facilitated by enabling the patient to more fully engage in their care and self-management, by providing choice about support for their needs, and giving a focus for discussions with HCPs.

1.6.1 Why undertake needs assessment?

There is evidence that physicians and nurses do not accurately assess what patients' concerns are, which can lead to distress and anxiety or inappropriate interventions (Di Maio et al., 2015). However, the introduction of routine use of patient-reported outcomes to assess symptoms, as opposed to normal care, can significantly improve QOL with 32.4% vs 19.3% (P=0.04) reporting a clinically significant improvement (Baratelli et al., 2019). Feedback from brain tumour patients highlights that some areas are not routinely discussed. These could be areas that patients find difficulty in articulating or feel embarrassed to mention, such as fatigue, loss of memory or emotional needs (National Institute for Health and Care Excellence, 2018a). To help ensure that patient concerns are detected, needs assessments should be reported by the patient and PROs are often used to gain this information.

Through undertaking an HNA, person-centred care can be facilitated by allowing the individual to identify their most important needs. This can potentially empower patients to have more open communication with their HCPs (Young et al., 2015).

HNA can be an important process as it should help to alleviate unmet needs and focus supportive care on improving wellbeing for the individual (Bonevski et al., 2000). Since HNAs were first introduced, numerous studies and reviews have demonstrated high levels of unmet needs. Psychosocial or practical needs are often the most commonly unmet among cancer patients around the world (Barg et al., 2007; Harrison et al., 2009; Hwang et al., 2004; Janda et al., 2008; McDowell et al., 2010; Pigott et al., 2009; Puts et al., 2012; Sanders et al., 2010). Many caregivers and patients do not communicate psychological concerns to their clinicians (Wen & Gustafson, 2004) and there is evidence that HCPs are not accurate at assessing these issues (Jiao et al., 2015). One study compared a validated tool (the National Comprehensive Cancer Network (NCCN) distress thermometer (DT) to the clinical judgement of cancer nurse practitioners in 401 mixed cancer patients (Mitchell et al., 2011). The nurses had a detection sensitivity of 50.5% and specificity of 80%, with an equal proportion of false negative as positive. While clinicians have expressed a preference for using their own clinical judgement, this disparity could have implications for either lack of support or inappropriate support (Mitchell et al., 2011). Based on a number of cross-sectional studies, high unmet needs correlate with psychological distress and poorer quality of life. They may also have a significant effect on the carer's wellbeing (McDowell et al., 2010; Soothill et al., 2001; Wen & Gustafson, 2004).

Additionally, there are also potential benefits in appropriate targeting of resources within a health service where these are finite. In the absence of individual needs assessment, there are two negative implications for resources:

- Patients are not given beneficial interventions or support (this will result in unmet needs and implications for services as they may be more resource intensive at a later date)
- Patients are given inappropriate interventions or support (if needs are accurately known, this releases resources).

1.6.2 Evidence of impact of needs assessment

Based on a review of randomised, quasi-randomised or controlled before and after studies which undertook interventions (supportive care) after the identification of unmet needs using validated measures of needs assessment, it was found that there was limited or no evidence to support the reduction in unmet need (Carey et al., 2012). A similar review with broader criteria also included non-randomised studies (but with a comparison group of any kind) and screening using any needs assessment tool (Carlson et al., 2012). This review also found that there was limited evidence of benefit. The strongest area of benefit noted was satisfaction with communication between patients and clinicians. However, the authors highlight that the results demonstrate either equality or a positive trend in terms of needs assessment screening, but that the chance of a Type II error was very high due to the small sample size. More recent studies have also not demonstrated significant findings. A study in lung cancer patients randomised patients to a structured intervention or normal supportive care with the aims of reducing unmet need, psychological morbidity and distress and improving QOL (Schofield et al., 2013). No significant effect was seen on any of the measures and the hypothesis that this would have an impact was rejected.

However, this does raise some questions about research into both needs assessments and the effect of interventions to change unmet needs. One area that may warrant further research is in relation to how we are measuring needs – does the scoring provide a reliable measure or an accurate reflection of the overall unmet need in the individual? (Carey et al., 2012). Or should research be more focused on the effectiveness of interventions to reduce or alleviate needs? (Carey et al., 2012).

These interventions are likely to be varied and complex which, particularly outwith the physical domain, may have large variations in context where effectiveness would be challenging to demonstrate. This view is supported by Catt et al., (2008) who highlight that there is a lack of research around the efficacy of many interventions in supportive care for high-grade glioma. A Cochrane review undertaken by Day et al. (2014), which examined interventions for preventing and ameliorating cognitive deficits in adults treated with cranial irradiation, highlighted that although some interventions may be promising, a lack of numbers, withdrawals and non-randomised studies do not provide conclusive evidence and more research is needed. These authors concluded that more research should be done in relation to the effectiveness of interventions due to the lack of quality evidence. However, some aspects in which there was limited evidence of benefit, such as staff training, should be an important focus for further research as well as more longitudinal studies and a focus on economic evaluations.

However, there may be another contributing factor in relation to HNA. A randomised trial was undertaken to examine the impact of knowledge of patients' functional status on consultations. PROs that included a validated global cancer QOL questionnaire, (the European Organisation for Research and Treatment of Cancer Quality of Life Questionnaire-Core 30) and the Hospital Anxiety and Depression Scale (HADS) were used. The researchers carried out content analysis of 792 consultations of 198 patients and 28 oncologists over three appointments. There were three groups – the first group of patients completed the questionnaires and had the results conveyed to the doctors, the second completed the questionnaires but the results were not conveyed, and the third group did not use the questionnaire. This study highlighted that, even when there are mechanisms to alert oncologists to patients' functional problems, they did not influence the discussion and social or role problems in particular were rarely discussed (Takeuchi et al., 2011). The authors of this study conclude that, despite the known impact of cancer on these aspects of functioning, there continues to be a barrier to discussion. There was, however, some effect on discussion of physical problems, but the authors conclude that targeted interventions to overcome the oncologist's barriers to enable them to discuss the psychological, social and other impacts of cancer. Holistic assessment of needs has the potential to improve outcomes for patients by identifying and resolving their issues quickly.

1.6.3 Challenges of needs assessment and the requirement for research in brain tumour patients

The needs of individuals with brain tumours are significantly different to other cancers as the tumour itself may cause physical and cognitive impairments. These impairments, such as cognition, behaviour, personality changes and other symptoms increase the complexity of assessing and addressing the needs of people with brain tumours (National Institute for Health and Care Excellence, 2018a). Many questions arise when assessing patient needs and arguably all of these require focused research on brain tumours due to their unique and complex impacts. What must be addressed includes a lack of clarity on best practices in initially identifying needs, measuring the importance of needs, the effectiveness of interventions when needs are identified, and how to evaluate this effectiveness.

The latest UK clinical guidance for the management of brain tumours (National Institute for Health and Care Excellence, 2018) has identified that low-grade gliomas have significant symptoms and unmet complex needs. Their review highlighted the gap of any high-quality research in this area. There are also significant gaps in the knowledge base for higher-grade tumours. A systematic review of supportive care needs for glioma patients and their carers found that there is a gap in the knowledge of the breadth of needs across “the patient’s whole illness trajectory and it remains unclear how needs might be best addressed, by whom, and at what point in their care” (Moore et al., 2013, p.152). However, the recovery package which has been integrated to a large extent within the UK, including the integration of HNA as a central component – with a focus on recovery after treatment and survivorship (Macmillan Cancer Support, 2018) – may not be appropriate for cancers with such a poor prognosis. Arguably, the assessment of needs here is even more important.

HNA has also been identified as a specific area of development needed within Scotland. The Scottish Cancer Taskforce for Brain Cancer (2018) identified needs assessment as an area of key importance but it cannot currently be assessed as it lacks quality performance indicators (or evidence-based measures) in the treatment of support for patients affected by CNS/brain cancer. They state that, although a needs assessment for physical, psychological, cognitive, functional, and specifically neurological function is required, there is currently a lack of evidence and measurement tools to support this action, which was identified in the first version of their document in 2013 but remains an issue in the third version published in 2018. Therefore, Chapter 2 will undertake a review of the existing PROs in brain tumours – which have attempted to assess patients’ concerns, problems or issues for use in a clinical setting – to appraise what tools might be used to support an assessment of needs.

1.7 Purpose of research

The purpose of the research presented in this thesis is explore how HNAs followed by appropriate support and care could improve outcomes for brain tumour patients. This will provide information on how healthcare professionals and individuals affected with brain tumours can utilise a holistic assessment of needs and then use this to address them and maximise wellness. This research will first explore the existing tools which could support the assessment of problems or unmet needs in this patient group. However, while it is important to accurately capture the most significant needs for this group, it is also important to ensure that any tool is acceptable to users who may have cognitive difficulties as a result of their brain tumour. This is followed by a qualitative study which explored patients' and carers' perceptions and experiences in relation to unmet needs and strategies which may address these (or what has not helped in addressing these). Finally, a realist synthesis will examine what the relevant contextual factors in an HNA programme are that need to be considered and what mechanisms, such as interventions associated with HNA, can improve outcomes.

1.8 Aim and research questions

The overall aim of this research is to explore how HNA followed by appropriate support and care could improve the outcomes for brain tumour patients. This has led to three main questions that will be the focus of this thesis. To answer these questions, three sequential studies were undertaken – a literature review examining HNA tools for brain tumour patients, a primary qualitative study of patient and carer experiences and finally a realist review focused on how HNA programmes could improve self-management for brain tumour patients.

The three questions were:

1. What tools are there to assess needs or concerns in brain tumour patients? Based on an evaluation of the psychometric properties and clinical utility of such tools as an HNA, what could be recommended for use in this population?

2: What are patients' and caregivers' experiences and perceptions of unmet needs and strategies which may address these (or avert problems before they become unmet needs?). This includes their perceptions of the tools identified from study 1.

3: How, and in what circumstances, might an HNA programme improve self-management in brain tumour patients?

1.9 Importance of this research

There are clear gaps in how needs assessment as part of The Recovery Package is implemented. Halpern, McCabe & Burg (2016) highlight the lack of evidence in knowing which survivorship care models provide the best care. In addition, another clear question is, what is the role of the patient or survivor in his or her own recovery and care? They also highlight that clinicians will be resistant to implementation of any strategies without a strong evidence base, proof of increased efficiency or attached resources. While the paper above discussed this as an issue for all cancers there is an even more pressing need for research in brain tumours due to their high symptom burden, high levels of psychological distress and comparatively poor patient experience. Evidence supports the significant and unique needs of brain tumour patients – however, there are clear gaps in how to assess and support these. The role of self-management and stratification of care, an integral part of cancer care in the UK, has received some focus and research in other cancer types – but at the time of writing there has been no examination of this for brain tumour patients. This thesis provides some initial evidence of how strategies for survivorship, focused on self-care, could have a positive impact for this patient group and will provide a basis for evidence-based pathways of care to be tested in further research. In addition, this research will provide the basis for development of brain tumour-specific assessments and interventions to maximise wellness for this group.

1.10 Structure of the thesis

This research was structured around three sequential research studies – a systematic review, a qualitative study of patients and carers, and a realist synthesis. The research initially focused on examining how to assess unmet needs in brain tumour patients and how to respond to these assessments or seek to address them. The first stage was the systematic review followed by a study which sought the views of patients and carers on their experiences and perceptions of unmet needs, how these have been supported (or not) and how strategies such as HNA might support this. The results provided some key considerations in relation to the next stage that influenced the question and development of the final stage. It was important to view HNA as a package of interventions, similar to the UK Recovery package (Macmillan Cancer Support, 2015), as assessment without responding to unmet needs is potentially detrimental. It also needed to consider the complexity of both the patient group and the HNA package of interventions and outcomes. Therefore, undertaking a realist synthesis which examines how HNA could work for brain tumour patients, and in what circumstances, to improve self-management would be a beneficial approach to answer these questions. Self-management was the initial focus in line with the UK recovery package which encompasses HNA.

1.10.1 Chapter 1

Chapter 1 provides the background to support and introduce relevant contextual information on brain tumours. This chapter covers information on the epidemiology and diagnostic and treatment pathways for brain tumour patients. It examines symptoms and areas of potential holistic needs, including the impact on the patient and their family. Also included is an overview of HNA and the potential role that HNA and associated interventions can have on unmet holistic needs. Finally, a summary of the rationale for research into this topic is presented.

1.10.2 Chapter 2

Chapter 2 presents a systematic review of potential tools that could be used in brain tumour patients, focusing on their psychometric properties and utility as an HNA tool. This includes methods, findings and discussion.

1.10.3 Chapter 3

Chapter 3 presents a qualitative study which explores the views of patients and carers on their experiences and perceptions of unmet needs, how these have been supported (or not) and how strategies such as HNA was perceived or experienced. This includes methods, findings and discussion from this study.

1.10.4 Chapter 4

Chapter 4 provides an overview of realist methodology and then a realist synthesis of examining how HNA could work to improve self-management in brain tumour patients and in what circumstances. This includes methods, findings and discussion.

1.10.5 Chapter 5

Chapter 5 brings together the findings of all the studies for additional discussion. This provides the basis for a conceptual practice model and implications in this research for practice, policy and further study. It also provides an overview of strengths and weaknesses and the conclusion of this thesis.

1.10.6 Chapter 6

Chapter 6 provides a brief conclusion for the thesis.

2 Chapter 2 - A review of tools which identify needs in brain tumour patients (Study 1)

2.1 Introduction

The aim of this review was to systematically identify and evaluate the psychometric properties and clinical utility of tools that assess needs, problems or concerns in brain cancer patients. This review sought to identify tools that were developed and tested specifically on a brain cancer population which could be used to support HNA. This defined HNA tools as those that identified an unmet need that prevented optimal well-being. The tools needed to look at a minimum of two of the domains of need in the categories of physical, psychological or emotional, spiritual, practical or social needs. The search was focused on tools designed to assess need in clinical care as opposed to outcome measurements purely for clinical research. This did include those classified as “symptom” assessment measures as these have been suggested and used as a strategy to support problem identification (Canadian Partnership Against Cancer, 2012; Watanabe, Nekolaichuk, & Beaumont, 2012).

2.2 The aim of the literature review

The aim of the review was to identify and evaluate the psychometric properties and clinical utility of tools, which assess needs or concerns in brain tumour patients.

2.3 Methods

A literature search was undertaken from February to May 2014. This was repeated 01 February 2018 to 27 February 2018 to check for more up to date publications and the findings from this final search are reported here. The following electronic databases were searched: MEDLINE, Cumulative Index to Nursing and Allied Health Literature (CINAHL), and PsycINFO. This was to identify the literature which reported the development, psychometric testing or clinical utility testing of tools that were developed as a PROM and undertake need or symptom assessments in brain cancer. The search strategy aimed at including all relevant literature; but was limited to the most common databases, studies on humans and studies on adults.

The following search terms were included:

(Needs Assessment) OR (Symptom Assessment) OR (Self-Assessment) OR (Patient Outcome Assessment) OR (inventory or tool* or measure* or instrument*) AND (Brain Neoplasms). Dependant on the database, Subject Headings which encompassed relevant concepts were used or some terms were limited to abstract and title to improve the sensitivity and specificity. The search strategy was proposed by JA and checked and revised by a healthcare university librarian. Citations from relevant research articles or systematic reviews of cancer HNA tools were reviewed for potentially relevant research studies. The corresponding authors of the relevant tools were contacted to request additional information.

2.3.1 Study Selection

Each paper was assessed for relevance by using the following inclusion and exclusion criteria. Inclusion criteria included primary research published in English which reported the development, psychometric testing or clinical utility testing of PROMs for the assessment of adult (age $\geq$ 18) brain cancer patients' needs or generic cancer needs assessment tools developed specifically on a brain cancer or brain tumour population. Studies that included benign brain tumours as well as brain cancers will be included due to the similar symptom profile. However, studies were excluded which only included patients who have brain metastasis due to the differing symptom profile which would be present due to their underlying primary cancer diagnosis. Tools were selected that identified unmet needs, concerns or problems. All literature, which contributed to the development of 'brain specific' content of a need assessment tool, was included in the review. The tools had to have been developed for completion by patients. There were no date limits to ensure all development studies were included. Exclusion criteria were any other PROMs which do not measure needs (for example health related quality of life (HRQOL) or satisfaction) or those that only looked at only one aspect of need (for example information needs).

Each identified article was saved onto a reference manager (Mendeley Desktop, (Mendeley Ltd., 2017) as it was retrieved. This also ensured that duplicate articles from previous searches were clearly identified.

2.3.2 Data extraction and quality appraisal strategy

Data from the selected studies was extracted using standardised forms and these were then transferred to the two collated tables. The tool characteristics are presented in Table 4 and the data which represented the psychometric properties of the tools is summarised in Table 5. This information was then used to support the evaluation of each of the tools psychometric properties and use as an HNA tool.

To evaluate the psychometric properties of each identified tool, the 'Consensus-based Standards for the selection of health Measurement INstruments' or COSMIN checklist was used (Mokkink et al., 2006; Mokkink et al., 2010). This checklist was developed through a Delphi study of 57 international experts to help select an instrument for use, to review studies, to design or report the measurement properties of a new tool (Mokkink et al., 2010). This checklist provides evaluation criteria for psychometric properties (Mokkink et al., 2010) which are then given a rating of excellent, good, fair, poor or not assessed by taking the lowest rating of any of the items that make up each attribute (Terwee et al., 2012). Interpretability and generalizability are not rated but a list of considerations is provided as no scoring criteria was developed for these properties (Terwee et al., 2012). Figure 3 below provides a pictorial representation of the four areas examined in the criteria and Table 2 provides a description of the psychometric properties.

(Mokkink et al., 2010)

Figure 3 - COSMIN taxonomy

Three studies have proposed evaluation criteria for HNA tools, which were used in systematic reviews and research (Bonevski et al., 2000; A. Richardson et al., 2007; Wen & Gustafson, 2004). Below is a summary of each of their criteria.

Wen & Gustafson, (2004) undertook a literature review and developed a conceptual model focused on burden, related constructs and identification of need. Utilising their conceptual model and adding relevant psychometric properties, they proposed the following criteria for evaluation of needs assessment tools:

1. Conceptual and instrument model, which includes examining the tools for development with a conceptual model, examination of dimensions (i.e., factor analysis)
2. Validity, with a focus on content and construct validity
3. Reliability, focused on internal consistency

4. Reproducibility, represented by test-retest and inter-rater reproducibility
5. Responsiveness
6. Burden, with a focus on time to complete and the administrative burden

Richardson et al., (2007) proposed the following evaluation criteria:

1. Validity, with a focus on construct validity and content validity
2. Reliability
3. Appropriateness, considers the match between specific purpose, population and setting for which a tool was developed and how this relates to the intended use
4. Responsiveness
5. Feasibility, examines the extent of effort, burden and disruption to staff and clinical care arising from use of a tool
6. Acceptability, considers how acceptable a tool is for respondents to complete

Bonevski et al., (2000) suggested the following six criteria:

1. Measure of the multidimensional impact of cancer on patients' needs
2. Directly and comprehensively assess subjective health-related needs
3. Measure needs within a defined temporal context
4. Demonstrate acceptable reliability and validity
5. Be user-friendly
6. Be system-friendly

Based on the above lists and details provided in each paper evaluation criteria were formulated for this review to provide a comprehensive evaluation of all aspects related to HNA tools in combination with the methodological and psychometric appraisals. These publications all included the need for good psychometric properties focused on validity and reliability, a consideration of user acceptability for patients and those administering and interpreting the results. All authors in their proposed criteria mentioned tools should capture the holistic dimensions of need - although the exact criteria varied. Ratings based on these criteria were determined as detailed in Table 3.

Table 2 - Definitions of Psychometric properties for PROMs

Psychometric property	Definition	Considerations
Validity	The degree to which an HR-PRO instrument measures the construct(s) it purports to measure	
Content validity:	Content validity seeks to assess if the component parts and tool measures what it is intended to	Initial development through the literature, expert opinion and patient input Refinement of item selection and phrasing through end users
Hypothesis testing or (Construct validity):	The degree to which the scores of the PROM are consistent with hypotheses (for instance with regard to internal relationships, relationships to scores of other instruments, or differences between relevant groups) based on the assumption that the tool validly measures the construct to be measured.	Any measurement tools used, as a comparator, should have adequate measurement qualities. Two criteria should be met; 1) that hypotheses should be stated in the methods including magnitude and 2) 75% of the results are in accordance with these hypotheses which should be reported as a correlation
Structural validity	The degree to which the scores of a PROM instrument are an adequate reflection of the dimensionality of the construct to be measured	This is appropriate for use when a measurement tool is based on reflective model - not for those based around a formative model. HNA is likely to be considered formative due to independent contributing factors.
Cross-cultural validity	The degree to which the performance of the items on a validity translated or culturally adapted PROM instrument are an adequate reflection of the performance of the items of the original version of the PROM instrument	

Reliability	Refers to the stability of responses over time or between respondents (reproducibility) and the consistency of the items in the tool. Reliability contains two measurement properties: Internal consistency and test-retest reliability	
Internal consistency:	The extent to which items in a questionnaire scale are correlated, thus measuring the same concept.	The most common method used is Cronbach's alpha and it should be calculated for each dimension separately.
Reliability: Test–retest reliability:	The degree to which repeated measurements in stable persons provide similar answers.	Correlation values between administrations of 0·70 are considered acceptable. Two weeks is often considered an acceptable time gap
Measurement error:	Refers to changes in the scores of the tool that are not attributed to a true change in the construct to be measured.	This will require two measurements usually about 2 weeks apart (to prevent recall) and the construct to be measured should remain stable.
Responsiveness: (also called sensitivity)	Refers to the ability of a tool to detect changes over time, which correlates with the construct being assessed.	
Interpretability	Refers to the ability to attach meaning (for example commonly understood concepts) to the tools results or changes in the tools results. This looks at the degree to which one can assign qualitative meaning to quantitative scores.	An important concept within this is the ability to detect the MIC which is the lowest level at which a patient feels a change is important to them. Interpretation of this is gives a clinically important level for clinicians assessing the effects of treatments or need for intervention (de Vet et al., 2011).

HR-PRO = Health-related patient reported outcome, PROM = patient reported outcome measure. HNA = holistic needs assessment, MIC = minimal important change Derived from the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) checklist (Mokkink et al., 2012; Mokkink et al., 2010)

Table 3 - Criteria for Tools for Holistic Needs Assessment

Needs assessment characteristic	Definition	Recommendations if applicable	Rating
Planned use of tool	Tools can be primarily designed for research or clinical use. The approach to needs assessment in most cases will be with a specific tool for this purpose	A holistic needs assessment tool should be designed with the purpose of assessment of patient needs for clinical purposes and developed and tested for the intended population (A. Richardson et al., 2007). – however, some strategies such as Canadian Partnership Against Cancer (2009) have advocated symptom assessment with an amended problems checklist as a reasonable approach	**Weak** – primary purpose is not needs assessment and/or not developed for clinical use **Strong** – primary purpose is brain tumour patient needs assessment in clinical practice
Identification of the dimensions of need	The literature clearly identified a number of domains to be considered which included physical, psychological, social, emotional, financial, sexual, functional and spiritual domains in HNA (Cleeland et al., 2000; Johnsen et al., 2011; Schofield et al., 2012; Waller et al., 2008).	HNA tools should assess the multidimensional impact of cancer. Within this criterion, reference to a theoretical or conceptual framework can help to identify factors, which are important to assess.	**Weak** – no theoretical framework, not covering the majority domains of needs **Moderate** – covering majority of domains of needs but no theoretical framework **Strong** – theoretical framework and covering the majority domains of needs
Psychometric properties	Demonstrate strong psychometric properties	The COSMIN criteria were chosen to assess the psychometric properties as described in the previous section.	**Weak** – Little evidence of psychometric properties **Moderate** – some limited evidence of psychometric properties **Strong** – good evidence of

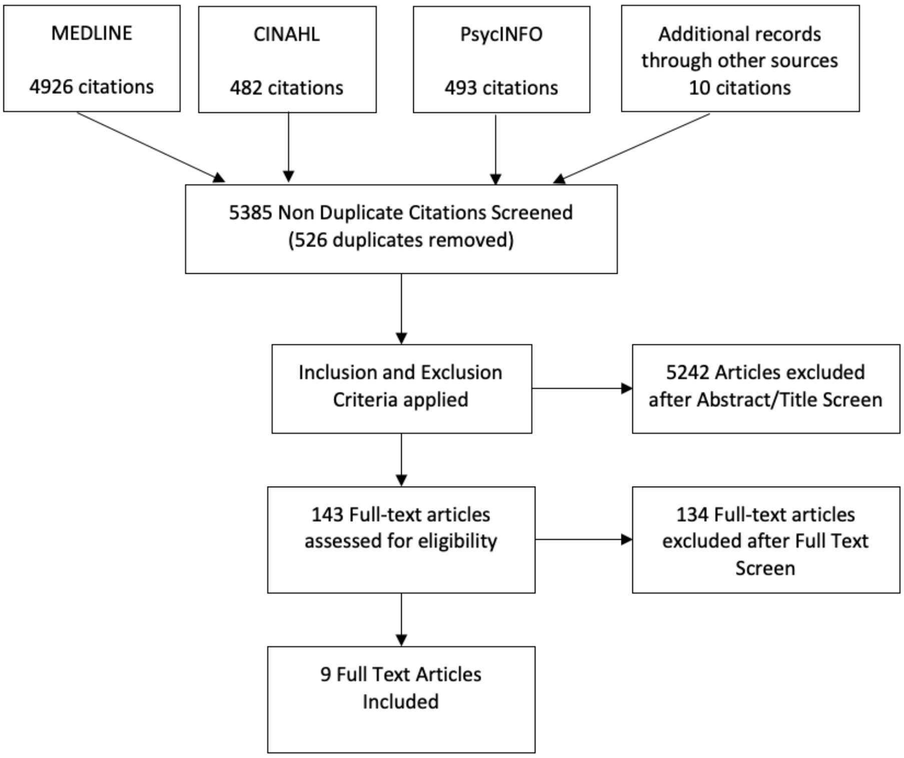
			psychometric properties
4 Be user-friendly	A number of elements should be considered such as the time to complete, ease of completion, comprehension, and perceived usefulness as a tool to communicate needs.	It is generally agreed that reading level for patient reported health outcome measures should not exceed 12 years of age (Streiner & Norman, 2008). In patients with PBTs, cognitive impairment is very common with 54% demonstrating some form of cognitive impairment often related to language, memory and attention (Zucchella et al., 2013). Lai, Jensen, et al., (2014)	**Weak** – no user assessment **Moderate** – some subjective assessment from users **Strong** – subjective assessment from users and objective assessments (for example encompassing assessment of reading level, acceptably form completion requirements and recall time frame)
5: Be assessor friendly	The time, energy and resources for those receiving and responding on questionnaires needs to be considered (Maguire et al., 2013). This criterion relates to the ease of interpretation and usefulness to clinicians to support HNA. It should also consider variables that may affect health care resources such as does it add time to the overall consultation	Vodermaier, Linden, & Siu, (2009) highlight that a short questionnaire of 5-20 items may have a moderate chance of use in busy clinics compared to longer formats, and there may be resources associated with scoring longer questionnaires if required.	**Weak** – no assessment **Moderate** – some subjective assessment from clinicians or objective assessment **Strong** – subjective assessment from users and objective assessments (for example encompassing usefulness, ease of interpretation and resource implications

HNA = holistic needs assessment, COSMIN = COnsensus-based Standards for the selection of health Measurement INstruments

2.4 Results

The search was undertaken from 01 February 2018 to 27 February 2018 and this process is illustrated in Figure 3. The total number of articles identified using the search criteria was 5901 and 526 duplicates were removed leaving 5375 articles to screen. After reviewing titles and abstracts there were 142 remaining articles screened by full text and 8 were included in the review. Reference lists were reviewed to search for any additional relevant citations, and none were found. Corresponding authors of tools were contacted, and this process provided one other relevant paper.

Figure 4 - Systematic search



In total, nine articles were identified describing four tools, which have the potential to assess needs, problems or concerns in brain cancer patients and a summary of their characteristics is presented in Table 4. These were the Supportive Care Needs Survey 34 plus brain subscale (SCNS34-BS); MD Anderson Symptom

Inventory – Brain Tumor Module (MDASI-BT); Brain PCI; and the National Comprehensive Cancer Network/Functional Assessment of Cancer Therapy-Brain Symptom Index (NFbrSI-24) and the studies which developed these tools are detailed in the following section.

Summary of tools and the developmental studies

Brain Patient Concern Inventory

There was only one study which reported the development of the Brain PCI (Rooney et al., 2014). This study described the process for questionnaire design; however, the number of healthcare professionals or geographical location was not specified for those involved in developing content. In the assessment of user acceptability, 45 patients were recruited from a cancer centre in Scotland. In addition, 21 feedback forms were obtained from healthcare professionals in this centre but it was unclear how many participants were involved (Rooney et al., 2014).

MD Anderson Symptom Inventory – Brain Tumor Module

There were four studies that reported the development of the MDASI-BT for clinical and research purposes. The first study undertook item generation and content validity (Armstrong et al., 2005), the second focused on reliability and validity testing (Armstrong et al., 2006), and two studies looked at the test-retest reliability (Armstrong, Vera-Bolanos, Acquaye, Gilbert, & Mendoza, 2014; Armstrong et al., 2012). All participants for all studies were recruited from a large cancer centre in Texas, with the exception of the inclusion of experts recruited to develop content validity with 50% recruited out with the institution (Armstrong et al., 2005).

National Comprehensive Cancer Network/Functional Assessment of Cancer Therapy-Brain Symptom Index

There was one study that reported the development of the NFbrSI-24 (Lai, Jensen, et al., 2014). However they utilised survey results from a previous study of 69 healthcare professionals from throughout the United States (Cella et al., 2003) in the development of content validity. The main developmental study recruited 50 patients with advanced PBTs from six National Comprehensive Cancer Network institutions along with ten physician experts (Lai, Jensen, et al., 2014).

Supportive Care Needs Survey – 34 plus brain subscale

There were two studies that reported the development of the SCNS34-BS (Janda et al., 2006, 2008) designed to look at supportive care needs. The first study focused on content validity and involved 18 patients and carers in item generation (Janda et al., 2006). The second study's main aim was to look at supportive care needs, however the relationship with anxiety and depression was examined and this aspect provides an assessment of construct validity (Janda et al., 2008). The studies were completed with a supportive care service in a region of Australia. In the following section, relevant psychometric properties of these four tools will be evaluated

The Supportive Care Needs Survey – 34 (SCNS-34) is a generic needs assessment tool for all cancer patients (Bonevski et al., 2000) with a specific supplementary brain tumour subscale (Janda et al., 2006, 2008). The MD Anderson Symptom Inventory (MDASI) is a generic assessment of psychological and physical symptoms which examines the impact of these in a number of aspects such as interference with activities of daily living (Cleeland et al., 2000). MDASI-BT was developed to include brain specific symptoms and the development and testing of this was undertaken in a further three studies (Armstrong et al., 2012; Armstrong et al., 2005, 2006). The third tool was a brain PCI developed by Rooney et al., (2013) based on a PCI that was developed for Head and Neck cancers (Rogers et al., 2009). The 24 item NFbrSI-24 was developed to identify symptoms and concerns which are most important to patients (Lai, Jensen, et al., 2014). The content validity for this tool was provided from two sources, first data were used which came from the development of the Functional Assessment of Cancer Therapy-Brain (FACT-Br) which is a specific QOL questionnaire for patients with brain tumours (Weitzner et al., 1995). Second, content validity from health professionals was provided from a study on advanced symptoms in cancer, which included data from a subgroup of HCPs who provided feedback specifically on brain cancer (Cella et al., 2003).

Table 4 - Summary of tool properties

Tool	Purpose	Domains	N of items	Response options	Recall period	Scoring	Time to administer
Brain PCI	HNA CU	Practical, family, emotional, spiritual and physical – with options to request referral and space to ask questions.	58 plus 4 free text questions	Tick box to 'issues that have been a concern'	'recently'	Tick box only	60% of patient report <15 minutes
MDASI-BT	SYM CU & R	Six affective, cognitive, focal neurologic deficits, constitutional, generalized symptom, and a gastrointestinal related factor.	29	Scaling – A 11 point Likert scale in relation to the presence and severity of each symptom in the, with 0 being ''not present'' and 10 being ''as bad as you can imagine" 11 point Likert scale in relation to level of interference.	Last 24 hours	Individual symptoms scored none, mild (1-4), moderate (≥ 5) and severe (≥ 7) and average for overall rating of symptom burden	Less than 10 minutes
NFbrSI-24	SYM CU & R	Three subscales; disease related symptoms (physical and emotional), treatment side effects and functional/wellbeing.	24	Scaling - A five-point intensity scale based on 'how it applies to you' from 'not at all' to 'very much'	Last 7 days	Individual symptoms/concerns are scored with higher scores reflecting less symptoms and concerns (negatively worded items are reversed). Each domain has a score as well as overall questionnaire.	NR

SCNS34-BS	HNA CU	**Format** - Brain specific 'add on' questionnaire to SCNS34. Five domains SCNS34 (Psychological, Health system and information, physical and daily living, patient care and support, sexuality); care needs. The brain subscale has no grouping of domains	50	Scaling **-** Five categorical responses, ranging from high need to no need.	Last month	Patients grouped according no or low needs and those with moderate or high needs.	NR

Brain PCI – Brain Patient Concern Inventory, MDASI – BT – MD Anderson Symptom Inventory and Brain Tumor Module, NFBrSI-24 - 24-item National Comprehensive Cancer Network/Functional Assessment of Cancer Therapy-Brain Symptom Index, SCNS34-BS – Supportive Care Needs Survey (Short Form) with Brain Subscale
HNA = holistic needs assessment, SYM = symptom assessment, CU = clinical use, R = research or clinical trial use
NR = not reported

Table 5 – Sample characteristics of developmental studies and psychometric properties

Tool	Source	Sample characteristics				Content process	Validity	Hypothesis testing/construct validity	Internal consistency	Reliability	Interpretability
		location	N* (subgroups if applicable)	Sex (%)	Age in years range (%) or mean = x						
Brain PCI	(Rooney et al., 2013)	UK One cancer centre	53	M (55), F (47)	18-34 (11), 35-59 (62), >60 (21)	• Other tools • Expert opinion		NR	NR	NR	NR
MDASI – BT	(Armstrong et al., 2005)	USA (multi location)	20 (16 = HCP 4 = PBT or C)	NR	NR	• Theoretical framework • literature review • Expert opinion • CVI > 0.80)		NR	NR	NR	NR

MDASI – BT	(Armstrong et al., 2006)	USA One cancer centre	201	M (57), F (43)	18-45 (52) 45-84 (48)	NR	Principal component analysis endorsed the six underlying constructs	6 symptom scales and interference scale (Cronbach's α = 0.87, 0.82, 0.72, 0.81, 0.69, 0.67 and 0.91)	NR	Correlation with KPS *p* < 0.001 And IP vs OP *p* <0.0005
MDASI – BT	(Armstrong et al., 2012)	USA One US cancer centre	230 (115 PBT and C dyads) (Test–retest subgroup N = 21 PBT)	PBT = M (63), F (37) C = M (27), F (73)	x = 48.2	NR	NR	NR	mean symptom severity, r = 0.952, *p* < 0.0001; mean interference, r = 0.783, *p* < 0.0001) 2 hours	NR
MDASI-BT	(Armstrong et al., 2014)	USA One US cancer centre	100 92 completing	M (62), F (38)	19-77 x = 48	NR	NR	NR	Day 7 Cronbach's α = **Symptoms (overall)**	NR

			three time-points						**0.91**, Affective 0.86, Cognitive 0.94, Neurologic 0.74, Treatment related 0.53, Generalized 0.68, disease GI (composite) 0.42, **Interference (overall) 0.93**, WAW 0.89, REM 0.88	
NFbrSI-24	(Cella et al., 2003)	USA Multiple sites	69 (all HCP)	NR	NR	• Literature review • Other tools • Expert opinion (ranking)	NR	NR	NR	NR

NFbrSI-24	(Lai, Jensen, et al., 2014)	USA Six cancer centres/hospitals	50 plus 10 HCP (for Content validity ONLY)	M (66), F (34) NR	x = 52.2	• Patient survey to refine items from 20 to 24 • Expert opinion (domains)	CV FACT General, physical, social, emotional, and brain tumour–specific concerns (ρ = 0.59, 0.57, 0.40, 0.35, and 0.50, respectively; Ps < 0.05)	full tool; disease related symptom subscale; functional wellbeing subscale; treatment side effect scale (r = 0.84, 0.79, 0.89, 0.65)	NR	The NFBrSI-24 and its subscales significantly differentiated patients with different levels of functional status - ECOG – PS: (F2,47 = 8.21; *p* < .001)
SCNS34-BS	(Janda et al., 2006)	Australia One support group	36 (N = 18 C, 18 PBT)	M (30.5), F (69.5)	NR*	• Literature review • Expert input • Focus groups • Interviews	NR	NR	NR	NR
SCNS34-BS **	(Janda et al., 2008)	Australia Mailing list of support group	75	M (46), F (54)	< 50 (53.5) >50 (46.5)	NR	Patient adjusted odds ratio with patients categorised as	NR	NR	NR

high need (95% CI)

Depression > 11 = OR 2.11 (CI - 1.10-4.03)

Anxiety > 11 = OR 2.89 (CI- 1.29-6.45)

Brain PCI – Brain Patient Concern Inventory, MDASI – BT – MD Anderson Symptom Inventory and Brain Tumor Module, NFBrSI-24 - 24-item National Comprehensive Cancer Network/Functional Assessment of Cancer Therapy-Brain Symptom Index, SCNS34-BS – Supportive Care Needs Survey (Short Form) with Brain Subscale

NR = details not reported in article

M= male, F = female

PBT = primary brain tumour patient C = Carer HCP = health care professional

X = mean

CVI = content validity index

DV = divergent validity, CV = convergent validity

FACT - Functional Assessment of Cancer Therapy

KPS – Karnofsy Performance status, ECOG-PS = Eastern Co-operative Oncology Group Performance Status

REM = relate-enjoy-mood, WAW= walk-activity-work

* All primary brain cancer patients unless otherwise specified

**Study also reported results of 70 carers separately

2.4.1 Evaluation of Psychometric properties

The developmental studies and their psychometric properties are outlined in Table 5 and the quality scoring based on COSMIN criteria (Terwee et al., 2012) is detailed in Table 6.

Table 6 - Psychometric testing quality rating

Evaluated measurement properties	Content Validity	Hypothesis testing	Internal consistency	Reliability test retest
Brain PCI	Poor	NA	NA	NA
MDASI – BT	Good	Good	Excellent	Good
NFbrSI-24	Excellent	Good	Fair	NA
SCNS34 – BS	Fair	Poor	NA	NA

Brain PCI – Brain Patient Concern Inventory, MDASI – BT – MD Anderson Symptom Inventory and Brain Tumor Module, NFBrSI-24 - 24-item National Comprehensive Cancer Network/Functional Assessment of Cancer Therapy-Brain Symptom Index, SCNS34-BS – Supportive Care Needs Survey (Short Form) with Brain Subscale, NA = not assessed

Validity

Content validity was developed in all tools with some advised approaches such as the use of literature, reference to other tools and the input of experts as well as end users (de Vet et al., 2011; Reeve et al., 2013; Scholtes et al., 2011). Item generation was completed with the end users for the SCNS34-BS (Janda et al., 2006), MDASI – BT (Armstrong et al., 2005) and the NFbrSI-24 (Lai, Jensen, et al., 2014) however, this important aspect was missing from the Brain PCI (Rooney et al., 2014). The next step of content validity should undertake a more extensive evaluation to assess comprehensiveness and comprehensibility. This was evident in the MDASI – BT (Armstrong et al., 2006) and the NFbrSI-24 (Lai, Jensen, et al., 2014) with both tools utilising qualitative and quantitative methods. The MDASI – BT which has met the criteria for a ‘good’ rating rather than ‘excellent’, as there were less than 10 participants from the end users (patients) and the NFbrSI-24 was rated as ‘excellent’.

Another aspect of validity is construct validity, or hypothesis testing, which is frequently undertaken with QOL, performance status, anxiety and depression, or distress (Richardson et al., 2007). The three studies that analysed this aspect were rated from ‘poor’ to ‘good’. The SCNS34-BS examined the relationship between supportive care needs and distress utilizing the Hospital Depression and Anxiety Scale (HADS) (Zigmond & Snaith, 1983). Multivariate logistic regression analysis was performed rather than recommended test of a correlation co-efficient resulting in the ‘poor’ rating. However, it should be acknowledged this was not the stated purpose of the study and this did demonstrate a relationship between these constructs. The NFbrSI-24 (Lai, Jensen, et al., 2014) was rated ‘good’ rather than ‘excellent’ as the sample size was < 100 and the MDASI-BT was rated ‘good’ rather than ‘excellent’ due to only recruiting from one centre (Armstrong et al., 2006).

No studies undertook measures of structural validity, however as HNA is a likely to be a formative model, as need is comprised of many unrelated factors, this property is not relevant (Terwee et al., 2012). There were no studies examining cross-cultural validity.

Reliability

The SCNS34-BS (Janda et al., 2008, 2006) and the Brain PCI (Rooney et al., 2014), did not undertake any testing of reliability in the reviewed studies. The MDASI-BT looked at internal consistency for each sub-scale (Armstrong et al., 2006) and demonstrated ‘excellent’ internal consistency within the recommended range (Mokkink et al., 2012). The NFbrSI-24 demonstrated acceptable internal consistency for the full symptom index however the treatment side effect subscale fell slightly below recommended limits (α = 0.65), which corresponds to a ‘fair’ rating for this property (Mokkink et al., 2012).

The inter-rater reliability and test-retest reliability for the MDASI-BT was first reported in Armstrong et al., (2012). As this review is focused on the use of tools as a PROM the inter-rater reliability is not relevant. The test-retest variable was measured on a subgroup of 21 patients using Spearman correlations between the two time points and the analyses supported test-retest reliability but it was an inadequate sample size for this psychometric property (Mokkink et al., 2012). However, a subsequent study of 92 individuals undertook test-retest measurements at 24 hours and 7 days (Armstrong et al., 2014). This demonstrated good congruence between both time intervals and based on the 7-day recall and a sample size of 92, a rating of ‘good’ was made for these criteria. Although there is not a specific time interval advised in the COSMIN rating scale some authors advocate an interval of about two weeks (de Vet et al., 2011). The time interval must be balanced between ensuring respondents do not remember their answers and the stability of the patient’s condition. Measurement error was not specified for any tool.

Interpretability

The COSMIN criterion does not provide ratings for this property; however, the MDASI-BT demonstrated a significant correlation with inpatient and outpatient status and both the MDASI-BT and NFBrSI-24 demonstrated a significant correlation between symptoms and performance status. This provides the ability to assign meaning to the changes through commonly understood clinical connotations. Responsiveness was not reported in any of the reviewed studies.

Generalizability

A further limitation of each of the tools assessed is that they have all been developed and tested only in one country, which may impact their generalizability to other regions. Even though all were developed in English, meanings can have cultural and language variations, therefore cross-cultural validity should be assessed. The MDASI-BT and Brain PCI have been developed for our target group, including all stages of primary brain cancer patients. In their development, the NFbrSI-24 focused on only advanced brain tumours and the SCNS34-BS included a significant proportion of benign tumours.

The MDASI-BT and the NFbrSI-24 demonstrated good psychometric properties while both the Brain PCI and SCNS34-BS are lacking evidence of reliability and validity. Of these the MDASI-BT, as developed for all brain cancer patients, would be most suitable, however additional development would be needed to encompass the holistic aspects of need.

2.4.2 Quality appraisal

The development of patient reported outcome measurement tools requires qualitative methods for item generation and initial development but also quantitative methods for assessing many of the psychometric properties such as reliability or hypothesis testing (Reeve et al., 2013). Literature using qualitative, quantitative or a combination of both methodologies in the same article may be used, which makes interpretation of findings difficult (Caldwell et al., 2011). Many critical appraisal tools such as Critical Appraisal Skills Programme (CASP) focus on only one method such as a randomized controlled trial which makes comparisons between the quality of articles using different methods difficult (CASP UK, 2018). Sirriyeh, Lawton, Gardner, & Armitage (2012) proposed a framework for evaluation of studies with diverse methodologies which allows appraisal and comparison across tools. This framework has demonstrated interrater reliability and therefore was chosen to assist with the critical appraisal of the relevant papers. This framework provides shared criteria for all studies and then specific criteria for qualitative or quantitative designs. There is a rating scale of 0-3 for each criterion (3 is the highest rating) to be assigned for quality with an overall percentage to be calculated. The rating using this criteria can be seen in Appendix 1, however , as the COSMIN criteria was specifically focused developed for measurement tools, this was focused on as the most robust appraisal.

2.4.3 Evaluation of tools for use as a Holistic Needs Assessment tool

The details of the evaluation of the four tools identified were summarised, in relation to their quality and usefulness as a HNA tool (Table 7). The first criterion was to comprehensively represent the common domains of need. The original supportive care needs survey-34 (SCNS-34) was designed using a theoretical framework developed with five constructs of need (Bonevski et al., 2000). Although this was not specifically examined for the additional brain tumour subscale, in combination with SCNS-34, there is representation of the common domains of need (Janda et al., 2008). Similarly, the MDASI-BT structure and design was underpinned by a theoretical framework of individual characteristics of patient burden and symptoms (Armstrong et al., 2006). This tool was designed to assess emotional and physical symptoms with aims of evaluating treatments and planning interventions to alleviate symptoms, therefore the focus is on physical and psychological problems and other domains of need are not covered. The Brain PCI appeared to cover the majority of the relevant domains of need, but the process for comprehensively representing these was not discussed (Rooney et al., 2014). The NFbrSI-24, similar to the MDASI- BT, was developed as a symptom questionnaire rather than a HNA tool therefore the focus was on physical and emotional symptoms.

The SCNS34-BS did not report any details of the user acceptability or how usable this might be in a clinical situation. The NFbrSI-24 and MDASI-BT did not formally assess acceptability, although completion time of the MDASI-BT was noted to take approximately 10 minutes. The Brain PCI did examine user and assessor acceptability and found that despite the presence of cognitive difficulties in many participants, 91% of patients found this questionnaire ‘easy’ or ‘very easy’ to complete. The Brain PCI was rated positively from clinicians with 19/21 feedback forms rating the tool as useful, however 14/21 stated that increased consultation time, although this was not formally assessed (Rooney et al., 2014).

In the context of the evaluation criteria for an HNA tool, the Brain PCI appears most suitable due the assessment of most of the domains of need and the assessment of the usability from the perspective of the patient and clinician.

Table 7 - Holistic Needs Assessment Tool Rating

Measure	Source	Purpose	Dimensions	Psychometric properties	User acceptability (methods and results)	Assessor Acceptability (method and results)	Overall assessment
Brain PCI	(Rooney et al., 2013)	**Strong** - has been developed for needs assessment with only brain cancer patients	**Moderate**- recognised domains covered, facility to add questions around needs. No theoretical or conceptual framework.	**Weak**	**Moderate** - feedback from patients sought on comprehensiveness and ease of completion	**Moderate** - feedback from clinicians sought and felt useful. Interpretation as tick box format straightforward. Verbal reports of increased time but not empirically assessed	This tool has very little psychometric testing. It is the only tool to examine user and assessor acceptability
MDASI-BT	(Armstrong et al., 2005) (Armstrong et al., 2006) (Armstrong et al., 2012) (Armstrong, et al., 2014)	**Weak** - has been developed for primary brain cancer patients but focus on symptoms	**Weak** - Has only focused on psychosocial and physical symptoms. Scaling of intensity and interference. Conceptual framework used	**Strong**	**Weak -** time frame for completion 10 minutes but no user subjective feedback	**Weak**- no supporting data on interpretability or perceived usefulness or ease of use	This tool demonstrates good psychometric testing but focuses on symptoms and omits some important aspects of need. There has been no evaluation of patient or assessor perceptions

NFbrSI-24	(Cella et al., 2003) (Lai, Jensen, et al., 2014)	**Weak** - has been developed for primary brain cancer patients but focus on symptoms and concerns – but has excluded items of concern not related to disease or treatment such as financial concerns	**Weak** - Has only focused on general wellbeing, psychosocial and physical symptoms or concerns. Scaling of intensity	**Strong**	**Weak -** no reported data	**Weak**- no supporting data on interpretability or perceived usefulness or ease of use	This tool demonstrates good psychometric testing but focuses on symptoms and omits some important aspects of need. There has been no evaluation of patient or assessor perceptions
SCNS-34 BS	(Janda et al., 2006) (Janda et al., 2008)	**Strong** - is designed for needs assessment in a brain tumour population	**Moderate**- recognised domains covered and scaling of level of need. No theoretical or conceptual basis	**Weak**	**Weak** - no reported data	**Weak**- no supporting data on interpretability or perceived usefulness or ease of use	This tool demonstrates minimal psychometric testing and good coverage of HNA but has not examined user or assessor characteristics

Brain PCI – Brain Patient Concern Inventory, MDASI – BT – MD Anderson Symptom Inventory and Brain Tumor Module, NFBrSI-24 - 24-item National Comprehensive Cancer Network/Functional Assessment of Cancer Therapy-Brain Symptom Index, SCNS34-BS – Supportive Care Needs Survey (Short Form) with Brain Subscale , HNA – Holistic Needs Assessment

2.5 Discussion

This is the first review of assessment tools developed for brain cancer patients that may be used to assess unmet needs or concerns. This review found four tools which could be considered for HNA, however none had strong psychometric properties, and the two that were developed for HNA had only minimal psychometric testing. Some of the studies that developed these tools were conducted prior to publication of the COSMIN criteria so other specifications may have guided their methodology, however these criteria now present a reliable and valid process to evaluate tools supporting the choice of this criteria. The lack of psychometric testing is not unique to brain cancer and is reflected in other reviews of HNA for generic cancer (Richardson et al., 2007) or other specific cancers such as lung cancer (Maguire et al., 2013). As previously discussed, most authors support the need for psychometric testing of HNA, however the authors who developed the Brain PCI (Rooney et al., 2014) shared the alternative viewpoint presented by Garssen & de Kok, (2008). They assert that the priority for research on HNA tools should not be the development of the psychometric properties but a focus on the feasibility of usage of screening tools in clinical practice and the effects of decisions made by the health care providers, ultimately focused on the outcomes for patients.

While it might be questioned whether tools that are developed as ‘symptom questionnaires’ such as the MDASI-BT or the NFbrSI-24 were appropriate to consider, they were included for a number of reasons. There were a lack of PROMs which facilitated the patient’s identification of concern in brain cancer. In addition, the use of a validated symptom scale in combination with targeted problem checklist focused on the other domains of need is an approach that has been adopted for generic cancer needs assessment in Canada (Canadian Partnership Against Cancer, 2012; Watanabe et al., 2012).

User acceptability for both the patient and clinician is a key component when developing questionnaires or assessment tools for clinical use. The ideal instrument should assess the perceived burden, usefulness and meaning in relation to improving the existing strategies for detection of unmet need. User acceptability and burden is important for all PROMs, but as previously highlighted, in brain cancer this is particularly important as completion can be impacted by the neurocognitive impairments. In clinical trials for brain cancer patients, QOL form completion can be poor, with either no form completed or missing items (Dirven et al., 2014; Walker et al., 2003). A recent study in Germany with patients diagnosed with glioma highlighted that even with support in clinics up to 10% of participants erroneously completed the SCNS-34 and this rises to 20% without support (Renovanz et al., 2016). They highlighted structure and comprehension as contributing factors but also noted that if patients were distressed more errors occurred. There is also data that suggests that if clinicians find the questionnaires cognitively demanding, burdensome or not clinically relevant, their support of implementation and response to any PROM could be sub-optimal (Gilbert et al., 2015). There clearly needs to be a balance between ensuring the relevant problems of a neuro-oncology population are assessed, while minimising burden.

The findings suggest two potential options that could be considered for HNA in Brain cancer. The Brain PCI, out with its psychometric properties, demonstrated moderate to strong characteristics in relation to HNA quality criteria and with additional psychometric testing may offer one approach. Alternatively, the MDASI – BT demonstrated the strongest psychometric properties and could provide the basis for an alternative approach. This tool could be combined with a more holistic assessment, similar to the approach the Canadian Partnership Against Cancer (2012) that uses the Edmonton Symptom Assessment System together with the Canadian Problem Checklist. However, based on the results of this review, that although progress has been made, no tool provides a comprehensive approach in identifying needs without further development.

2.5.1 Recommendations for further research on brain tumour HNA

Recently, an international multidisciplinary working group has been set up to evaluate and provide guidance on the use of PROs in neuro-oncology (Dirven et al., 2018). This may provide valuable information on the use of HNA or provide direction on what other PROMs could generate high quality evidence to help evaluate the impact of HNA processes in future research.

In addition, to the areas already discussed, there are many other aspects of HNA in brain cancer which would benefit from research. The use of electronic HNAs are increasingly used and may provide a basis for improving compliance and providing this information to a variety of health care providers. Electronic PROMs are acceptable to patients and have the potential to provide a variety of modes (e.g., internet based, hand held devices) and could be personalised based on patient preferences or capabilities (Gilbert et al., 2015). However, the IT systems to collect this data in a meaningful, accessible, and secure ways need to be developed and tested.

There is also a need to look at studies comparing different pathways for HNA and the impact of this process. For example, where is it completed, at what point in the treatment trajectory and which health or social care provider receives and reviews it. Most importantly, research should focus on the impact of HNA in improving outcomes such as reducing distress or increasing wellbeing.

2.5.2 Strengths and Limitations

This was a rigorously conducted review of tools that may support HNA in brain cancer. This review has some limitations. The diversity and quality of methodological approaches was challenging when comparing tools. There were also differences in the aims of tools and although they all aimed to measure some aspects of needs or problems, some may have not been designed specifically to undertake a holistic assessment.. Despite this limitation, their inclusion was useful, as if an existing tool was to be adapted – these could be considered. In addition, meta-analysis was not possible due to the diversity of tools and subject heterogeneity.

Input of an extra investigator would have been beneficial in undertaking the quality appraisal with the COSMIN criteria and evaluation for use as an HNA tool as the use of two independent raters would have given greater assurance of the reliability of these assessments. However, although not formally undertaken, the extensive discussions with the supervisory team helped to ensure that each rating was justified.

As mentioned, the COSMIN criteria have the drawback of only evaluating measurement properties and therefore the evaluation of the tools as an HNA tool was undertaken. This formulation and use of a rating scale were piloted for the first time in this study and there are no measures of the reliability of this. However, undertaking an assessment of the tools which is focused on their appropriateness for use, could be recommended.

In relation to the review of available tools there was a clear gap in the assessment of content validity. The development of the Brain PCI (Rooney et al., 2014) had a lack of input from patients, with recommended methods for establishing content validity such as patient interviews not undertaken. The other HNA tool had undertaken a very limited and insufficient assessment of content validity (Janda et al., 2006) and this is a necessary component (Terwee et al., 2018). There were also clear deficits with all tools on the patient and assessors' acceptability ratings – while the Brain PCI obtained a score of moderate all other tools were weak. In a population like brain tumour patients with high rates of non-completion of PROs (Dirven et al., 2014), this can help ensure that tools are fit for purpose and usable in practice or research. This important aspect was examined in more detail in Chapter 3.

2.6 Conclusion

Providing supportive care and meeting the needs of patients with brain cancer who in many cases have a poor prognosis, is challenging. HNA has been identified as an important strategy to facilitate this process. This review has provided a comprehensive overview of the content and measurement properties of four tools that could be used for HNA in brain cancer. Similar to other reviews of HNA tools in cancer, this review identified a variety of tools for assessing needs, however there is currently a lack of evidence to support what might be the best tool or even consensus on how to evaluate this in a clinical setting (Higginson et al., 2007; Richardson et al., 2007; Wen & Gustafson, 2004). It is clear, due to the lack of a clearly suitable tool in this area, that the evidence base to develop this area of supportive care is limited. Ideally a fit-for-purpose, psychometrically robust, and context-specific tool should be developed specifically for brain cancer patients, to be used in everyday practice to allow for meaningful communication to identify supportive care needs. The lack of a tool, which adequately meets these requirements, supports the need to further explore how HNA can be performed in brain cancer patients to optimize this intervention.

2.7 Summary and reflection on next steps of the thesis

This chapter has reported the literature review which was undertaken to identify and evaluate potential tools which could be used to support HNA in brain tumour patients. This review identified four tools – two that were developed as an HNA and two that were developed as symptom assessment tools which could be further developed for a holistic assessment. This followed a systematic approach to identify all available sources and then undertook a robust evaluation of the psychometric properties and utility as an HNA tool. Where there might be options to take this forward, this could only be through further developmental studies was needed. This lack of a suitable tool has implications, as in the absence of this, there may be suboptimal assessment of needs to better deliver supportive care. This review is now published paper can be seen in Appendix 2

This leads to a consideration of the next step of the thesis as I sought to consider these findings but also the other significant evidence gaps in the knowledge surrounding HNA in brain tumour patients. The systematic review highlighted significant gaps in seeking patient views about the acceptability, usefulness and feasibility of different tool designs. Also, earlier scoping reviews revealed no studies that explored how brain tumour patients' needs are currently being met and how they perceive an HNA might improve this. There were also some questions raised about the effectiveness of HNA in other cancers where reviews of interventions had been undertaken.

In a systematic review of interventions to reduce unmet needs as measured by HNA, none demonstrated a statistically significant reduction (Carey et al., 2012). These authors also noted that it appeared that HNA without subsequent interventions to address needs might be detrimental. Some of the possible reasons for this were suggested such as a lack of sensitivity of HNA tools, lack of effective interventions, heterogeneous samples, samples too small, interventions not administered as intended or that the population have needs which cannot be feasibly met within a health care system.

At this point, there was consideration that a fit-for-purpose, psychometrically robust, and context-specific, tool should be developed specifically for brain tumour patients, which can be used in everyday practice to allow for meaningful communication to identify supportive care needs. However, even within the tools identified in my systematic review, some had no patient involvement in development, or others did not look at acceptability or feasibility. It was important therefore to seek patients’ views on HNA tools, but also examine if they perceived that this intervention had some potential to improve outcomes. As there was very limited data on acceptability, usefulness or feasibility there was benefit of seeking patient views what tool might work best for HNA – an exploration of the views and experiences of brain cancer patients and their carers was proposed. To explore this a pilot study was proposed to help guide the next phase of research in developing HNA for brain tumour patients.

The next phase of the thesis was therefore designed to answer the following questions through a qualitative study:

- What are the unmet supportive care needs of patients in all stages of their brain tumour?
- How do patient and carers think a HNA should be undertaken?
- How do patient perceive this would improve their unmet needs?
- How do patients and carers think a HNA tool should be designed to identify patients’ unmet needs to health care professionals?

3 Chapter 3 - Assessing and supporting unmet needs; Exploring the experiences and perceptions of those affected by a Brain Tumours (Study 2)

3.1 Introduction

When planning and designing this primary empirical research study it was important to consider the relevant questions which could be addressed in relation to the implementation of HNA with brain tumour patients. Many obstacles exist to assessing patient needs including: a lack of clarity on the best tool and processes in initially identifying needs (Renovanz et al., 2016); what interventions are effective when needs are identified; and how to evaluate their effectiveness (Carey et al., 2012). While studies have examined these aspects in other cancers, there is no research amongst brain tumour patients which is essential given the unique neurocognitive impact and is a significant omission in the research (Renovanz et al., 2016).

There has been some research conducted on the use of PROMs in oncology in clinical practice to improve patient centred care and outcomes for patients. This was able to demonstrate certain patient benefits, such as increased discussion of emotional issues and symptoms, but also detailed explicit areas which could enable or impede successful implementation of PROMs (Howell, Molloyet al., 2015). There were many enablers such as having relevance including disease specific questions and simplicity, as the degree of disability could present significant challenges. To address these difficulties, it has been suggested that the involvement of the ‘patient voice’ in development and implementation of any patient centred programme has the potential to develop insights which could impact successful and meaningful improvement to patient outcomes (Vandermause et al., 2017).

The COSMIN criteria (Mokkink et al., 2010), as well as a number of other authors, clearly advocate involvement of patients in the design of any assessment tool questionnaire (Abernethy et al., 2010; McDowell, 2006; Reeve et al., 2013; Turner, et al., 2007). Of the tools that have been developed and reported here in Chapter 2, there was no individual tool that could be recommended for HNA. Moreover, there was a variation in the involvement of patients and carers in the design of the tools with one of these, the Brain PCI, having no involvement of patients and carers in development (Rooney et al., 2014). All the tools were developed within a distinct region or country (and health system) consequently having some influence on needs, for example financial needs could be more prominent if universal healthcare is not available (Armstrong et al., 2012b; Janda et al., 2006; Lai, Jensen, et al., 2014; Rooney et al., 2014). While several tools did have a limited satisfaction or some ease of use questions in their exploration, there was lack of questions which addressed detailed insight into patient views about how HNA could improve or impact the assessment of needs in improving outcomes for these patients.

While PROMs are increasingly viewed as an important component of both care and the evaluation of treatments, previously research has demonstrated that PROMs in this group are often not completed (Renovanz, Henchtner, et al., 2018). This study demonstrated significant motor dysfunction and poorer clinical condition were predictors of non-completion of PROMs, yet in light of their significant problems, these patients may have a greater need of assessment and support. Therefore, additional exploration was warranted, to consider the views of patients with brain tumours on their perceptions of HNA tools in the identification of unmet needs. Specifically, due to the unique neuro-cognitive deficits that could influence this feasibility or utility of these tools. These included exploring issues around; the length and complexity of the scale, relevance of the HNA to address brain tumour issues, and phases of the cancer journey which would be beneficial. In addition, administration issues including modes of completion, (electronic versus paper formats), place of completion (home versus clinics) and if assistance is needed, who and how might this be best be delivered (Howell, Molloy et al., 2015).

To allow HNA to be as widely accessible as possible for brain tumour patients, there might be a role for proxy completion by family members. Similar to other studies of patients with a PBT (Langbecker & Yates, 2016), it was expected that some patients might experience physical, cognitive or neuropsychiatric symptoms which could impede their capacity to participate in some aspects of the study. Patients were invited to also ask a caregiver or family member to join them in their participation. There is some evidence to suggest family members or significant others are reliable proxies for symptom assessments (Armstrong et al., 2012a) and QOL questionnaires (Giesinger et al., 2009), thus acceptability and feasibility of this should be considered in future research for this patient group.

Chapter 2 highlighted the lack of patient involvement in some tools, and this was an important area to explore. Furthermore, there have not been any studies that invited brain tumour patients to consider what existing design of tools would be feasible and useful for them to help in an HNA. Therefore, this study was designed to address this gap.

This chapter will first provide a description and justification of the research methodology used. This will be followed by the results, and this section will present the four themes which emerged from this study. Each theme will be discussed in turn, and this will include discussion and an illustration of how that theme contributed to a conceptual model of need in brain tumour patients. These findings will then be considered within policy and the wider evidence base for HNA in brain tumour, which will provide the basis for the next stage of this thesis.

3.2 Research Aims

This research was an exploration amongst brain tumour patients and their carers of their experiences and perceptions of unmet needs, how these have been supported (or not) and how strategies such as HNA might support this.

The study focused on patients (and their carers) who have been diagnosed with a brain tumour and explored their experiences of communicating needs, processes which supported or hindered the meeting of these needs and perceptions of HNAs and tools which support this process. The specific aims of this study were:

- To explore if patients and carers perceive they have unmet needs and if so what are their experiences of these. This includes the range and nature of concerns/unmet needs they would like to discuss in consultations.

- To explore the perceptions and experiences of patients and carers of communication and support in response to their needs with health care professionals and the approaches used in resolving problems.
- To consider how patients and carers perceive advised strategies such as HNA tools, other assessments or interventions contribute to the identification and support or resolution of unmet needs.

3.3 Research Design and Justification

This section will give an overview of the research design which provided a framework for the collection and analysis of the data. It had the best fit for the research question to permit the questions to be examined through an appropriate lens. The two broad research approaches are quantitative and qualitative, and the choice of approach was guided by the question. A quantitative or positivist paradigm seeks to control and measure data to provide reliable and replicable data (Denscombe, 2014). Alternatively, a qualitative approach seeks to study phenomena to make sense of it and describe or interpret this (Denscombe, 2014). As the primary aim in this study was to explore experiences and perceptions of patients and carers affected by a brain tumour, with a focus on their needs, how they are met and strategies that might help in the identification support or resolution of unmet needs, a qualitative research design was considered most appropriate to address the aim and objectives.

There are several different approaches within qualitative research which are underpinned by distinct traditions such as phenomenological traditions, grounded theory, ethnography or thematic analysis (Parahoo, 2014). Thematic analysis was selected for this study as it is not tied to specific disciplinary traditions or epistemological assumptions and hence was pragmatic in addressing the wide current study aims and research questions (Nowell et al., 2017). Furthermore, It is widely and successfully used in health research and has been used previously to explore unmet or care needs in cancer (Muntlin et al., 2018; Weaver et al., 2020). As a result of the literature review and gaps in the literature on HNA in brain tumours the following questions were established.

- Do patients and their carers have unmet needs? And if so, what are their experiences of unmet needs and concerns through their disease trajectory?
- What are the nature and range of concerns that brain tumour patients would like to discuss in consultations?
- What are the patients and carers experiences and perceptions of communication and support of their concerns or needs with health care professionals or others supporting their care and their approach in resolving problems?
- What are patients and carers perceptions of how advised strategies such as HNA tools, other strategies or interventions have contributed to the identification and support or resolution of unmet needs?

3.3.1 Patient and public involvement and input

Patient and public involvement (PPI) in research is valuable as it allows those on whom the research may impact, to influence the research agenda. It can cover a variety of activity which can involve consultation, collaboration or user-led research (Pii et al., 2019). Research does not often report or include PPI, yet a systematic review examining PPI found that it is feasible and valuable to involve patients in research even when the survival rate is low and that researchers should seek to involve patients, as the perception that they are too vulnerable is often misplaced (Pii et al., 2019).

PPI in this research was sought in the form of consultation into the aspects of design and relevance of current HNAs for those affected by brain tumours. PPI input was considered vital in this research to ensure the design and methods maximised inclusivity as much as possible and to allow those with a variety of neurocognitive deficits to participate.

Two support workers from Brainstrust, who have also been carers for patients affected by brain tumours, reviewed the proposed research agenda and commented on the suitability and feasibility of the questions. They also assisted with the recruitment through their support network. Their input influenced the inclusion of carers and the addition of interviews to support those who had higher levels of impairment. They confirmed the research questions were important and relevant for those who were affected by brain tumours, but also supported the importance of the inclusion of carers both for support, but also as their perspectives were likely to be valuable in providing information on unmet needs and strategies to might address these. They also reviewed the information sheets and topic guides to comment on the format, and understandability of these documents for a brain tumour patient and their carers. No changes on the language or content were required, however some suggestions in the conduct of the focus groups such as the location were utilised. It was advocated to hold focus groups in a central location (with good public transport links) and not within the health care institution (as parking was challenging and some participants found this a reminder of a traumatic events related to surgery and treatment). Although there were was no significant changes needed other than the areas mentioned above, this helped ensure the research was relevant, inclusive and appropriate for the participants.

3.4 Methods

This section will give an overview of the methods for each step of this research study. A justification of the chosen methods is provided to support the rationale for the process undertaken.

3.4.1 Sampling strategy

The sampling strategy was based on a number of factors that were influenced by both methodological considerations and underpinning practical issues with conducting research with this patient group. Practical issues are often based around convenience, cost, and time (Bryman, 2016) and in this research design and data collection technique, this also had to be considered. Unlike the focus on representative sampling in quantitative research, qualitative research does not claim to be generalisable, but rather seeks purposive participants who have experiences and views that may shed new insight in relation to exploring the objectives of the study (Parahoo, 2014).

Consideration of overall numbers in a sample can often be evolving in qualitative research but ethics approval committees will often require the exact details of the numbers of participants. This can be a challenge and may require updating the committee if numbers or sample methods change. Sample size in qualitative research should seek to be large enough to provide a textured understanding of meaning but due to the vast amounts of data that can be generated through interviews and focus groups, be manageable for the size of project (Fugard & Potts, 2015). Initially up to five focus groups (with a maximum number of 10) were planned for, however the numbers may have been decreased if data saturation was reached earlier or extended if data saturation not achieved. However as described in section 3.4.4 adjustments to the sample were required to reach data saturation and this was approved by the ethical review committees.

3.4.2 Ethical issues

The study was conducted in compliance with the protocol, Research Governance Framework for Health and Community Care (Scottish Executive Health Department, 2006) as well as the relevant UK and Scottish regulatory requirements. This study sought and was granted ethical approval from both Edinburgh Napier University, Faculty of Life, Health and Social Sciences Ethics Research Committee and South-East Scotland Research Ethics Committee. This was followed by site specific approval at each participating Health Board. All protocol amendments were submitted to all relevant committees detailed above prior to implementation. Copies of the approval letter for the study and then the amendment expanding the research to include interviews are included in Appendix 8.

One area of ethical concern was if it was possible that discussing client unmet needs might distress the participants. Prior to the focus groups and interviews participants were advised they had the option of not answering any question and to the right to withdraw at any time, to minimize distress. It was emphasized that participants only needed to discuss areas that they felt comfortable sharing. In addition, the details of appropriate support personnel such as the relevant clinical nurse specialist or support groups were given if needed. Several participants did become emotional during the interviews, which will be discussed further in the results and in all cases referral to additional support was offered. A few participants, however, did report that it was the first time that anyone had asked in such detail about their experience and participants highlighted this as a supportive interaction to talk about their experiences and how they were feeling.

An additional area of ethical concern was that within this group of patients there was the potential for significant cognitive impairment, which could have affected capacity for consent and participation. The existing care team and support workers were aware that only those that were competent for consent should be approached for participation in the research. If there were any concerns in relation to capacity to consent, potential participants would not have been recruited for the study.

3.4.3 Sample

Patients who received a diagnosed with a brain tumour were selected for this study. Inclusion criteria were that they needed to have an ability to understand English and be of at least 18 years of age. Participants also needed to be able to provide consent and not have a level of cognitive impairment which would prevent understanding during the focus group discussion or individual interviews. Inpatients were also excluded as this study was focused on support of needs when not in hospital or another inpatient setting. Carers or family members were invited to attend if they were at least 18 years of age and willing and able to consent. Carers or family members were invited to support patients who might have additional support needs to facilitate communication.

3.4.4 Sampling procedure

Participants were initially recruited for focus groups from an outpatient neuro-oncology clinic in Scotland, or through a patient support group in covering these regions after initial referral from their care team or support worker. After the first year of recruitment, a second neuro-oncology clinic was added with an option for interviews added due to poor recruitment to the focus groups. In the study setting, consecutive eligible patients were approached at their clinical visit. The patients from the support group were a sample of those that were willing and able to attend a focus group after an invite email was sent by their support worker. A purposive sampling strategy seeking patients with different stages of brain tumour, social circumstances and disability was used provide the maximum variation in their potential perceptions and experiences. Carers of patients who were taking part in the study were invited to attend as a pragmatic decision was taken that often support of the carer is needed. In addition, as highlighted through the PPI, the carers may add a different perspective.

3.4.5 Recruitment Process

Recruitment took place via two separate mechanisms. For the focus groups, participants were recruited from the one cancer centre and a brain tumour support group and for the individual interviews, participants were referred to the researcher from their cancer team at the two cancer centres. After an initial period of recruitment, the recruitment strategy was expanded from only focus groups to include interviews and a second cancer centre as approximately 90% of patients were unable to participate or refused to participate in focus groups at a set time. Although patients did not need to state a reason for refusal, the set time and place planned for focus group were a barrier for many due to challenges with mobility or travel, while others had concerns about speaking in a group (either due to privacy or cognitive challenges). Therefore, semi-structured interviews were added as a data collection technique after the completion of the second focus group in July 2015. This ensured that patients who experienced these challenges would not be excluded from this research.

At the cancer centres, if potential participants were willing to discuss the study further, they were referred at the end of their routine clinic appointment to the researcher to discuss the study at clinic. If the potential participant was recruited from the brain tumour support group their details were forwarded by the support group worker to the researcher to then contact them. At that time, a verbal explanation was given, and their eligibility checked and confirmed. The written information and consent form were given or sent to potential participants with a provisional schedule for focus groups or interview. In all cases if the patient wanted to have their carer or significant other involved, they were also given the information sheet and, if agreeable, all informed consent processes were followed, except informing their consultant or GP.

The two final approved patient and separate carer information sheets and consent forms can be seen in Appendix 3 for patients and Appendix 4 for carers. They contained slightly different information based on whether the participant was a patient or carer, and in addition in line with NHS ethics guidance at the time of ethics approval, GPs were informed of participation in the case of patients.

After a period of at least 24 hours potential participants were contacted again to confirm if they were still willing to participate, after any questions were asked details (place, time and date) of the focus group or interview were confirmed. After consent, the patient’s consultant and GP were informed of their participation in this study by letter in line with NHS guidance (see Appendix 7).

3.4.6 Procedures and data collection

This was a qualitative exploratory study that used focus groups, individual interviews and interview dyads to explore unmet needs of brain tumour patients. The rationale and processes for this will be discussed in the following paragraphs.

Focus groups are widely used data collection methods in qualitative research which allow participants to discuss key areas. Barbour, (2018) highlights the benefits of this approach for health services researcher in many areas, including exploring why some services are working or not working as intended or alternatively to provide insight into how services could be developed. The interchanges between participants such as discussion and debate can provide valuable data examining these aspects of care. Therefore, in the context of this thesis, focus groups provided a data collection technique that would provide understanding into how needs are met or not within existing care pathways and how HNA if introduced might be used.

Focus groups have been useful to explore specific outcome measures are perceived in routine clinical practice. An example of this is a study which explored perceptions of outcome measures (PROMs and clinician completed questionnaires) recommended for use in clinical care with parents or carers who cared for a child supported by Child and Adolescent Mental Health Services (Moran et al., 2012). Focus groups were seen a beneficial to develop the understanding of the role of these measures which was not familiar to the participants, but also provide valuable data on their perceptions of the tools including how each of the tools should be used and administered. The ensuing discussion and debate about the role allowed the participants to learn and refine their opinions together – while providing valuable insight into how these should be implemented and used in routine practice. As HNA was not routinely used in brain tumour patients at the time of this research, this was likely to be the same and this shared understanding could develop.

However, as previously mentioned, semi-structured interviews were added as pragmatic decision to offer participants a choice of method. Semi-structured interviews are very widely used in qualitative research and entail asking participants to elaborate on a set of questions and are particularly beneficial to explore experiences (Holloway & Galvin, 2017). In this research one of the benefits of this approach was its flexibility for patients with cognitive impairments, as it allowed the freedom for participants to explain their thoughts and ideas at a pace appropriate to them and take more time to articulate their thoughts if needed. It also provided a better platform for clarification than focus groups and this approach could have helped develop a more in-depth understanding of some of the aims such as their experiences.

This addition of interviews also allowed the pragmatic decision to allow carers to also be part of the interviews. However, this approach has some clear benefits in that there could be more in-depth exploration of responses, there was also an interactional element that provided some of the benefits of focus groups (Morgan et al., 2016) such as discussion how HNA might be implemented.

These methods were adopted for this study to address the aims of the research. Pragmatic decisions were made as detailed above to offer participants a choice of method. However, the use of three different data collection modes, interview, interview dyads and focus groups will have supported a broader understanding of the areas under study (Carter et al., 2014). Focus groups may allow participants to build or debate ideas while interviews will have allowed participants to more fully share their experiences.

The discussions were guided using the focus group topic guide or interview schedule (see Appendix 5). The content of the topic guide and interview schedule was informed by the research aims and research literature on unmet needs and HNA in cancer. It was then reviewed by a patient support worker in relation the comprehension, content and acceptability prior to finalisation. There were no changes suggested and they agreed this was suitable for the research.

The following areas were explored with patients and their carers:

- What are participants (patients and carers) potential concerns or needs during outpatient medical consultations? What is the importance of different concerns from a patient perspective?
- What are the participants experience and perceptions of the communication of concern or needs in consultations?
- What are the patients and carers experience and perceptions of communication and support of their concerns or needs with health care professionals or others supporting their care and their approach in resolving problems?

- Individual advantages or disadvantages of using an existing tool for needs assessment prior to consultations. (This question was aided by providing the participants with examples of tools.)
- Explore timing in a treatment trajectory at which an HNA would be most useful?

Prior to the end of the focus group, or interview, participants reviewed paper copies of four needs or symptom assessment tools which have previously been developed for brain tumour patients and identified in Chapter 2 (Armstrong et al., 2006; Janda et al., 2006; Lai, Jensen, et al., 2014; Rooney et al., 2014). The discussion of these tools focused on the participants' perceptions of these tools including potential benefit, content, length, general layout, or any other general comments that could inform the important areas to gain the patients' point of view.

Nine interviews and two focus groups were conducted in total. The focus groups were held in a private meeting room in a public building and the interviews were held in the homes of the patients. The focus groups lasted from 70-100 minutes with one held January 2015 and the other in July 2015 and the individual interviews were all held in September 2015 and lasted from 16-128 minutes. The length of the interviews or focus groups were determined by the participants and their exploration and disclosure of the question and topics.

Two focus groups were completed with seven participants in the first focus group (four carers and three patients) – there was one carer who participated without the patient as the patient decided not to consent at the start of the focus group, but the carer wished to stay. This group was recruited from both the support group and neuro oncology clinics. In the second focus group there were two carers and two patients, and this group was recruited entirely from the support group. In addition, there were nine interviews, and these participants were recruited entirely from the neuro oncology clinics. The participant characteristics and data collection method can be seen in Table 8 below.

Table 8 - Summary of participants and data collection method

Focus group or interview	Female	Male	Diagnosis of the patient	Age range of patient	Relationship status
Focus Group 1	Patient 1		High grade glioma (Grade IV)	50-59	Married
		Carer 1			Married to patient 1
		Patient 2	Low grade Glioma (grade II)	70-79	Married
	Carer 2				Married to patient 2
		Patient 3	High grade glioma (Grade III)	60-69	Married
	Carer 3				Married to patient 3
	Carer 4*				Mother to withdrawn patient
Focus group 2	Patient 1		Meningioma	50-59	Single
	Carer 1				Mother of patient 1
	Patient 2		Glioma	50-59	Married
	Carer 2**				Daughter (daughter of non-attending patient)
Interview 1	NA	Patient -	Astrocytoma	40-49	Married
Interview 2 (dyad)	Patient	Carer	Glioma (Grade IV)	60-69	Married
Interview 3	NA	Patient -	Glioma (Grade III)	60-69	Married

Interview 4 (dyad)	Patient	Carer	Astrocytoma	40-49	Married
Interview 5 (dyad)	Carer	Patient -	Glioma (Grade III)	60-69	Married
Interview 6	NA	Patient	Glioma (Grade III)	50-59	Divorced
Interview 7	Patient	NA	Glioma recurrent	40-49	Single
Interview 8 (dyad)	Carer	Patient	Glioma (Grade IV)	70-79	Married
Interview 9	NA	Patient	Glioma (Grade II)	50-59	Married

* Note focus group 1, patient 4 withdrew consent.

** focus group 2, Carer 2 came without a patient.

Initially focus group discussions were planned however after three months of recruitment, 40 patients had been approached through clinics, eight had agreed but only two patients (and their two carers) could attend the focus group times as the times needed to be rescheduled due to poor recruitment. The support group reported 48 email invites were sent to potential participants. Of these five individuals participated in the focus groups (one additional patient attended but did not consent). In total, approximately 88 individuals were approached, and 11 participated. Although it was planned carers would only attend with a patient, they participated without a patient in two cases. In one case this was due to the patient, not wanting to sign the consent and in the second, one carer who attended the second focus group without the patient. While this had not initially been planned, carers had valuable insights, so they were included in the study.

Therefore, semi-structured interviews were offered pragmatically to ensure that participants who felt unable to participate in focus groups (due to practical or personal concerns) were able to take part in the study. For the interviews: 14 patients were approached and nine agreed and all of these were recruited from neuro-oncology clinics. In total, there were 13 participants for interviews, including four carers and nine patients. The reason for participation or non-participation was not sought (if not volunteered) in line with the participant consent form that stated no reason was required. However, for the focus groups transport difficulties were mentioned by six patients and participating in group discussions by three patients.

3.4.7 Data Analysis and Data Handling

The focus groups and interviews were audio-recorded and transcribed verbatim, in keeping with qualitative research. After transcribing the recordings, all identifiable data were removed from the transcriptions. NVivo software (version 10) (QRS International, 2012) was used to store and organise the data. The focus groups were analysed using the six-stage framework for thematic analysis described by Braun & Clarke (2006). This approach aims to understand the underlying ideas, assumptions, conceptualisations, and ideologies through a process described in Table 9.

The first phase involved the familiarisation with the data. Through transcribing the initial transcripts and listening to them on multiple occasions ideas started to emerge. The gap between the first focus group and second allowed a longer period of reflection to consider the issues that were emerging. Notes to explore concepts were kept throughout this process, such as considering the significance of certain interactions. An additional example was one participant who gave a full description of her life before her diagnosis, and then highlighted all the changes. This seemed to be a process that illustrated changes and adjustments she had undergone. The notes reflected that this was not just to outline the loss of areas of her life, but also to demonstrate adjustment and positivity. Mind maps were also used to support how concepts might link together and this started the early consideration of themes or subthemes. An example of one of the mind maps can be seen in Appendix 9 demonstrating how the theme of ‘altered self’ was put together.

The second phase then involved generating initial codes. Initial coding was done through NVivo (QRS International, 2012) however after this it was also supported by manual analysis. This was done by printing out collated word documents of all quotes for a code, then re-reading and considering the meaning of the code. In some instances, the name of the code was revised if it did not represent the meaning conveyed across the sample of quotes.

Phase 3 of thematic analysis required a consideration of the themes and subthemes. This process followed the coding, but this part of the process involved often going back to the initial quotes to check and recheck that the theme captured the essence of the data set. A brief representation of the consideration of one quote in relation to codes and themes is presented in Appendix 10. At this stage, a number of themes and subthemes were proposed from the existing coded data. The names of these themes were revised on multiple occasions in consultation with the supervisory team. The author (JA) explained the rationale for this choice and then different possible interpretations were discussed (Phase 4 and 5). These discussions helped to support the final title of themes. These were checked again against the original transcripts, codes and subthemes. Subsequently, a selection of compelling exemplar quotes which support the themes and subthemes were selected that represented the data set. The report of the analysis of the themes is presented in the next section with the interpretation of the quotes (Phase 6).

The process and details of data analysis help to provide rigour and credibility for this study (Parahoo, 2014). The use of exemplar quotes is presented to support how the themes were developed and synthesised.

Table 9 - Phases of Thematic Analysis (Braun & Clarke, 2006)

	Phases	Description of analysis process
1	Familiarising self with data	• Narrative preparation and (re) reading data over and over for familiarisation – noting initial preliminary ideas
2	Generating initial codes	• Coding of data systematically with the support of NVivo in line with the research aims

		- Collating data relevant to each code and reviewing data within transcripts to help verify meanings
3	Searching for themes	- Collating data into potential themes and sub themes - Gathering and reviewing data for each potential theme
4	Reviewing themes	- Checking if themes work in relation to the coded extracts - Checking if the themes work in relation to the entire data set - Reviewing the transcript and initial coding to search for additional themes
5	Defining and naming themes	- Ongoing analysis to refine the specifics of each theme and describing each theme and subtheme to avoid overlap - Two researchers reviewed the themes and examined the data to verify the themes. Different interpretations were discussed and when agree the researchers defined and named the themes.
6	Producing the report	- Selection of compelling exemplar quotes which support the themes - Final interpretation of the extracts and relating the analysis back to the research questions

3.4.8 Establishing trustworthiness

Trustworthiness has been suggested by (Guba & Lincoln, 1989) as one of the important key methods of convincing others of the legitimacy of qualitative research. They have suggested that credibility, transferability, dependability, and confirmability are criteria that should be considered, as a parallel to validity and reliability in quantitative research.

Credibility is the consideration of whether the interpretation of the data is appropriate and truthful in presenting the findings (Mills et al., 2012). To help demonstrate credibility, several strategies were undertaken. Research triangulation through the interrogation and review of the coding, themes and interpretation was supported by the supervisory team. The preliminary findings and interpretations were continually checked against the raw data and exemplar quotes were chosen to support the interpretation, which also included listening to the recording again to consider them in the context in which they were discussed. This was particularly useful for the focus groups as it helped to interpret agreement or discord with some of the individual viewpoints. Finally, the findings were discussed with the representatives from the brain support charity at various points and as carers of brain tumour patients they provided a forum to discuss the interpretation of some aspects of the data and consider alternative views.

Transferability of data is concerned with how these findings can be applied to other studies or contexts. While qualitative studies do not seek to have external validity in the same way as quantitative research, the ability to use the data elsewhere is a key aim of any research. Credibility as described in the previous paragraph helps to achieve this. Another element is the transparency of the research and the description and illustration of the research process in this chapter have helped to achieve this. In addition, representation of the diversity of the population the study represents is important (Coghlan & Brydon-Miller, 2014). Through expanding the recruitment strategy which supported those with higher degrees of disability this was achieved.

Confirmability is focused on ensuring the participants contributions are appropriately interpreted by the researcher (Given, 2012). To achieve this, during the focus groups and interviews the researcher sought clarity of meaning for some of the participants discussion. In addition, as a researcher, throughout the data collection and analysis process reflective notes were kept minimising researcher bias. Another method that can help with this to ask the participants to review the analysis and consider whether this adequately represents their view (Given, 2012). Although this was not undertaken, as this could have been challenging for individuals with cognitive impairments. However, the reviews from the brain tumour support group representatives and supervisory team helped to minimise the impact any researcher bias which was undetected by the researcher.

The last concept to be considered is dependability. The aim of this aspect of trustworthiness would be that if another researcher undertook the study in the same context that there could be similar results (Given, 2012). This was achieved through ensuring the research process was logical and documentation was in place to support each step (Nowell et al., 2017). These elements of trustworthiness were embedded to help establish the acceptability and usefulness of the results described below.

3.5 Results

In summary, there were two focus groups and nine interviews in total. Focus group one had three patients and four carers. Focus group two had two patients and two carers. There were five individual interviews and four joint dyad interviews of the patient and their main carer or significant other. The total number of participants included in analysis was 24. This next section will present the findings that emerged after completing the thematic analysis aligned to Braun & Clarke, (2006). These all contributed to the combined analysis and the results presented below.

Thematic analysis helped frame the codes, into subthemes and finally the themes into four areas represented:

1. ‘Altered self’ – impacts of a brain tumour
2. Impacts of others - Responses and Actions to unmet needs
3. Impacts of patients and carers - Perceptions of coping, self-management or distress
4. The role of HNA tools and strategies

Quotes are used to illustrate the themes and subthemes and to protect anonymity the number of the focus group or interview and patient or carer number (if applicable) were used and these can be viewed in Table 8. As each theme is discussed its contribution to a conceptual model of need in Brain Tumours is described and depicted. This is continued through each theme with the final *Conceptual Model of Needs in Brain Tumour Patients* presented in Figure 8.

3.5.1 Theme 1:'Altered self' - Impacts of a Brain Tumour

This theme encompassed the impacts of the diagnosis and treatment and the alterations in the participant's life. This was influenced by a perception that for some patients that they were not the 'same' person as before their diagnosis. This 'altered self' was relevant for the patient, but also pertinent to the carers in terms of the changes within their role, responsibilities and their relationship as a consequence of that diagnosis. This theme encompassed four subthemes, neurocognitive changes, psychological distress, perceived 'loss of self' and changed patient and carer relationship. The first subtheme of neurocognitive changes was often the source that was the precipitating factor for the areas discussed in the other three subthemes.

Neurocognitive changes

All patients reported neuro-cognitive problems. These included loss of hearing, mobility, memory, concentration, and seizures all of which have changed the daily activities they could do such as employment, hobbies or activities of daily living. One participant in his 40s described some of the changes he experienced.

> *'my speech was so bad, it was like I had a stammer all the time, couldn't get words out. And I couldn't remember my pin number (for his bank card) … the words and numbers, everything was jumbled. They said it would be a short-term thing, it came back but I still have problems. I can't spell, even simple words ... and have to read things over and over to understand them. Int 1/Pt*

This was echoed by others who highlighted significant problems with simple reading and spelling when fatigued for example:

> *'I've still got problems every now and again if I'm tired, with my speech and I can't spell words. A, a simple word I really have problems trying to focus to see what word it is, you know what I mean?' Int9/Pt*

Many also highlighted the distress this caused, and this linked with the constraints on other areas of their life due to these changes. For example, as a diagnosis of epilepsy (caused by the brain tumour) will result in the loss of a driver license in the United Kingdom. The participant from interview 1 had to change career due to loss of licence associated with seizures and for another participant this loss had a profound impact on their ability to leave their own home and contributed to their isolation. This was emphasised below in the 'Bungalow' and the powerful image of the 'caged lion':

> *'[my doctor] said I would lose my driver's license for a year and a year to me is 365 days. In a bungalow – I was like a caged lion because I couldn't get anywhere.' Int6/Pt*

The neurocognitive changes also impacted on the carers in terms of a compensatory role, doing things for the brain tumour patient that they would have previously done themselves. For example, one of the brain tumour patients indicated his wife would take over discussions, as finding the right words and speaking were now problematic.

> *'Socially with my speaking I sort of go in the background … when my speaking is not as fluent, I sort of give a wee (small) signal to [my wife] and she takes over the conversation.' Int 9/Pt*

Another patient and her mother described the impact of the changes on their lives due to the weakness and mobility problems and the dependence this brings.

> *Patient 1: 'I can't move in the morning … Mum brings my breakfast and my pills.'*

> *Carer 1: ‘I come down (with her breakfast) and I sometimes think, some days I might want to lie in, but I know she needs these and will fall over if I don’t bring them.’ FG2*

There appeared to be an openness of discussion between this carer and patient of the impacts and changes on the mother’s life, but during the focus group they often seemed to use humour to help deal with these impacts making jokes about some aspects, such as their descriptions of the patient’s falls. For example, the mother at one point said no matter what is in place, the daughter will find a way to fall. The humour did seem to contribute to a positivity and closeness in this relationship which seemed to help them both cope with the impact of the brain tumour. However, in many instances this seemed to contribute to distress for the patient and carer which are highlighted in the next subtheme.

Psychological distress

Carers and patients both experienced significant emotional impacts such as feelings of isolation, anxiety, depression and feelings of distress. One patient who was currently diagnosed with depression after a recurrence of his brain tumour emphasised the emotional impact he was still experiencing. Although his treatment was completed and he was back working, many of the people he knew thought he was back to normal, but he did not feel back to normal.

> *‘[People] think because you have had a brain tumour everything should go back to normal after and it doesn't quite work like that – because it is a mental thing, if you have a broken leg everyone sees and makes allowances’ Int1/Pt.*

Some of the patient and the carers shared their experience that their emotional wellbeing was often not asked about or the services for psychological support were quite limited. One patient was felt her medical care and physical needs were well supported but psychological support was limited.

> ‘*…it’s like there’s a hospital and there the medical side of it, but it probably not sufficient for the psychological help that you really need, like I did.’ FG2*/Pt 1

This patient drew attention to some of the challenges that they were adjusting to such as the loss of work, hobbies (due to loss of sight) and dependency on her parents as challenging areas post diagnosis. However, she highlighted the role of friends and family in this as support that did help her psychologically. Carers also described a lack of emotional support from professionals. For example, a carer was distressed and crying throughout much of the interview when discussing her experiences of caring for someone with brain tumour and their own unmet emotional needs which had been overlooked by health professionals.

> *'I think emotional is one of the things …that's a big one, sometimes they don't…and sometimes…it's not addressed. Well, I presume they don't have time for us (crying).' Int8/Carer*

This carer highlighted that no one spoke to her directly except when he was getting diagnosed and she felt very isolated and felt there could be more communication with her. She said although she recognised her husband was the one with a diagnosis, no one had considered how she was coping and perceived that she was not a priority for HCPs. She spoke about being 'terrified' about caring for him when discharged after surgery and said if only she had someone to phone to ask about things, it would have helped. Other carers also highlighted the need for responsive and timely advice to help them manage the care as not knowing what to do to manage or improve symptoms caused distress.

Loss of self

The loss of self was associated with the perception of the earlier person prior to diagnosis crumbling away along with some of the positive and valued aspects of this self-perception by patients themselves. There were many descriptions of loss which related to socialisation roles, hobbies and independence and several of the male participants in particular highlighted how loss of employment had changed their perception of themselves. A woman in her early 50s who was now wheelchair bound and blind described her loss and sense of identity below through the multiple losses of different aspects of herself until all that was left was someone with hospital appointments.

> *'… because you were losing a bit of yourself, losing your identity, I wasn't a teacher anymore, and I had lost my sight so I couldn't read books, I was bored out of mind, and I couldn't sing, I couldn't get access anymore (it was not accessible without a lift), I couldn't do Sunday school, my diary was empty except for hospital appointments. I found that really strange' FG2/Pt1*

However, the patient and her mother started to elaborate on how they overcame some of the challenges simply through purchasing a magnifier for reading.

> *'And she got a [a magnifier] with a bit of a light on it … I was conscious of wanting to try and address all your issues…' FG2/C1*

Her mother, who was also her carer, expressed that she felt a need to help solve the problems, and both the mother and daughter were able to provide examples of how they attempted to improve the situation in small ways to allow certain activities to be undertaken such as hobbies like reading. However, they also highlighted how other areas such as financial issues, or continued employment could not be easily solved.

Changed patient and carer relationship.

Psychological distress was evident to both carers and patients, however there was also a significant emotional and practical burden to carers that both patients and carers were aware of. Carers in many cases, had financial, and family obligations which in one case contributed to a relationship breakdown, for example:

> *'I still hold her in adoration as a saint because she kept a roof over our head on a part-time salary and managed to feed us as well. But there was one day, she was filling in some DWP [Department of Work and Pensions] forms, and one of them was disability living allowance, and I walked in, and she'd been crying. I said, what's wrong;*
>
> *[she said] 'it just mentioned when you're expected to die'….well, that was breaking my heart. I mean she…she's a star, but that's us divorced.' Int6/Pt*

The patient had clear admiration for the role that their spouse undertook, but they seemed to link the change of roles and emotional impact of this to their marriage breakdown. Although we cannot know in this instance if this was the cause, there was clearly a practical and emotional burden for this carer that was precipitated by the brain tumour diagnosis.

In a subsequent interview with a carer and patient, they were both aware of the change of role with the carer taking on ‘everything’. In the excerpt below the patient and carer discuss the practical tasks, but at other times during the interview discussed the care needs such as urinary catheter care, dressing, bathing and almost all personal care needs for his spouse.

> *Carer: ‘Someone at our local church said, ‘how are you doing … as you will be taking the brunt of it’. Well, it certainly is a change for me’*
>
> *Patient: ‘Aye, cause you’re doing everything now aren’t you’*
>
> *Carer: ‘Yes but my thoughts are on [patient’s name] all the time’*
>
> *Patient: ‘He washes the clothes, irons them, cooks, buys food, everything’*
>
> *Carer: ‘I start at 9 am and finish at 6 pm and that’s now my day.’ Int 2*

Personality changes in the person with brain tumour also seemed to impact some relationships. However, interestingly not all patients seemed find these changes distressing, one patient who was interviewed on his own was discussing some of the neurocognitive changes and how these were perceived differently by himself and his wife:

> *Personality changes, I feel I am OK but [my wife] thinks I’m not the same person, personally, I feel OK… I look at things differently now and in that way I feel happy’ Int9/Pt*

This patient was interviewed alone without their wife present, therefore the impacts of this on her were not explored from her perspective, however, would be likely to involve some adjustments with a degree of challenge for the family member. As patients were present in all instances, this might have been a challenging area to discuss, and hints of the challenge came through with one carer becoming frustrated with the patient when they would not turn off the television for the interview saying.

‘Look what I have to deal with now [indicating husband]’ Int8/Carer

The emotional impacts of isolation for this carer were discussed under the subtheme of psychological distress, and this change in their husband seemed to also contribute to this.

These neurocognitive changes were linked with the loss of some aspect of their physical function, emotional, social or practical functions. Interestingly, these were often not clearly identified by the patient as an ‘unmet’ need when the participants were questioned about this during the study – but rather a symptom that they needed to cope with themselves, however distressing this might be for the patient or carer. In most, but not all cases, the patient and carers seemed to view the symptom as unchangeable and had to live with it but in many cases, this did seem to lead to distress. The next paragraphs will describe how this theme contributed to a conceptual model of Needs in Brain tumour patients.

3.5.1.1 Conceptual Model of Needs in Brain Tumour Patients (phase 1 – Altered self)

The theme of 'altered self' was one of the most significant aspects of this research, because it highlighted the difference in managing this illness compared to other cancers largely due to the neurocognitive impacts or symptoms. This alteration was seen in individuals' view of themselves with their internal perception of how things have changed, but additionally how they thought they were perceived altered in the view of the 'world'. This change in the individual was also experienced by the carers. However, it was experienced somewhat differently to the patient, with some loss of the dynamics of a relationship prior to diagnosis, increased responsibility and decision making by the carer, or in some cases the management of behavioural changes which had emerged. Also, the patient’s personality may have changed, and these changes in 'losing' the relationship of 'before' caused distress and other impacts on their wellbeing. In Figure 5 below, we see the start of the development of a conceptual model examining the theme of altered self and neurological impacts on unmet needs. Subsequent themes will be added to the model to provide an overall representation of the key themes and how these integrate into this conceptual model.

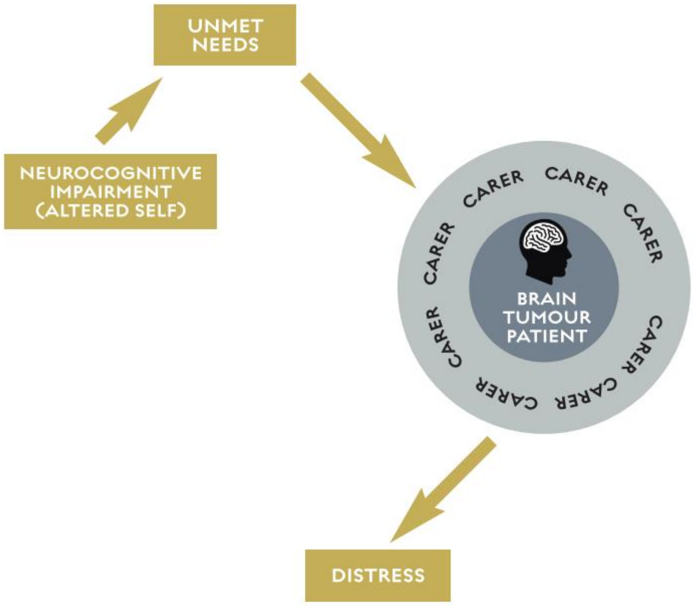


Figure 5 - Model of Needs in Brain tumour patients (phase 1 – Altered self)

The above figure depicts a conceptual model of how an ‘altered self’ impacts patients with a brain tumour and their carers. The first yellow box represents the significant and detrimental impact of neurocognitive needs or deficits which often are a causative factor for other unmet needs. For example, some psychological needs like depression are linked with increased neurocognitive impairments. The needs are central to the patient, which we see represented by the first inner circle. The carer encompasses the patient, as many of the unmet needs also impact them in their caregiving role, however they also may have additional individual needs that have emerged as a result of this role. For example, as a carer they take on additional responsibility such as decision making or managing financial issues. These unmet needs often may result in the outcome of distress for both the patient and carer represented by the final yellow box. This helps to illustrate how unmet needs might link to distress and highlights the unique impact of neuro-cognitive impairment as a causal factor.

Unmet needs can be variable and from many of the holistic domains – but for brain tumour patients the 'altered self' is the source of the most burdensome concerns and problems. This change in 'self' can undermine an individual's confidence in how they can manage many aspects of their life as they know their limitations in cognitive functioning. Memory loss, loss of executive function, challenges with reading or using technology can often be the manifestation of this for the patient. Assessment that detects their unmet needs or most burdensome problems such as HNA has the potential to improve wellbeing through appropriate interventions, referral or measures to support self-management. Through these measures that are undertaken to address there is the potential to decrease distress. The next theme explored how some of the responses and action of others were perceived or experienced by the patients and carers.

3.5.2 Theme 2 — Impacts of others - Responses and Actions to unmet needs.

It was clear these impacts of the disease in many cases could be modified – particularly through actions or responses which is represented by the theme ‘Impacts of others’. This theme observed how others could influence the outcomes for the patients, in relation to decreasing distress or meeting patient or carers needs. When referring to ‘others’ this was represented by formal care providers or services. There were two main subthemes within this theme; *Healthcare professionals – the role of specialist knowledge and trust* and *Focus on patient’s agenda and needs*.

Healthcare professionals – the role of specialist knowledge and trust

Almost all patients and carers commented on challenges in relation to the quality or knowledge of care outside of the special neuro or neuro-oncology providers. Access to specialist knowledge and trust in the practitioner’s knowledge was highly valued compared with other health practitioners such as GPs or other non-specialist services. This was a discussion in relation to managing their seizures in relation to epilepsy after they had completed their primary treatment.

> *'I think it's so specialist… I don't think the GPs are really up to speed, some of the finer points of this kind of thing. [They] suggested to take a medicine which I was already taking, that obviously wasn't a good suggestion, then the medicine that I got that didn't do anything, either, and I finally, the senior consultant at the [cancer centre] said this is what we need, the expertise was there.' Int3/Pt*

Another patient also reported that they felt that their relationship with their GP had changed – and this could be linked to not knowing how to manage the brain tumour and that this made her GP avoid her. This exchange from focus group 1 was part of a discussion of the support that patients get from their primary care team and their GP.

> *Patient 1: 'I was disappointed with her [her GP]'*
>
> *Carer 1: 'Oh aye, shocking'.*
>
> *Patient 1: 'Kind of, or that she'd missed out. So, she felt like she'd sort of, she just felt… …a wee bit uneasy because she thought she maybe missed something… But she didn't even ask how we were on the phone, you know'…*
>
> *Carer 1: 'Just, I think she was just that shocked that she was just stunned …she felt guilty a wee bit, you know.'*
>
> *Carer 3: 'Doctors are human too'.*
>
> *Carer 1: 'And it was because she had never come across it before and she was just totally gutted'. FG2*

This patient expressed disappointment – and both the patient and carer felt this was due to her the embarrassment of the GP due to the delayed diagnostic period. Delayed diagnosis or suboptimal management could result in a change in the relationship with other health care providers -this could be due to a lack of confidence or mistrust from the patient – although understanding for this reaction from other was sought – such as acceptance that mistakes are made.

In both focus groups, individuals highlighted a perception that general practitioners (GPs) may not have specialist knowledge. While this could have implications for the relationship between the patient and/or carer and the GP such as lack of trust in their knowledge, there was an acknowledgement of the challenges with managing care for such a rare disease.

> *'My GP said one in his career usually is what they get... that's why they know so little about it. Because they're not coming across it very often.' FG2/Pt2*

Many commented on how they avoided seeing their local doctor as they felt they couldn't manage their neurocognitive side effects. However, for others, compassion and support, from the local primary care team provided reassurance and support.

> *'Our own doctor…he's good. He's been round a couple of times to see us, changing steroids, asking how things are. So, he's there; I know if I seriously need something I would call [him] and he's very good. He'd be able to arrange something for us.' Int2/C*

Almost all participants seemed to value the provision of knowledge or support from the specialist neuro-oncology team. In particular, the roles of specialist nurses or therapeutic radiographers were mentioned by many patients as having a very important role in support. One of the carers highlighted this which was then agreed with most of the others in the group.

> *Carer 2: 'The specialist nurses are the backbone of the health service.… I know they are not always appreciated within the nursing hierarchy and what they do for the patients and they are absolutely critical. They hold the whole service together…and the patients depend on them.'*
>
> *Patient 2: 'they said to contact them any time and I have done…they are lovely.' FG1*

It was detailed that the ability to call them for advice 'anytime' was particularly valued, but it also seems that their interaction was viewed positively with the comment 'lovely' being echoed by two other participants – which may indicate that the patients perceived compassion or care from these professionals. The importance of having a knowledgeable healthcare professional to call when problems arose was valued by those that seemed to have this resource, while others while some participants who did not have this in place, voiced it was needed.

> *'I found it really hard … we had left umpteen messages and we were told we needed to wean him off the pills [steroids] and there were all these pills and didn't know what to do. We panicked but the district nurse didn't really know what do either.' FG2/C2*

The care of individuals with a brain tumour and their significant others is challenging due to the rarity and the complexity, particularly managing neurocognitive symptoms. Lack of continuity of care and communication between healthcare professionals was seen as problematic particularly between the specialist providers and GPs. The data from this study suggests in some instances that patients might have benefited from additional specialist management or advice and the care and support strategy should take this into consideration. In some instances, there also could be a lack of understanding of how and who could be providing support, from the patients and carers. In other cases, psychological support referral pathways may need to be defined.

Focus on patient's agenda and needs.

Participants shared that they were grappling with new disabilities which caused a multitude of different problems including practical concerns, such as how to take a bath or filling in forms related to their brain tumour. A number of participants felt that their multifaceted needs were not currently considered.

> *'When somebody's had all their operations and leaves hospital [they have] all the practical things they suddenly have to cope with …The biggest thing I found was I find it very difficult to remember things …that person isn't the same as from when they went… So, there's a lot of practical things that they could do to help, sort of like, build a big package for the person' Int8/C*

For some participants, it felt like consultations had a set agenda from the HCPs rather than their concerns for some participants. One of the carers highlighted their frustration of trying to get support or referral for weight gain after long-term steroids and the focus on medical issues.

> *‘I find it frustrating that those kinds of things tend to get minimised – it all about the medication or the scan… their focus might be very different from what your focus is. We have tried to bring it up but no one really wants to know… whether it’s because they don't think they can do anything or they don't want to try, I don't know.’ Int4/C*

Healthcare professionals who ‘really listened’ or helped seek solutions were viewed very positively. However, a few highlighted negative experiences after raising an issue with a HCP that then seemed ignored. Participants speculated that the healthcare professional did not have time for them or that the maybe it wasn’t their remit to help with that and this was viewed negatively.

In a few instances care or support seemed to be rejected. In one instance, social care workers who were providing care were cancelled due to the disruption to ‘normal’ life which did not fit with the patient’s daily schedule, even though this presented considerable challenges to the carer.

> *‘They were coming at quarter to seven at night – saying ‘we will get you into your night clothes and put you to bed.’ I don’t go to bed then. … And in the morning, I told them not come as I needed to be dressed and up before then and [my husband] can do all that.’ Int2/P*

Although her carer (husband) agreed formal carers were no longer needed, he found caring tiring and that he was unsupported.

> *‘Nobody’s been here for me. Nobody… British stiff upper lip; you have just got to get on with it.’ Int 2/C*

The delivery of personalised care to support brain tumour patients is not always achieved. Actions, or the lack of them and responses to these were clearly identified as helping or hindering patients and carers wellbeing. In this next section the impact of others builds on the conceptual model of Needs in Brain tumour patients and help to demonstrate the influence of this theme.

3.5.2.1 Conceptual Model of Needs in Brain Tumour Patients (phase 2 – Impacts of Others)

For many of the patients and carers, the knowledge and support of their specialist neuro-oncology teams were highly valued, and several reported that care delivered out with this team was less than ideal. These opinions were often prompted by a perceived delay in diagnosis or mismanagement of side effects once treatments started. Previous interaction and experiences have a considerable impact on trust in the knowledge and advice in that health care provider. Subsequently, lack of trust had the potential to result in avoidance of care from providers that were no longer perceived as trustworthy.

It also became clear that many patients felt their clinical interactions were determined by what the clinician wanted to talk about or do, rather than with their concerns, and they perceived that there was not a holistic view of care. In other instances, carers felt that they were not included or heeded during consultations which they found distressing as they too had needs or suggestions for care that were not acknowledged. Such an approach created barriers with patients and families seeking support elsewhere or from each other rather than from their specialist team. This demonstrates the importance of the focus on the patient and carer's agenda. Through the provision of informed care needs and information, a therapeutic trusting relationship develops. Alternatively, if care is received, which has been less than optimal, the patient and carer may no longer regard advice or indeed accept or seek care from this provider which has implications for longer term care. For instance, after primary treatment has been completed, there could be challenges to shifting any care provision and support to the community if advice and information are not perceived trustworthy. Nonetheless, such a rare and complex condition does pose challenges for those in general care to provide that level of knowledge. This also raises important considerations around communication between care providers and this issue will be considered in section 3.5.4 in examining the role of HNA.

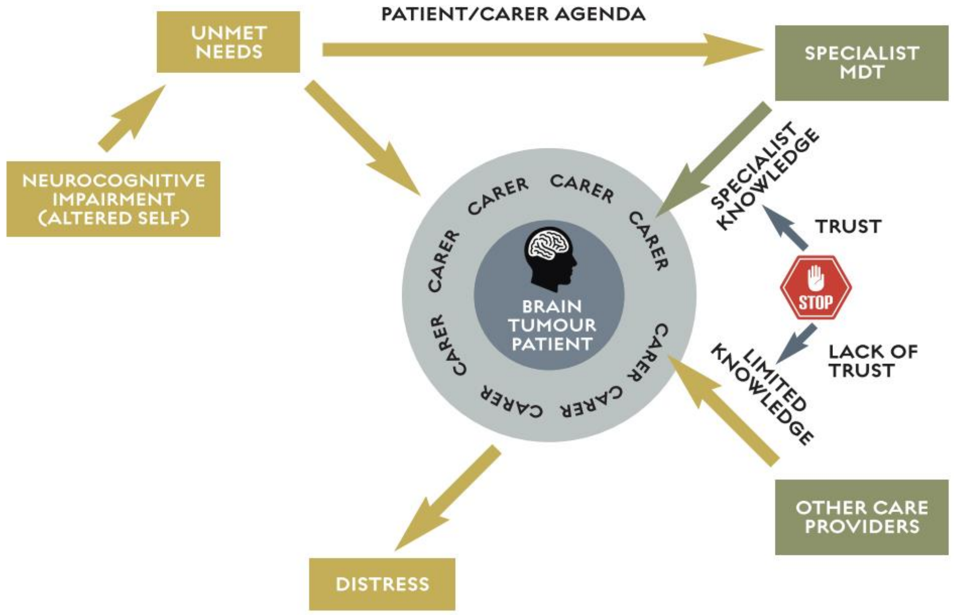


Figure 6 - Model of Needs in Brain tumour patients (Phase 2 – Impact of Others)

The findings in this theme enabled building on the model of need to illustrate how others could help to reduce unmet needs and distress. Or alternatively, in other cases, why the actions (or inaction) of healthcare professionals may not help. The first addition to the model is represented by the dark yellow arrow which represents the importance of identifying the patients and carers focus and agenda to the healthcare professionals. This includes needs, but also requires healthcare professionals to have good communication skills to ensure that the voice of both the patient and carer can contribute to planning care. With a focus on this agenda and the patient's critical problems, knowledge from the specialist can help mitigate unmet needs. Thus, the model now emphasises imparting of knowledge, appropriate referral and listening to the patient can help to develop trust. The model illustrates how channelling this to the patients and carers can help support their needs based on their agenda. However, in other cases, we can see that with providers that might not have the specialist expertise or the knowledge of the patient/carer agenda, trust might not be developed or could even be lost if in previous encounters, some care was perceived by the patient as inappropriate. This is shown as the red stop sign blocking the development of trust (Figure 6). This lack of trust in non-specialist providers, such as primary care, may result in gaps in care as support for unmet needs is not sought.

3.5.3 Theme 3 – Impacts of patients and carers - Perceptions of coping, self-management.

This theme focused on the role of the patients and their family or carers in managing their needs or coping with the multifaceted impacts resulting from a brain tumour. This theme looked at how patients and carers strategies in coping or managing could influence the outcomes for the patients, in relation to maximising QOL and decreasing distress. While the previous theme focused on external variables, this theme focused on the individual or collective responses of the patient and/or carer. There were two main subthemes within this theme; *Effective strategies in coping* and *Seeking self-management* which helped to explore this theme.

Effective strategies in coping

A positive outlook or ability, in spite of disability, was commonly referred to and was seen as an important element to wellbeing. Often after a period of adjustment to being not quite the same some patients remained positive and happy.

> *‘I feel okay. I've still, I've still got my, disabilities with my speech, but I must admit, I feel okay that way. And I never have sadness. My outlook, now, is that I've had that operation, I could have been worse. And every day, I look at things differently, now.’ Int9/Pt*

Patients and carers reported a variety of sources of support, both formal and informal which impacted on their wellbeing. Most of these interactions were seen as positive, however, some participants noted that some from friends or significant others drifted away, and in some cases, this was down to discomfort with the change the patient or feeling unable to cope.

> *'He just disappeared, they literally couldn't cope and we never saw them again. He literally just disappeared and never came back … but I think he couldn't cope with it'. FG2/Pt1*

Despite the loss of previous networks such as work or some friends were no longer there, but often other networks were sought such as support groups and two patients mentioned the support from their church as excellent. Family members were mentioned most often and the majority of participants had very limited support outside of their family unit.

> *'I think we have sort of really stayed as family unit through this and are able to get the support we need from within'. Int5/Carer*

However, despite this, as explored theme 1, there may be an excessive emotional or practical burden that contributes to psychological distress so seeking ways to minimise this is important. Coping by the patients and carers were often accompanied by the actions that sought to manage the impacts of brain tumour.

Seeking self-management

Some respondents seemed to manage without additional support outside of routine appointments with the ethos that 'we take care of ourselves'. Other respondents reported that they had refused offers of additional support. One of the reasons for a refusal for support was that they see themselves as independent and not in need of support. In one interview the carer was describing their response to being offered a Macmillan nurse

> *'We refused didn't we, because we didn't think it was necessary … I feel we are kind of independent and we wouldnae be needing services. We would waste them when someone else was needing them.'Int8/Carer*

The reason for refusal was that they felt independent. However, this same carer highlighted having no one to call to discuss caring for her husband and the isolation she felt as a carer. This might suggest that there is a lack of understanding of the role of some professionals, and that there is a perception that unless there is a need for physical care, that nursing support is not justified and helping clarify support pathways for those caring for support and advice may be needed.

However, others also spoke of their frustration and distress at being unable to assess advice for distressing symptoms caused by the tumour such as loss of vision. This was challenging for the carer, as it was hard to interpret what a change in vision might mean and knowing if this was something that needed urgent intervention or not.

> *'I would like someone on the end of the phone … it runs itself the NHS within its own time constraints … we a have a need now but we can't doing anything' FG2/Carer 1*

However, this same carer also highlighted the lack of knowledge outside of the specialist team and the contact they would prefer to have is someone with specialist neuro-oncology knowledge. A few of the carers highlighted the need for help to manage at home some symptoms, knowing what actions were appropriate, but also to provide some emotional support for them to discuss the challenges with someone expert.

3.5.3.1 Conceptual Model of Needs in Brain Tumour Patients (phase 3 – Impacts of patients and Carers)

Individuals and their significant others had to respond to meet their needs. Several responses included an adjustment to a new normal and seeking alternative ways of navigating their lives. However, for those who had adjusted well, unmet needs were viewed as a problem they had to overcome independently, and in many cases, they felt they had some control over this. This resulted in patient and carer developing solutions to their unmet needs and self-managing some aspects of their care. For example, even with significant physical symptoms such as the loss of sight, they came up with a working solution or with emotional impacts, feeling they had the resources and confidence to deal with this. These problem-solving approaches and perception of self-management were viewed positively by the participants, and possibly mitigated some of the psychological impact. However, in other cases, individuals reported the distress that a lack of engagement from their healthcare team limited abilities to advance their wellbeing. For example, one participant was very concerned about weight gain (due to long term steroid use) and wanted dietary and exercise advice, however, was told there were no resources for this.

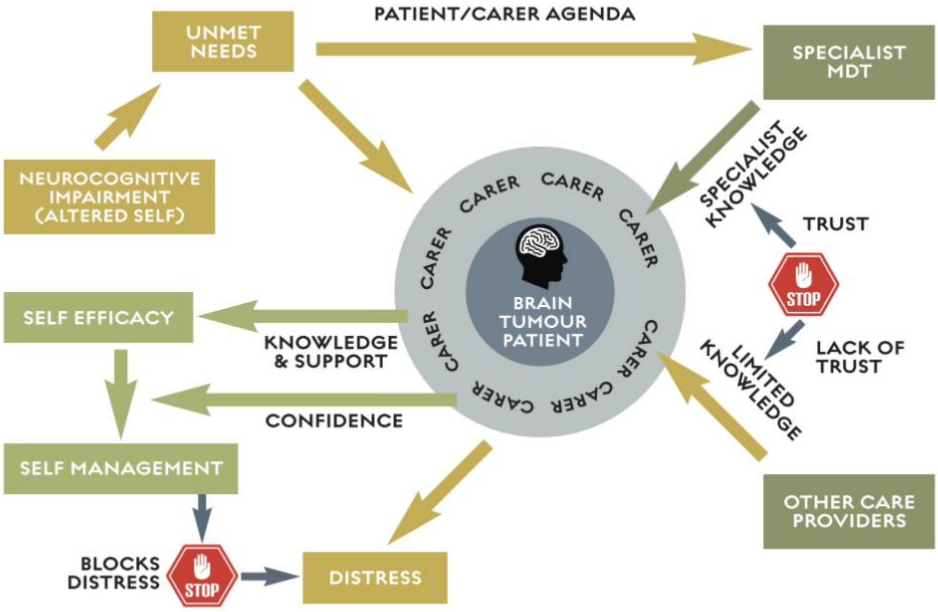


Figure 7 - Model of Needs in Brain tumour patients (Phase 3 – Impacts of patient and carers)

Figure 7 continues the development of the conceptual model and builds on the previous model by depicting the influence of family and carers on reducing their own unmet needs and highlights both the actions needed to do this and the outcomes that may develop. The first addition in this Figure is the green arrow which represents how appropriate knowledge and support delivered from HCPs to the patients and carers can help them manage the impacts of brain tumours. The term ‘appropriate’ implies at a level participant could understand, linking with capability for decision-making and undertaking care that is focused on the individualised knowledge they need as outlined by their agenda. If delivered in this way by the HCP, this has the potential to support the development of self-efficacy depicted in the green box which in turn leads to self-management actions by the carer and patient. In some instances, self-management which links with a sense of control over some aspects of a situation, can block or mitigate distress. So, by adopting this process of supporting and information sharing at an appropriate level the detrimental psychological outcomes for those affected by a brain tumour might be decreased.

3.5.4 Theme 4 - The role of HNA tools and strategies

This theme was informed by the perceptions of participants on potential brain tumour specific questionnaires or PROMs. In the latter part of both the interviews and focus groups, copies of four tools which could be used as part of a HNA, reviewed in Chapter 2, were distributed for the participants to provide their feedback and perceptions about the use of these. The feedback about these tools were informed by three subthemes; *Value of HNA, Barriers or Challenges of HNA tools* and *Implementation considerations*.

Value of HNA

All patients and carers universally agreed HNA was useful. The majority of patients included comments that suggested that these tools can give a chance to ask about concerns and remind them what they want to say. Another important benefit of these tools seemed to be providing information on what might be expected with a brain tumour and possibly even legitimising some of the areas that individuals may experience problems with and providing a platform for discussion of these.

> *'The only thing I did mention to [my wife] the other day; they asked me if I had any symptoms I said I wish I knew what kind of symptoms I was actually looking for – nobody's ever told me what symptoms to look for' [comment when looking at the Brain PCI] Int2/Pt.*
>
> *'I think [HNA] have their place, don't they? Yes, it certainly brings out things in your life that have changed' Int2/C*

Many patients also supported that it was positive that the questionnaires were focused on brain tumours.

> *'A person who has had an operation for a brain tumour has made out these lists as they are asking the right questions' FG2/C1*

There were numerous discussions of individuals going through the questionnaires discussing the problems, symptoms or unmet needs that were reflected on the forms. While they were not specifically asked to do this, many participants undertook filling in the forms and having discussion about what they had experienced as a result of their brain tumour.

A number of participants highlighted that it could improve information to other HCPs such as GPs and keep them in the loop if this was shared with them.

> *'I think it would be good for my GP to see this – they don't really know what is going on' Int5/Pt.*
>
> *This would be good…. I think the GP and, and oncology and all that, should be singing from the same sheet like. With this I know they will in a way, FG1/Pt3.*

This seemed to be viewed as way of bridging some of the knowledge gap between different care providers.

Barriers or Challenges of HNA tools

Despite the positive aspects of a brain specific HNA, the majority of patients highlighted that they would have challenges with completion with some or all of the tools. This was due to a wide variety of neuro cognitive problems such as reading, concentration, and difficulty with holding pens, or visual problems.

Scaling seemed to introduce complexity which many found challenging. Some of the tools presented had a variety of scaling methods including Likert and numerical rating scales and both were identified as problematic in defining the magnitude of need.

> *'You don't know what the scale is from one to five. What does that mean? I mean how am I to know whether……'some need'. That's actually very unclear' Int3/Pt*
>
> *'the absolute numbers are actually quite hard to fill in – because what is a 10 as opposed to a 5?' Int4/C*
>
> *'To me your just as well ticking a box – because you can't be specific for all it looks like you can be, where one being the worst and the best. You have to find a spot in between – I can't stand them'. Int8/C*

Some participants commented that it was hard to know what the different Likert levels meant, and others found just seeing a line or numbers harder to interpret. One participant also highlighted annoyance at questionnaires, and this may be a factor in lack of the completion of questionnaires like this.

An interesting perspective that came out from a few participants who felt that this might reveal too many problems for health care staff to deal with.

> *'Because you don't go on about it doesn't mean you don't have it. A lot of people will think, the doctor won't support me in this, He will say you never reported this before – but I could probably tick almost all of them'. FG2/Pt2*

In this focus group there was widespread agreement with this statement. This seemed to indicate that some patients do not volunteer information on the holistic impacts of the disease as they do not think that support may be possible due to the medical focus of consultations. In addition, this could indicate that there are so many life changing needs that patients have to be selective in what they mention as support for all aspects would not be possible. However, this also may support the important role a systematic HNA can fulfil in providing a comprehensive assessment in revealing the unmet needs.

Implementation Considerations

There were diverse responses on where to complete the tool (at home or in waiting rooms), the mode of administration (paper based, apps or tablet-based responses). While some participants felt completion of this in the waiting room before consultations would be the best option for them. Others highlighted that this would be stressful and potentially cause anxiety.

> *'To be honest if I got this in clinic, we are too anxious there, I would just tear it up – but it would be good to send out before' Int8/C*

This seemed to be linked to situations where their responses to treatment or prognostic information might be discussed after a scan or other investigation. This could indicate that HNA might need to be a specific focus or targeted for a time not related to these appointments.

Many of the patients highlighted practical challenges with completion due to coordination problems, concentration or impaired vision.

> *'I think it's the brain … what you're dealing with, forms and too many pages, you just switch off … I couldn't be completing it' FG1/Pt1*

During these discussions it was explored if electronic versions such as a touch screen on a tablet device or computer, but here seemed to a lack of consensus on this, with a few patients reporting neither would be possible for them.

> *Challenges with completion ‘I would not be able to do it (electronically)…my hand is quite volatile and trying to coordinate where I should touch the screen… it doesn't work’. Int4/Pt*
>
> *‘I can’t hold a pen – a touch screen might be better.’ Int9/Pt.*

While there was lack of consensus on where, when or how these should be completed, the agreed benefits from the participants would support implementing this for patients. It may be that flexibility around implementation using a patient centred approach for options for completion might allow as many patients as possible to have access to this assessment.

3.5.4.1 Conceptual Model of Needs in Brain Tumour Patients (Phase 4 – The role of HNA tools and strategies)

The final part of this study involved participants assessing the benefits, drawbacks and feasibility of using HNA in clinical practice through a review of the four tools identified in Chapter 2. As previously reported, there are challenges with HNA completion, but in supporting a holistic assessment, several benefits were identified. These included giving permission to bring up issues and problems – and focus discussions on their needs. A few also highlighted how this HNA tool could be of benefit in helping their primary care team or general practitioner know what was 'going on' with their brain tumour.

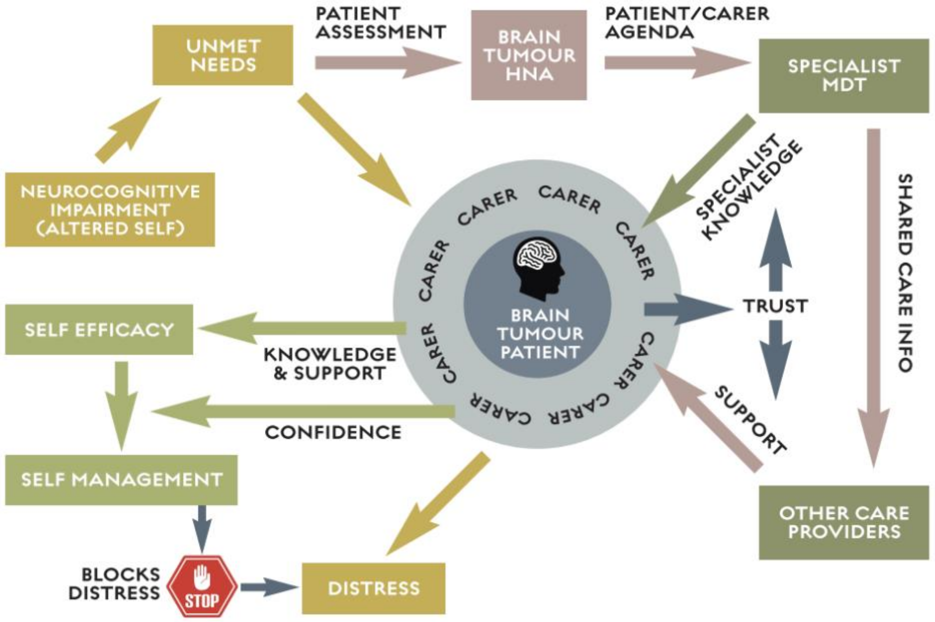


Figure 8 - Model of Needs in Brain tumour patients (Phase 4 – The role of HNA tools and strategies)

The final stage in the development of the conceptual model is represented above in Figure 8 and depicts how inclusion of HNA programmes can help improve the actions and responses to unmet needs by those affected by a brain tumour. The first light pink arrow in the diagram represents the inclusion of a brain specific HNA used as part of an assessment process. This will improve holistic assessment and support the identification of the needs most relevant to the patient and care. While this is useful for the specialist healthcare providers in terms of planning care and support, this knowledge can also be shared with other providers as represented by the light pink arrow demonstrating the shared information between specialist and other care providers. This sharing of information with providers such as those in primary care can include needs as identified by the HNA, but also the plan of care so support of the patients and carers is shared appropriately where possible between care providers. These steps also will help ensure that the knowledge and support delivered to the carer and patient are more representative of their needs thereby improving self-management to a greater degree and reducing distress.

This last addition to the conceptual model is important as there were clear gaps identified in the delivery of patient-centred holistic care when professionals do not accurately assess symptoms or do not ask about other needs such as psychological or practical needs. While patients and carers can be quite accepting of consultations being primarily focused on the healthcare professionals' agenda or perceptions of the patient needs, it often leads to unnecessary suffering and distress that could be relieved in some cases if a more patient centred agenda was undertaken. There are also meaningful opportunities to exploit and improve the information sharing to primary care, allowing them to provide appropriate support. The challenges for primary care in dealing with clients with rare tumours with complex side effects are clear. Within the UK Recovery Package, there is a very defined pathway for sharing End of Treatment Summaries with primary care and then for primary care to undertake a Cancer Care Review to discuss their needs and support self-management (Macmillan Cancer Support, 2020b). This is arguably more important with brain tumour patients to ensure the specialist knowledge is shared and with the professionals in primary care to provide appropriate support that could mitigate the higher levels of need and distress in those affected by a brain tumour. The patients may require more support due to complex symptoms and poor prognosis and ensuring that patients feel they can get knowledgeable care and support could result from good implementation of this pathway.

Prior to discussing these findings in the wider context, it is important to highlight some differences in the data collected by the different methods. There were some differences between the focus groups, dyad interviews and individual interviews. In the focus groups, patients and carers often supported and validated other participants coping strategies. In three of the dyad interviews, the carers dominated the narrative which may have been related to the speech difficulties for the patient, however two of these carers verbalised that no one listened to their story before. The individual interviews, in a number of cases, reflected that there was no significant family carer and these participants spoke about feeling isolated a bit more than those with families. There were no significant experiences based on gender, except for the frustration in the loss of independence or job seemed to be more of a focus for the male patient participants.

3.6 Discussion

This was the first qualitative study to undertake an in-depth analysis of how patients with a brain tumour and their carers perceive the patient's needs and how concerns are currently detected and supported. The study also explored the perceptions of how advised strategies in detecting and supporting unmet needs should be implemented or improved. Participants revealed that the detection and support of psychological and practical needs are often overlooked with a focus on primarily physical concerns and symptoms, or the reporting of prognostic/medical information. The participants gave accounts of the impact of this which resulted in inappropriate care, a lack of advice on management of problems or a perception that their emotional needs were not a priority which in a number of cases was upsetting to them.

Importantly, the lack of knowledge and appropriate support for patients outside their specialist teams was one of the key findings in this study. This related to the quality of care for brain tumour patients in NHS England's (2017) National Cancer Patient Experience survey of 69,072 cancer patients that included 389 brain tumour patients. This survey highlighted that only 54% of brain tumour patients felt they had been given adequate care and support from health and social care services, the second lowest of any cancer type (NHS England, 2017). In the analysis of the data on variations of cancer types, the report acknowledged that in the analysis of 10 key questions, that patients with brain tumours reported the lowest scores on four of the nine questions, and notably the overall quality of care question. This survey also examined ethnicity, deprivation, age and gender and the authors highlighted tumour type presented the widest range of scores. While initiatives and funding might be focused on redressing the balance in some areas, such as deprivation, the focus on initiatives to support the imbalance of experience with brain tumour patients is not obvious. In Scotland, due to the small sample size of less than 50 patients, subgroup analysis was not reported for the most recent Scottish Cancer Patient Experience Survey in 2018 and patients were grouped with ‘other cancers’ for the comparative analysis between tumour groups (Scottish Government, 2019). However, the results of the 2015 survey which included 39 patients with a brain or CNS tumour out of 4835 (Scottish Government, 2016), reported 74% of patients rated their care as good or very good, which is 11% below the next tumour area and compares to an overall average of 90%. The majority of indices of good care were lower for brain/CNS patients than the grouping of all tumours, however, due to the small size, this subgroup analysis of brain tumour patients’ responses needs to be interpreted with caution.

The complexity of managing the neuro cognitive symptoms is likely to be a contributing factor due to the challenges for primary care to manage these uncommon symptoms and problems. The plan to transfer care to the community and increase the self-management and self-care after HNA of patients through national UK initiatives such as the ‘Living with and Beyond Cancer’ may be more challenging for this patient group (Macmillan Cancer Care, 2013). Some additional provision which may include better access to specialist teams which should be afforded to this group of patients.

The findings reporting here also showed that psychological distress was very common, while sometimes this was detected, in other cases emotional wellbeing was not routinely asked about. One study found 38% of patients with an intracranial tumour have psychiatric disorder with the majority diagnosed with an acute stress disorder and 78% reporting some level of distress (Goebel et al., 2011). This is linked with a reduced quality of life, poor health behaviours, higher healthcare utilisation and decreased ability to self-care (Gao et al., 2010). There are interventions which may alleviate this and without detection they will not be offered. Distress is often multifactorial and caused by physical, psychological, practical or spiritual issues so when detected for supportive care, interventions and/or advice is able to be planned. Therefore, solutions such as HNA, particularly when combined with a simple measure of distress such as the distress thermometer can be useful in the first step of assessment and detection (Goebel & Mehdorn, 2011).

Although this study did not initially aim to focus on carer's experiences and views, the emotional distress found in this group was an important finding as it has implications for the patient as well as the carer. The detection of distress for carers is vitally important as they may experience more distress that the patients (Seekatz et al., 2017). Carers provide emotional, physical and social support which is vitally important to the individual but often decreases the use of health and social care services. As this study has shown without this support this can result in significant psychological distress for them or in some cases the breakdown of relationship.

This study, similar to others, also demonstrated that increasing cognitive difficulties could cause social isolation, decreasing support networks (Madsen & Poulsen, 2011). However, the impact of social isolation came through from many patients, and the role of support networks such church groups or cancer support groups were of clear benefit for those who had accessed them. Referral to other agencies is an important consideration for healthcare professionals, particularly for patients who may not be more isolated.

It was initially planned that this study would help provide some information about how HNA tools, other strategies or interventions can contribute to the identification and support or resolution of unmet needs. It was clear that participants perceived that HNA would be useful and beneficial in helping them identify and discuss unmet meets with HCPs. The use of a tool that specifically captured the unique symptoms of a brain tumour population was clearly an important element. However, based on a review by the author and colleagues (Afseth et al., 2018), tools which might be considered for a brain specific HNA all would need further development to encompass all the domains of a comprehensive HNA and/or to further develop the psychometric properties. However, this study supported that it would be very challenging to further develop psychometric and comprehensive HNA tools due to the length and/or complexity of the tools. Completing PROMs may be more challenging in this patient group as non-completion is higher than other cancer groups in clinical trials and a factor may be the underlying neurocognitive deficits (Dirven et al., 2014). In this patient group, the needs are very diverse and there appeared to be benefit in having a large simple list (such as a tick box), which can highlight areas for discussion. While a tool like this is unlikely to provide a psychometrically valid patient reported outcome measure, it would support the view of many of the patients and carers in this study, who viewed this as strategy in order to enhance communication of their needs.

Future research needs to examine the impact of HNA as of a communication aid to elicit areas of concern. While HNA is already widely used in cancer, studies which have examined the impact of HNA or various proxy measures that could indicate improvement or success have generally not shown benefits of this process in randomised controlled trials (RCTs) (Carey et al., 2012). The authors of this systematic review stated, ‘need surveys, while being helpful in identifying particular patient concerns, are not appropriate as a focus for intervention development or outcome measurement (p219)’ and in brain tumour patients, some research is needed on what are the meaningful outcomes in this area.

3.6.1 Strengths and Limitations

3.6.1.1 Limitations

There was a high refusal rate for the focus groups from patients with decreased social support for transport or who had higher degrees of disability so their experiences may not have been fully represented within the focus groups. Early in the recruitment process it became evident that those with limited support or with more symptoms or poorer performance status were either not referred or refused participation at the time of discussion with the researcher. However, the addition of interviews as a data collection method facilitated the inclusion of patients with more significant symptoms and neurocognitive impairments as the interviews could be conducted in the patients' home. However, it is likely that there remains a cohort of patients with even more severe symptoms or worse performance status who were not able to participate and their views and experiences were not captured in this data. The inclusion of carers to some extent was a pragmatic decision, however this helped enable a more cogent view of what the patient or the 'family' unit have experienced and what they might need. This was an emergent finding, but the inclusion of the carers in a cognitively impaired population may have benefited from a more considered approach to inclusion and exclusion and could have also included those who might have experiences of caring for those who are more severely impaired.

In addition, the use of the brain tumour support group as one of the mechanisms for recruitment could have potentially included more individuals who actively seek support. This could have influenced some aspects of the analysis as it is possible that those that access these are more active in problem-solving or alternatively have greater support needs. It would be beneficial to seek verification of the findings here of these samples including the unrepresented group above, but also through similar research in other locations.

There are issues that were highlighted in the course of these interviews and focus groups in which distinct differences emerged between participants in many areas such as coping, self-management, emotional wellbeing and relationships with healthcare professionals. Although the research was designed to explore experiences and perceptions, this approach did not explicitly explore underlying potential generative mechanisms, such as how the experience of distress might be a barrier to self-management. The exploration of this might have been beneficial to help provide information on what interventions or approaches might help support greater self-management. The use of a different method that seeks to uncover what might work for different groups, such as realist methodology was therefore used for the next stage of this thesis.

3.6.1.2 Strengths

The use of three different data collection modes, interview, interview dyads and focus groups will have supported a broader understanding of the areas under study (Carter et al., 2014). Focus groups may allow participants to build or debate ideas while interviews will have allowed participants to more fully share their experiences. The inclusion of carers and patients added a further dimension which focused on the shared experience of that dyad.

As discussed in the methodology, a number of measures were introduced to help establish the trustworthiness and credibility of this study. In particular, the exemplar quotes help to support the presented findings and allow readers views some of the data that supports the resulting themes.

The sample was not restricted to any point in their care journey, so it allowed exploration of their experiences from many perspectives. The inclusion of carers offers three valuable additions: 1) pragmatically it allows and facilitated the inclusion of the patients themselves who needed support to take part in the research study and this is the first study to do so. 2) provides carer’s perceptions of HNA tools as one option for widening access may be proxy completion by carers and 3) carers of this rare condition have also been neglected in research of need assessments in brain tumour and their unique perspectives offer valuable insights.

3.7 Conclusion

This study provided significant insight into the experiences of patients with brain tumour and their carers in relation to unmet needs. This study provided a unique perspective through the inclusions of both carers and patients in brain tumour research in relation to the problems patient and carers might experience in relation to communication around their needs and strategies to address these. This study highlighted needs are often not clearly identified due to gaps in communication of problems or unmet needs between healthcare professionals and patients as well as between care providers. A brain specific HNA can support this communication and this can then allow appropriate care, support and referral to be planned and implemented.

It was also the first study in cancer research that allowed patients and carers to review existing HNA tools and give feedback on their perceptions of design, usefulness and feasibility. The carers and patients in this study provided unique insights into how PROMs or questionnaires like HNA might need to be designed to maximise accessibility. Poor completion rates for PROMs in neuro-oncology is a recognised issue (Rosenlund et al., 2019) and this research provides some important insights into this. This research seemed to indicate that a personalised approach around technology, proxy completion by a carer and place and time of completion should be considered.

The management of the treatment and neurocognitive symptoms is highly specialised and although primary care is important in supporting patients and their families, specialist input is often required to resolve problems. This study found that in many cases the needs of patients and their carers are not fully addressed by any care professionals through existing supportive care strategies, particularly emotional or practical needs. Distress is a significant problem for both patients and carers and is often linked to an area that often was causing a great deal of concern in their lives. HNA could be viewed as a beneficial process to help facilitate the identification of problems and help patients and carers provide information to guide consultations, and plan further care and support.

3.8 Summary and reflection on next steps of the thesis

The systematic review of tools and results of this qualitative study prompted an evaluation of the next step in this thesis. It was important to examine the findings to date and consider what the priority for research in this area is to build the understanding of how unmet needs for this patient group can be met.

3.8.1 Implications of the research in this thesis to date

Analysis of findings in this chapter prompted reconsideration of the direction of the PhD proposal. Initially this thesis was to be focused on the on the tool development including assessment of psychometric properties for HNA in brain tumours. Chapter 2 set out the criteria for designing an HNA which was psychometrically valid measurement tool, comprehensive and inclusive of the diverse needs of brain tumour patients. However, Chapter 3 revealed significant challenges for HNA tool development and more importantly utility as a measurement tool. Due to neuro-cognitive impairments completion for some patients, with any of the possible tools might be challenging. However, the participants in this study viewed HNA as beneficial process that might elicit improved communication for discussion of their problems, concerns or unmet needs with their health care team. These two considerations were reflected on with the support of the wider literature and are discussed in the next sections.

3.8.2 Why are HNA tools not an appropriate measurement tool for brain tumour patients?

It was clear based on this research in this thesis that designing a psychometrically valid tool for HNA would be unlikely to be useable as an outcome measurement, however there is also a concern that they may also have limitations as a measurement tool at all. While needs and problems can be examined and recorded on a measurement tool, they are very individual and varied, which may render them inappropriate as a ‘measure’ for outcome due to these factors. While HNA is used to identify and address the unmet needs – studies which have used either reduction of needs on the same scale or various proxy measures that could indicate improvement or success have generally not shown benefits of HNA (Carey et al., 2012). The authors of this systematic review stated, ‘need surveys, while being helpful in identifying particular patient concerns, are not appropriate as a focus for intervention development or outcome measurement’ (p217)

Other measures have also been suggested as appropriate outcomes measures but there is a lack of consensus of what success means in terms of HNA. Some studies that evaluated the impact of HNA and associated interventions focused on the reduction of distress or other psychological benefits (Carlson et al., 2012; Snowden et al., 2011). The effect on wellbeing has also been suggested as an aim through improvements in QOL (Morrison et al., 2012). However these other measures have not demonstrated positive outcomes as supported by a number of reviews of needs assessment (Bidstrup, et al., 2011; Carlson, et al., 2012). These reviews include studies that looked at using a HNA tool and providing this information to the health care team (compared to not using a tool), but with no specific interventions being tested, did not demonstrate any significant improvement in meeting unmet needs (Boyes et al., 2006; Girgis et al., 2009; King et al., 2009). This may indicate that by simply providing the team with information on unmet needs has no effect on decreasing needs.

However, even with interventions designed to alleviate unmet need there have been no significant effects with randomized controlled trials (Allen et al., 2002; Aranda et al., 2006; McLachlan et al., 2001). There have been a number of suggestions for the lack of efficacy such as, a lack of uptake of support, design of interventions was not appropriate, or interventions may be inappropriate for some patients due to individual patient factors. In addition, needs flux and wan, and while an intervention may fully meet one need, other needs can emerge which may also limit the sensitivity of this as an outcome measure. Therefore, the planned development of an HNA tool was ceased and alternative areas of research aimed to focus on how this could be beneficial for patients.

3.8.3 How could HNA be beneficial for brain tumour patients?

The first consideration of how this could be beneficial came from some of the review of HNA that suggested assessment without the resulting actions to address HNA did not demonstrate any significant improvement in meeting unmet needs and in some cases appeared to have a trend of being detrimental (Carey et al., 2012; Schouten et al., 2019). Therefore, assessing needs without appropriate actions to resolve or mitigate needs is unlikely to have an effect. So, to consider the benefits of HNA, this must be considered as part of programme inclusive of HNA and the subsequent actions. However, the process of providing support for unmet needs for any cancer is complex and arguable for brain tumours more very complex due to the unique neuro-cognitive impacts. The model of need in Brain tumour patients as illustrated in Figure 8 proposed a pathway of care that considered the contextual factors relevant to brain tumour patients. This model proposed how HNA could improve the identification of needs and the care, support and referrals could be implemented support favourable outcomes such as improved self-management and decreased distress. However, to explore how this conceptual model could translate into practice, an approach which considered the complexity and variability of an HNA programme for patients with brain tumours was needed. The next chapter will undertake a realist review to propose how an HNA programme could be implemented within the current context of policy and practice.

4 Chapter 4: A realist review of HNA in brain tumour patients

This chapter will first give an overview of the justification for using realist methods and then provide a general overview of realist methodology. This will be followed by the methods and results of a realist review focused on:

4.1 Justification of a realist approach

As previously discussed, there is considerable evidence that patients who have experienced cancer have unmet needs and brain tumour patients may have higher needs than others (Carey et al., 2012; National Cancer Survivorship Initiative, 2014; Rooney et al., 2013). HNA has been presented as a strategy and potential solution to detect the unmet needs of patients with cancer to allow strategies to be put in place to address these, improve patient-centred care and better target supportive services (National Cancer Survivorship Initiative, 2014). Despite the absence of evidence of efficacy, this continues to be the supported strategy of many national cancer agencies, networks and charities. Within the UK it has been supported through considerable investment from Macmillan and the UK governments (Macmillan Cancer Care, 2013; National Cancer Survivorship Initiative, 2014). There is recognition of a need for evaluation of this process and specific interventions to determine support and how this can be delivered in the most effective way (Macmillan Cancer Care, 2013).

As previously reported, no reviews or RCTs for HNA for cancer have successfully demonstrated the positive impact of HNA, or interventions after HNA assessment. However, it could be argued that there is a missing aspect in terms of the impact of reasoning and personal choices of those receiving and administering the interventions, the environment of the individual and variables in their care delivery. Tremblay et al. (2014) argue that, in offering appropriate support and care which is based around an individual assessment, there is likely to be a large amount of variability due to interventions, patient characteristics, practitioner characteristics, and the service. Given the nature of needs and its multimodal support strategies, people with cancer receive care and support from multiple professionals from different disciplines, working in a variety of settings, whose services may be provided either concurrently or at different points in time. In RCTs, where the aim is to minimise variability delivering support for unmet needs will present challenges to this study design.

An alternative approach may to be to undertake a more comprehensive evaluation, which recognises this variability. Shiell, et al., (2008) describe non-drug interventions as ‘complex’, which can refer to the property of the intervention or the system in which the intervention is implemented. Pawson and Tilley (1997) highlight that, in complex programmes where there are multiple variables that could influence outcomes, experimental models seek to control influential variables. Instead, exploring these variables could reveal how these might influence outcomes.

4.1.1. Complexity in HNA programmes

First of all, it will be useful to define what is meant by ‘complex’ and ‘complexity’ and how HNA programmes fit into this. Complex interventions are defined as being composed of parts that make the whole intervention and – in isolation or combination – generate the power or outcomes in response to the intervention (Clark, 2013) or interventions with several interacting components, such as those that occur in the health service (Craig et al., 2008). However, in considering complexity in healthcare, it is not just the intervention, but also the system or systems these interventions operate within (Greenhalgh and Papoutsi, 2018). These authors note that the definitions and framework used by the Medical Research Council (MRC) has also evolved. In 2008, the MRC’s framework and guidance for developing and evaluating complex interventions was focused only on the components of the intervention. However, the latest guidance from 2021 recognised that complexity has other dimensions that need considered, such as the system, behaviours of those receiving and those delivering interventions, and variability that can be influenced by context (such as location, time, recipients etc.) (Skivington et al., 2021). However, despite the changes in how to view complexity from influential funders such as the MRC in recognising the multi-dimensional aspects of complexity, much of the research community remains wedded to earlier definitions and focuses only on the actual intervention (Greenhalgh and Papoutsi, 2018).

As highlighted above, the earlier MRC frameworks for designing and evaluating complex interventions were focused only on the complexity of the actual intervention and supported methods such as RCTs to test and evaluate these. However, this is insufficient as it ignores the emergent causality with impact on outcomes through the combined and separate influences of organisational, social, cultural and geographical systems on intervention effects (Campbell et al., 2007). Complexity informed health services research requires research methods which seek to understand the dynamic, inter-relational, and emergent and multi-causal aspects of research issues in health (Greenhalgh and Papoutsi, 2018).

The opposite of this might be illustrated by a simple analogy; if you have A, and you do B, you will get C. For example, if an obese person eats a very restricted diet, they will lose weight. However, it has been demonstrated that, despite this very clear formula, other variables will influence the likelihood of weight loss being achieved. Therefore, some researchers might propose that this is more ‘complicated’. They may look at adding aspects and making this a specialised area of study. Specific diets, such as high protein, and psychological interventions might be tested in the context of a RCT. But, despite many years of research into this problem in the developed world, obesity is rising and the response to interventions is inconsistent at best.

This may be that research into programmes to manage weight loss need to consider the complexity which may be due to relationships, the environment, and other elements, such as individual demographics. With many complex interventions, like HNA, the empirical positive outcomes have not been demonstrated. A different research approach may be more appropriate. For example, rather than question what intervention works best, it might be more appropriate to focus on why an intervention might work for some and not for others. What are the factors that can influence this? What might be a plausible explanation for why they work? The consideration of systems, relationships and behaviours of those administering and those receiving HNA programmes are areas that need to be considered in studying this phenomenon.

With this complexity, many interventions or programmes are not easily evaluated by traditional experimental designs (Bonner, 2003) as these do not take account of the systems or behaviours which influence outcomes. Researchers are increasingly urged to use evaluation designs which not only capture outcomes through quantitative data alone, but to also gather qualitative data which elucidates implementation processes, and contextual factors (internal and external) that may influence the success (or otherwise) of an intervention. Realist methodology recognises these factors and stems from critical realism. Realist methodology was used in this thesis.

4.2 Critical realism – a philosophical approach

Bhaskar (2008) who is widely recognised as the first author to set out a meta-theory of critical realism for social sciences, described several tenets of critical realism in 1975. The first is that we have an individual reality in our mind or mind-dependent reality – however that is not the same as physical reality or mind-independent reality. The next premise is that reality is stratified and generative. We have actual events that happen, and these occur through processes that might be viewed as empirical. However, the way something happens (mechanism) and how an individual experiences it 'gets in the way' of these empirical processes. Mechanisms, events and experiences are the overlapping domains of reality. There should be a focus on 'why' it happens and the explanation of what might be causing this to happen (or not). Within this explanation there should be recognition of the effects on the individual and environment. There can be a reality and it is not chaotic, but due to the open and natural system that our interventions occur in, it is complex. There will be trends that make an event more likely and there will be exceptions – we should aim to understand why this is. Reality is generative and complex. To understand these complex interventions our research approach should be eclectic and use whatever means we need to employ to understand reality.

A number of researchers and theorists have ascribed to critical realism to help develop an explanatory ontology in various fields. An examination of these will help illuminate the benefits of these in different fields and also provide some indication of how this philosophy could be beneficial in examining HNA in brain tumours.

One of these from the field of economics was Tony Lawson who criticised the traditional economic mathematical modelling. His main criticism was that it ignores the open and social world that economics operates within and does not look at the field holistically (Lewis, 2009). He asserts this has resulted in economics failing to deliver explanatory or predictive theories as these have ignored the nature of the social world (Lawson, 2003). Alternatively, he suggested that the socio-economic model of economics recognises the role of human choice and behaviour in response to certain conditions, or human agency. He uses the term 'social reality' to denote the phenomena whose existence is dependent to some extent on human agency. An example might be a structure in economics, such as a mortgage, that is dependent on human agency in terms of how someone decides to take a mortgage, pay it etc. But human agency can also evolve and change in response to structure, so these interactions are part of the social reality which needs to be considered. Another area that Lawson highlights, is the need to consider the holistic nature of society – and with this he refers to the network of interconnectedness that can have an influence on human agency and is influenced by structure. For example, through capitalist and worker or parent and child. The inclusion and consideration of these additional aspects described above ascribes to critical realism. This philosophical view rejects the purely empiric, traditional mathematical modelling and instead suggests the socio-economic model can provide potentially better predictive and explanatory frameworks. By recognising the role of all causal mechanisms, including those that may be non-empirical, explanatory frameworks can be better formulated. For example, in traditional modelling, if you introduce a stimulus that has been demonstrated to support economic growth, it should produce comparable outcomes in similar settings. However, in reality this rarely is the same. Searching for the unidentified causal mechanism(s) that account for these differences in growth, for example, differences influenced by the population or society, might provide a more accurate explanation. This may be a mechanism of human agency and the response to structures, for example, behaviour may vary in a different cultural setting. This approach is more complex but may provide a better and more sensitive model for this field.

Williams (1999) from the field of sociology has examined the 'body' through the lens of disability and chronic disease. Using this lens, he explains how critical realism provides a beneficial lens through its '*deep underlying structures and mind-independent generative mechanisms'* (page 798) providing a way of viewing these issues that does not have the limitations of other theoretical models which seek to compartmentalise the 'body' into a specific category. To help explain this Williams explores how the view of the 'body' in chronic illness has been categorised based on different theoretical approaches. For example, foundationalism may consider the body's organic matter, social constructivists may examine the body as discursive output of power or knowledge. Historically there has been a biomedical focus on the study of chronic illness and disability. This focused on the somatic or physical body and to what extent the body was impaired. This was challenged by social constructionists and is a view shared by the disability movement, that the focus is not about the 'body' and its limitations, but rather by the premise is that disability is caused by the dysfunction of society that has not removed barriers and prejudice to allow full participation. This premise holds that it is not about the body itself but rather social oppression. However, this view is very limited and lacked the integrated consideration of the physical and social, material and cultural, experiential and representational of health and illness, and critical realism can provide a basis for providing this integrated approach. There are some key points this paper highlights and these points also are useful for this thesis to highlight the benefits of critical realism.

With chronic illness or indeed cancer, there is an empirical impairment of the body that exists, independent of the mind (Williams, 2003). However, the experience of this impairment does impact the relationship of the individual with society and this can help us rethink of these interactions. This supports Bhaskar's premise of stratified reality. So, for example, whereas social constructivists who ascribe to the construction of social entities (social rules, culture, family relationships), critical realism recognises social entities exist *empirically* and are mind independent. However, these may not be experienced by all, but when experienced become *actual,* however, only become *real* through experiences, events and the interaction with the generative causal mechanisms (which may be undetected). The recognition of this stratification is an important aspect as this recognises and legitimises the impairment caused by brain tumours. When interventions are introduced, critical realism can help explain *how these are experienced* and provide an explanation of outcomes through searching through the causal generative mechanism.

4.1.1 Complexity and linkage with Critical Realism

Sturmberg (2016) proposed there are two approaches to explore issues and problems in healthcare, through a lens of reductionism or complexity. Reductionism seeks to break 'reality' into parts and then study these individually to develop the understanding. Complexity in contrast explores the relationships and interdependencies between component parts in a certain situation. Complexity is present in most healthcare situations as it is influenced by human choice and behaviour and therefore is not easily predictable or linear.

Complex systems recognise a number of agents are networked and this is all part of a larger system and therefore all the elements in the system need to be considered holistically. Additionally, these systems also adapt over time in response to these agents and systems. Interactions between agents are fed back to create feedback loops which could be re-enforcing or stabilising – maintaining the status quo. Alternatively, there may be a destabilising influence and in this case, the systems are emergent which further increases the complexity.

There is some clear alignment between critical realism and complexity. They both recognise the holistic system – rather than component parts. There is also the importance of consideration between interaction of structure and agency and how human behaviour or responses might influence this. This human behaviour may be the undetected generative causal influences. There is also a clear recognition of the open and evolving nature of the areas of under study. Self-management clearly requires behaviours or responses in relation to intervention which may lead to positive or negative outcomes, therefore in this ‘complex’ field, critical realism does provide a good lens to research this issue.

However, with this complexity, that can increase exponentially as more agents or structures are brought in, which is likely to be the case for using HNA and responding to unmet needs in brain tumour patients. And with both complexity and critical realism there is a recognition that constant event patterns are unlikely be obtained. However, within open complex systems, critical realists contend there may be recurrent patterns where some prevailing factors are place and these can lead to relatively enduring outcomes and these have been labelled as demi-regularities (Pawson and Tilley, 1997). These demi-regularities provide evidence and direction to the potentially identifiable mechanisms in play (Scambler, 2001). This is one of the important aspects discussed in realist methodologies which use a critical realist approach and provides a framework for researching complex interventions.

4.3 Realistic methodology

Realistic evaluation or synthesis is underpinned by critical realism and was first described by Pawson and Tilley in 1997. This has been proposed as a suitable methodology for evaluation of ‘complex’ health interventions (Marchal et al., 2012). While there are a number of theory-based evaluation methodologies, realist evaluation has a focus on critical realist philosophy. It is concerned with the nature of reality, how causation works, and what these assumptions imply for evaluation design, methods and utilisation.

A realist review recognises the variability of contextual influences and human responses (Pawson, 2006b). This is beneficial to use when the evidence is heterogeneous, as this process will allow the reviewer to explore and focus on the complexity of programmes of interventions, recognising the various influences such as context, mechanisms (defined as resources and responses) and outcomes. A realist review applies realist philosophy, using a theory-driven method and iterative process, to the synthesis of findings from data sources that have a bearing on the research question.

This approach is a theory-based evaluation and has a starting point of defining the ‘programme theory’. These theories postulate how programme activities are understood to cause (or contribute to) outcomes and impact – for example, if we deliver a ‘programme’ in a certain way it should bring about an improvement. Another core concept is that interventions achieve their effect through the action of the individual. Therefore, in part, its success or not will be linked with the personal choices and actions of the participants (Pawson et al., 2005). This contrasts with RCTs, which try and protect against this causal interference. Realist evaluation should then track these theories to evaluate their relevance (Pawson et al., 2005)

Finally, realist evaluation postulates that ‘intervention theories’ will have had a long journey, often beginning with policy and then passed on to practitioners. Different groups are crucial to implementation and success depends on the integrity of the implementation chain. Flows, blockages and points of contention are vital to this (Pawson et al., 2005).

Some important core concepts in realistic evaluation as outlined by Pawson and Tilley (1997) need to be highlighted to understand this approach.

- *Programmes* is the terminology that describes the package of intervention(s), being aware that these are introduced into social context. In this case it can be considered as an HNA programme. This is represented by the Recovery Programme (Macmillan Cancer Support, 2020b) in the UK
- The aim of realist methodology is to explain the *regularities* – which can be the *outcomes*, patterns, associations or correlations that are deduced (Pawson & Tilley, 1997)
- *‘Programme theories’* are used to explain these *regularities* – which can be the *outcomes*, patterns, associations or correlations deduced from the context, mechanisms and outcomes. These theories are therefore explanatory statements ‘attached’ to a certain context that theorise how interventions can lead to a specific outcome (Shearn et al., 2017)
- *Mechanisms* are a key element of realistic evaluation. Outcomes only work through the actions of mechanisms, which can be resources and reasoning (Dalkin et al., 2015)

- *Context* is a crucial factor to be analysed in terms of realistic evaluation – what prevailing social contexts help or hinder successful outcomes (Pawson et al., 2005)
- The final concept is of *change.* This occurs when those involved in the programme start to realise that certain contexts are relevant and a choice of approach can make differences to the outcome.

Causation (how programmes cause change) and attribution (whether observed changes can be attributed to the programme or were caused by other things) are critical questions in realist methodologies. It is also important that a 'realist' understanding of causation – cognisant of mechanism and context – is required in a realist review (Westhorp, 2014).

The central premise in realistic evaluation is that context (C) + mechanism (M) = outcomes (O) and this will generate the programme theories. However, there are likely to be several theories to support this generation. For example, $C^1+M^1=O^1$ then $C^2+M^2=O^2$ etc. There will be multiple indices for both groups in this trial that could be surrogates for 'success' and proposed as outcomes.

Realist approaches assume that nothing works everywhere or for everyone, and that context really does make a difference to programme outcomes. Consequently, policy makers and practitioners need to understand how and why programmes work and don't work in different contexts, so that they are better equipped to make decisions about which programmes or policies to use and how to adapt them to local contexts. Consequently, realist evaluation does not ask 'what works?', 'does this work?' or (retrospectively) 'did it work this time?' This review seeks to unpack the context-mechanism-outcome relationship, thereby explaining examples of success, failure, and various eventualities in between. Through examining these there may be semi-predictable patterns, but these are variable as the human choices or in some instances contextual differences influence the outcomes (Pawson and Tilley, 1997). These demi-regularities can help theorise linkages to explain why programmes may or may not work. These theoretical explanations are referred to as 'programme theories'.

4.3.1 Rationalisation of this approach in the context of HNA

The aim of improving outcomes through assessing needs and then responding 'appropriately' would in most cases require highly variable interventions with a wide array of individual and contextual factors. This would present challenges to an experimental design which seeks to minimise or eliminate these variables. There is widespread support for the implementation of HNA – but this is despite the lack of positive outcomes in randomised trials or other quantitative studies. Therefore, the use of realist methods which incorporates the exploration of different contexts and mechanisms, and how they link with positive or negative outcomes is a valid approach to help figure out what might work for whom, in certain circumstances (Pawson & Tilley, 1997). This is supported by Westhorp (2014, p.7), who asserts:

> "Realist impact evaluation is most appropriate for evaluating new initiatives that 'seem' to work but where 'how and for whom' is not yet understood; initiatives that have previously demonstrated mixed patterns of outcomes; and those that will be scaled up, to understand how to adapt the intervention to new contexts."

4.4 Introduction to the realist review

4.4.1 Rationale for focus area

As demonstrated in the previous chapters, there is no ideal tool for HNA in patients with a brain tumour. In addition, there are unique needs and implementation considerations for HNA with brain tumour patients and their caregivers. These occur as a result of the increased burden of symptoms or other unmet needs, compared to most other cancers. These needs are often directly related to neurocognitive problems or the consequences of these. One example would be the loss of a driving licence (due to seizures) that can lead to loss of independence. Previous chapters have noted that unmet needs are common in cancer patients and often not assessed or explored in routine consultations. Therefore, national policies aim to address these needs by recommending the use of HNA.

The national policies discussed in Chapter 1 all support the use of HNA for cancer and have an overarching aim of assessing unmet needs to then alleviate unmet needs (Appendix 11). However, the outcomes or goals that are desired through meeting needs, are variable, and include, reducing distress, improving QOL, improving self-management, or decreasing unplanned care identified as other goals of HNA. These are related to the focus of cancer policy in that region.

In the US and Canada, the aim is largely focused on reducing distress (Bultz et al., 2011; National Comprehensive Cancer Network, 2018). A review of the effectiveness of needs assessment and distress screening that explored the impact on reducing distress was inconclusive, and the authors advised that research was needed into certain aspects of HNA, such as the methods of assessment, timing, cost effectiveness and impact of staff training (Carlson et al., 2012). Although there were no conclusive results, recommendations were made, such as the need for staff involvement in implementation, administrative support, clear supportive care provision for identified needs and acknowledgement and provision for clinical judgement. These recommendations could be relevant for all HNA programmes – even where the primary goal is not distress management, for example within the UK.

In the UK, HNA is not a focused intervention for decreasing distress. It is multifaceted, with the aims of improving care after treatment, including improving knowledge and care planning, continuity of care between secondary, primary, social and community care and improving self-management (Macmillan Cancer Support, 2015; Scottish Government, 2016b). This is similar to policies in Australia, which directly advocate self-management or allude to it through phrases such as ‘empowerment’ (Clinical Oncology Society of Australia, 2016). Self-management is a clear goal of HNA packages and many authors link the implementation of self-management strategies to realise the benefits of HNA (Primeau et al., 2017; Wells, Cunningham et al., 2015). However, there has not been a review of how self-management might be impacted by HNA in cancer. Therefore, the main focus of this realist review will examine how HNA programmes can influence self-management in brain tumour patients and their families. Self-management is closely linked to the concepts of self-efficacy and coping through theoretical frameworks – this will be discussed later in this chapter. In addition, the relationship of these concepts or traits to each other was also a significant finding of the work contained in this thesis (Chapter 3).

Understanding variability in implementation and response to an intervention is the aim of realist research, which helps to understand what might work for who and in what circumstances. This chapter will focus on using a realist synthesis methodology to understand the variables that might have an impact on the success or failure of an HNA programme.

4.4.2 Holistic needs assessment programmes

As previously discussed, an HNA is the first step of a larger programme to improve outcomes for individuals living with cancer. Very broadly speaking, an HNA programme is the actual assessment, but also the actions that follow in response to these needs. Therefore, in this review, the actual assessment will be referred to as an 'HNA' and the HNA and its responses will be referred to as an 'HNA programme'. This review will consider this intervention in the context of the UK model, which is explicit in England as the 'recovery package' (Macmillan Cancer Support, 2015). The reason for this is that the UK model has self-management as a key outcome – this will be a focus along with self-efficacy and coping. This model is also followed in the other nations such as Scotland (Johnston & Campbell, 2018b), however, with a more flexible approach at times. While many aspects of this programme are relevant, this *exact* programme was not evaluated, as this HNA pathway is much more variable in Scotland. At the time of writing there is no defined HNA programme implemented for brain tumours.

The core elements of the UK model are:

- Holistic needs assessment (HNA).
- Care planning.
- End of treatment summaries which give both the patient and other providers information.

- Health and wellbeing events.
- Information provision.

Figure 2 in Chapter 1 provides an example of implementation of an HNA programme in the context illustrated by the UK Macmillan Recovery Package (2015-2020) (The Independent Cancer Taskforce, 2015).

Since the aim of this review is to determine what works for whom in which context, global literature is included, as most other countries have similar approaches that may have relevance – in particular, literature from Australia on this topic, as HNA or needs assessment has been widely implemented to support self-management.

4.4.3 Self-management and self-efficacy

The concepts of self-management and self-efficacy are linked but with subtle differences. As key concepts in HNA, these need to be clearly understood. Self-management can be seen as how an individual behaves in a given context (Baydoun et al., 2018) and can be defined as the activities and strategies that individuals undertake to deal with the physical and psychological consequences of cancer (McCorkle et al., 2011) and which promote survival, health and wellbeing (Foster et al., 2015). Self-management has been demonstrated to be effective in cancer and other long term diseases, improving health behaviour, reducing demands on services, improving communication between patients and health care providers and improving self-efficacy (Davies, 2009). In contrast, self-efficacy is a perception in capability that can be a moderator of behaviours (Baydoun et al., 2018).

Perceived self-efficacy is also a key determinant of how individuals might behave in self-managing their problems. Bandura (1999) theorises that those with high self-efficacy respond to challenges differently than those with lower levels. For example, individuals with high self-efficacy, when faced with obstacles, respond more positively and may work hard to overcome these as they have a strong belief in their capabilities. Thus, those with high self-efficacy are able to moderate their own behaviours through setting goals and evaluating their performance, increasing their motivation and performance attainments. There is evidence that high self-efficacy leads to improved health behaviours including adherence (Baydoun et al., 2018). However, Foster et al. (2015) highlights that self-efficacy is not a fixed trait and varies according to the context and the activity. As an example, in their study cancer survivors reported high self-efficacy in relation to finding information, but lower levels in relation to managing fatigue.

Fatigue is the most common symptom reported in patients with PBTs (Day, Yust-Katz et al., 2016). Individuals diagnosed with a brain tumour and their families often have significant input from health and social care professionals to help manage fatigue or other symptoms. However, there is limited evidence of efficacy of any medical therapy for fatigue but interventions such as exercise may have benefits in reducing it (Day, Yust-Katz et al., 2016). Therefore, self-management remains vitally important. In most cases they will be at home, striving to manage and coping independently for much of the time. Self-management can include guidance from professionals but also other activities that patients and their caregivers consider will improve their wellbeing. It also includes the task of navigating services and working with healthcare professionals for information and support in self-management.

Supporting those affected by a brain tumour in self-management strategies can improve perceptions of self-efficacy. This may have other benefits, such as improving psychological outcomes. There is evidence to suggest that those with higher self-efficacy are associated with improved wellbeing while those with lower self-efficacy are associated with higher levels of depression and increased perception of threat from their illness (Foster et al., 2015). This study across a variety of contexts found that socio-demographic factors such as social support were positively associated with self-efficacy. Interestingly, clinical aspects such as tumour type or co-morbidities did not have such an association. These authors conclude that those most at risk of low self-efficacy are women, those experiencing higher levels of pain, depression, lower wellbeing scores (measured by QOL), who did not own their own home, had low levels of social support and a more threatening perception of illness. This study did not include brain tumour patients as a distinct group. However, many of these variables may also be significant for brain tumour patients, such as the threat of illness.

As reported in Chapter 3, the ability of individuals and their families to cope was strongly intertwined with their perceived self-efficacy, which appeared to support their ability to self-manage. This link between coping and self-management connects to psychological influences and past experiences may also play a significant role (Richardson et al., 2017).

4.5 Objectives and focus of the review

The aim of this review is to develop and propose a theoretical framework that depicts context-mechanism-outcome (CMO) configurations (programme theories) of the effect of an HNA programme on supporting self-management for brain tumour patients. The aim is to answer the research question – how, and in what circumstances, might an HNA programme improve self-management in brain tumour patients?

Research questions

1. How to define the barriers and facilitators (or mechanisms) related to the effective use of HNA (implementation, assessment and responses) to facilitating self-management?
2. Can we define the contextual factors which relate to enhancement or detraction from outcomes?
3. What are the preliminary hypotheses – based on the literature that proposes the underlying triggering mechanisms or interventions in different contexts – that lead to desired outcomes, such as improving self-management and self-efficacy through HNA?
4. Can these hypotheses be interrogated to produce a theoretically based explanatory framework on how HNA in brain tumour patients works best, for whom and in what circumstances, to improve self-management?

4.6 Stages of the realist review process

The **Realist** And Meta-narrative Evidence Syntheses - Evolving Standards (RAMESES) for realist reviews has guided the methodology of this review (Greenhalgh et al., 2015). There are a number of steps which are considered good practice (Rycroft-Malone et al., 2012) and the following will be elaborated on and described in the next sections.

- Defining the scope of the review and purpose of the review
- Scoping searches and identifying initial programme theories to then be refined to preliminary candidate theories
- Iterative literature search strategy
- Selection of articles
- Analysis of literature and extraction of data related to candidate theories
- Synthesis of findings resulting in refined programme theories
- Writing the narrative and drawing of conclusions

4.6.1 Scoping and identifying preliminary theories

4.6.1.1 Overview

The aim of this first stage is to identify initial 'programme theories' which provide a starting point to describe the mechanisms that are likely to operate, the contexts in which they might operate, and the outcomes that will be observed if they operate as expected. The focus is to suggest hypotheses that underlie the use of HNA and that may support self-management for this patient group.

Many realist reviews can start with searching around studies in the area of interest to generate ideas about relevant programme theories and test their relevance (Greenhalgh et al., 2014). There are extensive policy documents and literature on HNA but very limited literature on HNA in patients with a brain tumour. Therefore, a modified approach was taken, with several sources of data used. With the support of data from Study 1 in Chapter 3, initial programme theories were proposed.

The generation of 'initial programme theories' was supported by a number of steps. Three sources of publications were used. Policies of countries with widespread implementation of HNA for cancer were reviewed. These provided the descriptions of policy aims or outcomes and relevant components of this intervention (Appendix 11). The second source was reviews of HNA programmes (Appendix 12). Finally, middle range programme theories from nursing, psychology and sociology that could help explain the ‘chains of inference’ for how this programme may or may not work in certain situations or explanatory frameworks were considered (Table 10). These theories were explored to help illuminate key mechanisms for behaviour change processes that are important in self-management (Mills et al., 2014).

These ‘initial programme theories’ were presented in a number of meetings of the supervisory team, cancer researchers experienced with HNA, and realist methodology experts. The formulation of these also drew on the author’s experience as a senior nurse, manager and researcher in the area of cancer for 20 years. Specific expertise in HNA has been gained through the research on this dissertation as well as Scottish and UK cancer advisory work. This knowledge supported the formulation and sense checking of the initial programme theories. After a period of refinement, this resulted in *candidate programme theories* that were the focus of the in-depth literature review.

4.6.1.2 Defining the scope and components

The scope of this realist review was defined through consideration of the relevant interventions. These were interventions linked to a holistic assessment of needs and then the response(s) to this assessment. As with most realist reviews, the complexity which may accompany these interventions was considered. For the purposes of this review, the interventions considered as part of the core programme for HNA were:

- An HNA.
- Associated care planning.
- Referrals, advice, communications with individuals, caregivers and other care providers, supportive care interventions and wellbeing events.

The assessment processes, as well as the actions for the development of self-management which followed, were the focus of this inquiry.

4.6.1.3 Formulating initial programme theories – detailed methodology

The detailed process of how each source was used in the generation of initial programme theories is described below. A consideration of the 'conceptual model of need' as presented in Figure 8 (Chapter 3) provided some causal chains of inference to help shape the initial programme theories.

Policy documents

A review and analysis of policy and practice documents was undertaken for countries that have wide-scale adoption and/or national support of HNA programmes. The countries included were the United Kingdom, United States, Canada and Australia. These policy documents are important to consider as they reflect the proposed ideals of how the policy makers would like to see these programmes work. They largely encompass ideas of holistic and integrated patient-centred care that take into account the patients' priorities and concerns. Features such as shared decision making, empowering patients and self-management were prominent. Details of these were included, such as the underlying aims and characteristics of programmes, interventions and intended outcomes (Appendix 11). The supporting documents for these policies were analysed for potential components of the CMOs that are also reflected in this appendix.

Systematic reviews and selected literature

A scoping search was then undertaken, focused on systematic reviews which evaluated the impact of HNA, and relevant data was summarised (Appendix 12). These reviews are important to consider – while policy might be clearly stated, less is known about how these programme work (or do not work) in meeting the needs of cancer patients to improve wellbeing, such as HRQOL or reducing distress. The contribution to initial theory was considered and coded based on CMO configurations or, in many instances, two aspects of these, such as 'CO' or 'MO'. It was also considered whether the mechanisms were a response or a resource and if this was enabling or disabling in relation to the intended outcome (Dalkin et al., 2015).

Theoretical underpinning

The process for programme theory generation involves deduction from existing middle range theories – such as Orem's self-care theory – or grand theory, such as humanistic theory. These theories are often examined through deduction with research and practice based findings used to propose new middle range or situationally based theory (Im, 2018). They can be a useful strategy to support the analysis in for a number of stages. These include helping to guide initial programme theory, shaping the review protocol for theory refinement and supporting the conceptualisation of outcomes (Jagosh et al., 2014). This can provide a theory-based framework against which the researcher can extract contextual and effectiveness data based around the set outcomes. This has been advocated as an approach in realist reviews (Booth & Carroll, 2015). Based on the behaviours of interest (self-care, self-efficacy and coping), a number of models, theories or frameworks were considered (Table 10). Social cognitive theory (Bandura, 1999) and the theory of symptom self-care management for adult people with cancer (Baydoun et al., 2018) were chosen both as widespread empirical research in cancer helped to reinforce the concepts of these theories. The consideration of various mid-range theories is presented in Table 10.

Table 10 - Theories, frameworks or models considered to help shape initial programme theory

Theory or concept	Reason for consideration	Inclusion or exclusion	Reason for decision
Orem's theory of self-care (Denyes et al., 2001)	Focused on self-care as maximising wellness	Excluded	Focused on 'medical' issues and not full holistic needs. Did not seem to look at shared or collective mechanisms for wellness
The Individual and Family Self-Management Theory (Ryan & Sawin, 2009)	Focus on the individual and family with purposeful incorporation of health-related behaviours into an individual or family's daily functioning. Self-management prevents or attenuates illness or facilitates the management of complex health regimens in ways that reflect individual	Exclude	Lack of emphasis on processes and underlying self-efficacy. However, does consider context so potentially useful in synthesis

	and family values and beliefs in personally meaningful ways. (Can be in collaboration with health professional)		
Wagner’s Chronic Care Model (Wagner *et al.*, 2002)	Includes a focus on community as well as secondary health services. Looks at interaction between the individual and health care team	Exclude	Has some good elements, such as a focus on decreasing healthcare utilisation and adherence to healthcare advice. In practice, increases patient burden and lacks focus on QOL (Boehmer et al., 2018)
Ajzen’s Theory of Planned Behaviour (Ajzen, 1991)	Focus on three clear areas of influence (attitude towards behaviour, subjective norms, and perceived behavioural control). Behaviour is ultimately self-regulated	Exclude	Is focused on mainly the individual – and not external influence

The Corbin and Strauss Chronic Illness Trajectory Framework (Corbin & Strauss, 1991)	Focused on diseases that are likely to last for the rest of an individual's life. Moves through phases and is useful for planning care and ensuring patients' perspective is considered. Adapted to cancer survivorship (Klimmek & Wenzel, 2012)	Exclude	Focused on goals and actions rather than internal reasoning (or self-efficacy)
Social Cognitive Theory (Bandura, 1999)	See below	Include	Focus on individual, proxy or collective agency. Recognises the role of self-efficacy and coping, which were linked with 'empowerment' of HNA programmes. It also has flexibility, which allows the consideration of context and mechanisms from multiple viewpoints
Theory of symptom self-care	See below	Include	Builds on Bandura's social cognitive theory with a cancer focus. Self-management is the central concept with self-regulation and self-efficacy as key moderators

management for adult people with cancer (Baydoun et al., 2018)	

The framework outlined in Bandura’s Social Cognitive Theory (Bandura, 2001) was utilised to help group the behavioural response through the lens of ‘agency’, which can be divided into personal agency, proxy agency (through the efforts of other individuals) or collective agency (group effort). HNA programme support empowers the individual, through individual, family and community (such as wellness events) (Doyle & Henry, 2014). Therefore, this focus on how agency can contribute to beneficial outcomes could be a suitable lens to help build strategy.

There are many examples of interventions to support self-management in long term conditions and Bandura’s Social Cognitive Theory has been the basis of the majority of these (Boger et al., 2015). The focus of this theory is self-efficacy and this is a central component of self-management. Foster and Fenlon (2011) define cancer-related self-efficacy in relation to self-care as the belief that one can undertake the actions needed to have the expected (or desirable) outcome in relation to impacts of the tumour and/or related treatments.

These are also linked to Bandura’s concepts of personal agency, proxy agency (through the efforts of other individuals) or collective agency (group effort). The assessments and interventions associated with HNA programmes have a focus on moving from a proxy (professionally directed), to moving to joint decision making (professional with individual +/- family) to, ultimately, where possible, empowering the individual. This theory seems to have multiple components of relevance to help support theory generation.

Aspects of Social Cognitive Theory were important in examining how the different agencies can support behaviour responses to a positive or negative outcome when an intervention was introduced. This offers a framework to consider how these could give insight into how each of these ‘agencies’ might interact with interventions, as well as the context to move towards improved self-management and other relevant outcomes.

However, while undertaking this review an additional theory was published, the Theory of Symptom Self-Care Management for Adult People with Cancer (Baydoun et al., 2018). This builds on Social Cognitive Theory and its authors highlight that existing theory use for cancer self-management is focused on symptom management and self-care, rather than self-care management. They also cite the increasing emphasis on empowering patients, shared decision making and improving survivorship care. In terms of a useful theory in this review, the potential mechanism of self-efficacy and self-regulation are important – not only as moderators of behaviour, but also as there is some evidence that cognitive changes can have an impact on self-efficacy. Although this theory is new and still requires testing and refinement, it has the potential to help guide interventions to ensure that patients are active partners in their management and through this adherence to beneficial behaviours can be improved.

These theoretical frameworks provide a prompt to consider initial chains of inference but were also considered in later phases of analysis and exploration.

4.7 Initial steps – from 'Initial Programme Theories' to 'Candidate Programme Theories'

To refine the initial steps of this review, a robust process using realist methodology was followed to arrive at the Candidate Programme Theories presented in Table 11.

Table 11 – Process for arriving at Candidate Programme Theories

Step 1: Action: Scoping searches, preliminary data analysis, component analysis of initial theory consideration Purpose: Identifying outcomes, mechanisms and contexts, and linkages	This first step involved searching and collating relevant papers, in this case, reviews and policy documents relevant to this practice area. These were read and re-read and data extracted which might contribute to the programme theory areas. There was a particular focus on identifying the positive and/or negative outcomes that might occur for the patients. The outcomes of interest focused on the patient/caregiver are self-management, self-efficacy, coping, and reducing distress. Based on these outcomes, contexts were examined and the interventions, responses and reactions which may lead to the identified outcomes were considered. The middle range theories selected helped to formulate these, in particular Bandura's Social Cognitive Theory. Appendix 13 provides these initial theories and the sources of information which informed each.

Step 2 Action: Review of data Undertake consultations with experts Propose and prioritise candidate theories for refinement Purpose: To refine linkages of CMOs to provide candidate theories to take forward	The relevant components (CMOs) were considered, with input from the advisors and the literature reviewed further (when necessary). At this stage, the contextual factors which were positive or negative influences on the outcomes were clarified. These ‘contexts’ included the patient, caregiver circumstances, or service and provider characteristics that might affect actions and decisions when the interventions are introduced. The focus was on ‘contexts’ which allowed certain mechanisms that resulted in outcomes to activate or ‘fire’. The literature specifically for brain tumour patients with HNA is limited so, whenever possible, these were considered in expert discussions and through the knowledge of the author. Expansion of this is detailed in the section below. Based on this analysis, this process resulted in the candidate theories. Those that focused on relevance to self-management were prioritised for further refinement.

The first step resulted in ten initial programme theories based around CMO configurations which influence the success or failure of HNA programmes to facilitate self-management (Appendix 13). The components within each initial programme theory have been organised under their 'response' mechanism or 'resource' mechanism (Dalkin et al., 2015). Each initial programme theory was supported by behaviours proposed in policy or suggested through literature and research.

These initial programme theories were then presented to five experts or stakeholders. They included a brain tumour clinical nurse specialist, a cancer network clinical lead for HNA programme implementation in Scotland, the evaluator for HNA programmes in Scotland, the director of studies (DoS) for this PhD, and a realist methodology expert. The DoS and evaluator for HNA in cancer in Scotland were done jointly and the others were done individually. The experts were asked for their views on the initial programme theories with scrutiny of each element of the CMO. This provided a 'sense check' and questioned the chains of inference between context, mechanism and outcomes. During this consultation comments were sought to refine, extend, and prioritise them. In addition, feedback was obtained to prioritise the most influential initial programme theories to guide the selection of the candidate theories. As part of this process, a few of the initial theories had similarities. These were considered together with a focus on the outcome of interest. An example of the notes from one of these meetings that included the DoS and HNA evaluator can be seen in Appendix 14.

Finally, two theories were not taken forward with the first focused on the burden of need.

If patients present with many needs (C) – particularly high levels of psychosocial needs causing a significant burden and discomfort (M - response) – this will have a positive impact on adherence (O) and uptake of interventions (M - resource) improving patient activation (O).

The burden of symptoms in this group of patients has been demonstrated to be higher than other cancers and result in significant distress (Ford et al., 2012).

Therefore, this theory was not taken forward, due to the significant level of need and higher levels of distress previously discussed for brain tumour patients. Adherence and activation, which can be influenced by need or distress, may not be as variable, as most brain tumour patients have high levels compared to other groups of cancer patients.

The second theory that was not taken forward was focused on the use of peer support for development of self-management.

> Peer-supported (C) self-care interventions (M - resource) may show efficacy in improving outcomes through building trust relationships (M - response) based on shared experiences that allow a forum for exploration and validation of options (M - resource) that may have a positive outcome (O).

This has been a widely used platform for self-management and care of chronic diseases, with many interventional strategies developed around it. However, there can be barriers in this patient group due to communication challenges and mobility problems which limit the application of interventions which harness peer support. The relevance of this to brain tumour patients may be less of a priority.

Finally, the four reformulated theories were refined through a tutorial with a realist methodology expert. The purpose here was to provide a confirmatory check around the CMO configurations with the correct ‘fit’ for each of the elements. For example, are the mechanisms actually mechanisms (as opposed to outcomes) and phrased in an appropriate way? HNA, as previously explained, is a complex programme starting with an assessment that leads to various responses dependent on the identified need. To give a manageable and clear focus to this review there was a focus on theories that related to needs assessment (but not to the exclusion of other components of this assessment) and how these can improve self-management.

These are represented in Table 12 below with four candidate programme theories which have been evaluated as the most relevant to the questions posed by this review. The table includes a title for each theory area, an explanatory paragraph and then the programme theories, coded with the proposed contexts, mechanisms (with ‘resource’ or ‘response’ added) and outcomes. This will provide the basis for the next phase of theory refinement and interrogation.

Table 12 - Presentation of candidate programme theories

Candidate theories – How can self-management with HNA be supported in brain tumour patients	
Bandura Agency categorisation	
	Candidate theory (for interrogation and refinement)
Proxy	Disease-specific HNA as a mechanism to legitimise needs and support shared decision making/problem-solving The use of a disease-specific HNA can help individuals to voice their problems (in brain tumours these are often neurocognitive problems not seen on general HNAs). Giving the individuals a platform to help identify their most significant problems, with practitioners then able to direct discussions towards these, can increase engagement and underpin shared decision making around strategies that might relieve these problems. This in turn can lead to self-management. This is achieved through developing the patient's feelings of confidence in articulating issues and then receiving responses which reinforce the importance of their voice in deciding how these problems can best be managed.

	1. Patients with brain tumours have needs specific for this condition (C). When practitioners use a brain tumour-specific HNA that supports individuals in the identification of their most significant unmet needs (M - resource), this can lead to a feeling of being legitimised (M - response) and help the individuals and their families to direct discussions towards their most significant problems (M - resource). This process can increase the willingness of patients and caregivers to engage (M - response) and develop confidence (M - response) in finding strategies to alleviate problems through a joint approach in solving them (O).
Collective	Caregiver distress and problems – unique challenges of brain tumour caregivers in supporting self-management Significant neurocognitive problems in the patient with a brain tumour sometimes have more of an impact on the family/caregiver (i.e., distress, increased financial burdens, becoming a caregiver, managing personality changes). Additionally, this can be exacerbated due to a perceived 'loss of the person' as they used to be prior to their brain tumour. Caregivers often need to have a significant role in managing care. However, they may not be invited to engage fully in the sharing of care decisions or contribute to problem-solving by healthcare professionals. But, without their support, many strategies may be unlikely to succeed. Input from the caregiver in any self-management plans is needed for them to be manageable. This input, as well as listening to the caregiver, may support feelings of empowerment to 'rise to the challenge' of caring, support self-management and minimise negative emotional responses. 2. Family members or caregivers often feel a greater sense of distress and anxiety than individuals with the diagnosis (C). This is partly due to the impact of a significant caring burden (C), but they may also feel distress at neurocognitive changes in the patient, such as personality changes, memory loss or communication challenges. This may make the caregiver feel that the individual diagnosed with a PBT is 'not the same person' (C). A focus on the patient within the context of a patient-centred healthcare system may mean that a

	caregiver’s needs or capacity for caring are not assessed (C). Caregivers may feel emotionally supported (M-response) through early and continued involvement in brain-specific HNA, followed by care planning and support for self-care (M - resource) that also is supportive of their personal or caring needs (M - resource). This helps caregivers to rise to the challenge (M - response) and develop their capacity to support self-management (O). This also helps to prevent detrimental impacts on their wellbeing (O) and ‘carer burnout’, where formal care services would need to increase support (O).
Individual	The role of self-efficacy and activation in positive outcomes within HNA programmes Individuals and their families will have pre-existing levels of self-efficacy and activation which may manifest in differing abilities to understand successful self-management, what barriers to self-management they face, and what strategies they employ to manage their condition and to cope with stress. This can be at a very low level, where individuals may (or may not be) compliant and being asked to solve problems may increase stress. Where there are higher levels, individuals may actively solve problems and have the confidence to share decision making over options or solutions that may address them. Practitioners need to assess and understand a patient’s activation level, their level of knowledge, skills and confidence to manage, and ability to solve problems. This will help to support self-management to an appropriate level, which minimises anxiety. 3. Patients and their caregivers will have pre-existing levels of self-efficacy and of activation (readiness for change) (C), which can influence confidence in shared decision making (M - response) and facilitate problem-solving to self-manage (O). When undertaking a disease-specific HNA, healthcare professionals invite patients and caregivers to participate in HNA-related planning and discussion (M - resource). If practitioners assess the

	activation as part of the HNA discussion, based on their perception of the individual/caregiver's current situation (M - resource). They should then provide a personalised approach depending on level of readiness (M - resource), this supports the engagement of the patient/caregiver (R - response). The level of input invited or sought over decisions around care and self-management. What levels of support are needed can be gauged to an appropriate level to reduce feelings of anxiety and/or build confidence (M - response). It can also increase problem-solving by patients and caregivers' and adherence to self-management strategies (O).
Collective	Practitioners as a barrier or facilitator to successful implementation of HNA Practitioners may be resistant to using or fully implementing HNA programmes for a variety of reasons. They may perceive that such programmes do not improve communication or assessment of problems, they may not see it as their role, or they do not have the knowledge or resources to support identified problems when elicited. Introduction and delivery of HNA programmes without supporting and training clinicians may result in poor implementation. The impact of this could be that practitioners ignore or mismanage problems. This may cause distress or a lack of trust in the patient/caregiver, if they ask about problems which are ignored or not managed. However, with the use of a disease-specific HNA that provides a disease-specific assessment focusing on the unique needs of this group, the benefits in problem identification may reveal improvements over routine assessment that can challenge this belief. Through providing evidence-based education and training that includes communication skills around problem management and shared decision making and knowledge of referral pathways, the confidence and competence of practitioners may increase. This can result in improved patient-centred assessment of problems and provide the basis

	to have shared discussions (with the degree of shared decision making gauged for each patient) about solving them through care strategies, appropriate referral and self-management strategies. 4. Practitioners may have preconceptions about HNA related to the usefulness of this intervention (C), their role in supporting the process (C), and their knowledge in responding appropriately to patients' holistic problems (C). This will influence their engagement, implementation, and their attitude to undertaking HNA (M - response). A brain-specific HNA focused on the relevant problems of this patient group, that supports a holistic assessment of relevant needs (M - resource), can build acceptance of the benefits (M - response). If practitioners are provided with knowledge of appropriate support and referral options (M - resource), as well as training in shared decision making to respond to problems (M - resource), this can improve the confidence and competence of the practitioner in undertaking HNA (O). For the patient and their caregivers, this can result in feelings of engagement in proactively managing their identified problems (M - response) when they are involved in decisions about solutions to alleviate their needs (M - resource) with practitioners, which can improve their confidence in self-management (O). Alternatively, if the practitioner has negative perceptions about the HNA process (C), and suboptimal delivery results in needs being ignored or mismanaged (for example, with solutions that do not work for the patient/caregiver) (R - resource), this can result in feelings of disengagement (O) and distress (O).

C = context, M = mechanism, O = outcome

4.8 Iterative literature search strategy and selection of articles

The next stage of this review involved the interrogation of the four candidate programme theories for further refinement. A purposive and iterative literature search was undertaken. The literature had to be relevant to part or all of a theory to be considered – and ideally, the subject of the literature would be focused on all key elements; patients with a *brain tumour,* exposure to an *HNA* and how this impacts *self-management.* However, as previously discussed, the literature is limited, so there were three key areas of search – one was a broad search on brain tumours and HNA, one search focused on self-management and HNA and cancer, and one focused on brain tumours and self-management. The evidence which informed the initial programme theory generation was also reviewed for additional information which could provide refinement of these theories, particularly studies from the reviews reported in Appendix 12. The following paragraphs describe the first steps – however, the literature search continued throughout all stages of this review to allow specific interrogation into an element of each theory. A list of relevant and related search terms was produced (Rycroft-Malone et al., 2012) – however, many papers contributed to theories in more than one area.

The dates were limited to literature from 2007 to 2020, as programmes that integrated HNA into cancer care were not well established before this. With the establishment of these national policies, the importance and emphasis on more holistic care – including non-physical problems and particularly psychological problems – became more widespread.

In the UK, this was first integrated through the national survivorship initiative in 2007 (Macmillan Cancer Support, 2015), although some cancer centres were using the distress thermometer and problem checklist for some time. In Canada, in 2008, key national agencies agreed a countrywide approach to distress management, which included the need to complete the problems checklist (as well as the distress thermometer and Edmonton Symptom Assessment System) (Bultz et al., 2011). Wide-scale adoption of distress screening using a problems checklist was not fully adopted in the US until 2012 (National Comprehensive Cancer Network, 2019).

The first search was conducted using the electronic databases Medline, Cumulative Index to Nursing and Allied Health Literature (CINAHL), Cochrane and PsycINFO. The search terms used were Cancer OR Neoplasm OR Tumo*r* or Oncol* AND Brain OR Neuro* OR Glioma AND SCNS OR "Supportive Care Needs Survey" OR “Patient Concerns Inventory” OR “Pepsi Cola Aide Memoir” OR “distress thermometer” OR problems checklist OR Concerns checklist OR holistic needs assessment OR "HNA" OR "Sheffield profile for assessment and referral for care" OR Needs Assessment OR Symptom Assessment OR Self-Assessment. There were 1006 articles retrieved for review. In this search the names of commonly used HNA tools were included as listed above.

The second search was conducted using the electronic databases Medline, Cumulative Index to Nursing and Allied Health Literature (CINAHL), Cochrane and PsycINFO. The following search terms were used: need* OR concern OR *problem* AND holistic needs assessment OR HNA OR problems checklist OR “Supportive Care Needs Survey” OR “SCNS” OR “distress thermometer” AND cancer OR tumo*r* OR neoplasm AND self-care OR self-manage* OR self-efficacy. There were 96 articles retrieved for review.

The third search was conducted using the electronic databases Medline, Cumulative Index to Nursing and Allied Health Literature (CINAHL), Cochrane and PsycINFO. The search terms used were: Cancer OR Neoplasm OR Tumo*r* OR Oncol* AND Brain OR Neuro* OR Glioma AND self-care OR self-manage* OR self-efficacy. There were 150 articles retrieved for review.

The fourth search focused on the practitioner’s role in HNA using the search terms Needs Assessment OR Symptom Assessment OR Self-Assessment OR Patient Outcome Assessment AND inventory OR tool* OR measure* OR instrument* AND neoplasms OR oncology OR cancer AND nurse OR doctor OR medical staff OR clinician OR allied health professional OR AHP AND attitudes OR perceptions OR opinions OR thoughts OR feelings OR beliefs OR experiences. There were 175 articles retrieved for review.

Nine additional sources also contributed to the theory refinement. This literature was obtained through previous research retained in earlier stages of this thesis – grey literature linked to policies such as evaluation reports of HNA in various regions and reviewing reference lists of retrieved literature which had relevance to the theory. Citations of relevant systematic reviews of cancer HNA tools were also followed up for potential research studies. Titles and abstracts were reviewed – if there was a consideration that they could contribute to theory refinement they were retained for a full-text review. The study details were extracted according to the data extraction form in Appendix 15 and the original literature in many cases was reviewed again as chains of inference evolved.

4.8.1 Process of selection, appraisal and data extraction

A realist review has several fundamental principles that are key differences from a traditional systematic review. These are important to highlight (Rycroft-Malone et al., 2015).

- The involvement of stakeholders is vital in the development of programme theories – so not everything will be reflected in the literature
- The search for evidence is purposive and theoretically-driven with the aim of refining theory. This will also be relevant to the appraisal and is often bespoke based on the focus of the theory
- Many different types of evidence or data sources may be included
- The process is iterative and may change throughout
- The outcome is explanatory – the findings should explain to a reader why or why not a particular intervention may or may not work to help enable decisions that inform further practice or research

There are challenges in this process, particularly for the novice researcher, due to its flexibility. The synthesis process uses a variety of sources, which may include research studies of any design, but also policy documents, opinion documents and other grey literature. The first challenge is in deciding the relevance or the boundaries of what is useful in advancing the theories. For example, does a study which demonstrates an HNA programme in breast cancer survivors has successfully shown higher uptake of a specific self-management activity, have relevance? Or are the contextual factors too different to have significant value?

In terms of this study, initial theory development was informed by policy and grey literature. However, refinement (this stage) will be informed by peer-reviewed literature as the knowledge of how policy makers hope this programme 'works' has been considered in the initial programme theories.

The second challenge concerns the rigour of the data being used and what questions should be asked about its quality. As Pawson argues in his paper *Digging for nuggets: How 'bad' research can yield 'good' evidence*, he provides a criticism of critical appraisal for realist syntheses, particularly concerning qualitative data (Pawson, 2006a). The paper proposes that a consideration of the rigour of the whole research process and looking at a study as a whole using a structured critical appraisal framework is irrelevant. In a realist review, it is unlikely that the whole paper will be testing the programme theory. Instead, a portion of that paper may provide a 'nugget' or contribution to a part of a programme theory. In this paper, Pawson concludes for all research that: “The only feasible approach is to make the appraisal in relation to the precise usage of each fragment of evidence within the review. The worth of a study is determined in the synthesis” (Pawson, 2006a, p.141).

To address these two challenges, the following approach was taken in reviewing data which could support the refinement of the proposed candidate theories. Data to be considered needed to inform at least one theory area with a contribution towards the linkages of contexts, mechanisms or outcomes proposed in an HNA programme *(or similar programmes)* and development of self-management *(or related concepts).* For example, similar programmes that assess symptoms to improve self-efficacy or coping might be relevant. In all cases, the quality appraisal was not formally undertaken, but principles such as those outlined in the Critical Appraisal Skills Programme (CASP) quality appraisal process were considered where appropriate (CASP UK, 2018). So, if it appeared that a finding of a qualitative paper that used thematic analysis appeared to contribute an explanatory framework for a candidate programme theory, the following question from the CASP quality checklist for qualitative research would be considered. Any relevant quality concerns were considered as part of data extraction. This is indicated in the summary of the literature in Appendix 15.

The following levels of relevance were applied to help in the decision-making process for papers selected for inclusion in the analysis.

High

This category is for papers with high relevance to the candidate theories of the realist review. These typically include studies or other literature that focus on brain tumour patients' context of an HNA programme *(or similar programmes)* and the development of self-management *(or related concepts).*

Moderate

This category is for evidence that has some relevance to theory areas but might have a different framing. For example, it could be an HNA package for cancer which looks at improving self-management, or interventions in brain tumour patients with the aim of improving coping or self-efficacy. There should be linkages of some aspects of the C/M/O in any combination that could enrich the theory output.

Low

This category is for evidence that appears to have some relevance on first reading but links between the C and M (for example) are not entirely clear. However, it may have rich information on a certain aspect (such as context) and some ideas that may be useful for conceptualising theories. This could be a paper looking at symptoms and sequelae of brain tumours or focused on exploring the challenges of self-management but does not have an intervention or mechanism to provide an explanatory framework for a programme.

Exclude

This is for papers that demonstrate some promise on reading the title and citation but do not have any content corresponding to the candidate programme theories (the data will not be extracted for this, but a list of excluded papers from this stage will be retained).

The appraisal of the literature used to refine the programme theories is represented in Appendix 15. These forms summarised potential contribution to theory generation based on the realist framework of relevant context, mechanism or outcomes with their possible linkages or chains of inference (Rycroft-Malone et al., 2015). The guiding principle in this stage was; What is the intrinsic logic of HNA? or, Why is this assumed to be a beneficial intervention? This appraisal included an overview of the study and consideration of some comments about the trustworthiness or the rigour of the research. It also contained an assessment of the usefulness and contribution to programme theories, looking at the relationship of study components that had relevance. This data extraction also contained some analysis of literature that could contribute to overall interrogation and refinement of the programme theories. For example, it could add to the potential CMOs in play or combinations of various iterations of these, such as the context and outcomes.

The appraisal of all papers was combined to synthesise the body of evidence with each of the candidate programme theories. A summary of the literature used to inform this can be seen in Appendix 16. This was done through seeking demi regularities in more than one piece of evidence or linkages of chains of inference that might be relevant. These were interrogated to look for confirmatory or alternative evidence to refine further and give insight into how these complex interventions and components may work (or not). This analysis provided refinement and focused on determining what contexts might trigger mechanisms to fire. The analysis focused on how this might support the success or be to the detriment of the interventions, relevant to the programme theories (Wong et al., 2013).

4.9 Findings of purposive searches

The full text of 152 papers was reviewed and data was extracted from those that were relevant using bespoke forms based on study characteristics and 'theory synthesis forms' for the different data types (see Appendix 15). Searches and theory development were undertaken from December 2018 to March 2020. In total 46 papers were used in the final theory refinement.

4.9.1 Analysis

On examining the available literature on this topic, it was clear that there was limited evidence to explore how HNA in brain tumours could support self-management. However, there was relevant literature from similar areas that could contribute to chains of inference to help explain these theories. Brain tumour studies of HNA (not focused on psychometric testing) were included, but also, where relevant, literature that looked at interventions linked to self-management or self-efficacy in the context of supportive care needs in PBTs was considered. There was a wide array of literature looking at HNA programmes in cancers more generally and several articles that focused on specific cancers, mainly prostate and breast. This was synthesised to refine the theories using an iterative process based on an appraisal in relation to its usefulness. The process used in the individual appraisal of each article is illustrated in Appendix 15 and the contributions in Appendix 16.

Analysis, interrogation and refinement of each of the programme theories is discussed in the following sections. The theories were interrogated through 'chains of inference', first examining links to the contexts (mechanisms and/or outcomes) and then from the view of outcomes (with any links to mechanisms and/or contexts). Where possible, multiple sources which supported chains of inference (demi-regularities) were sought. However, in many cases, there was a paucity of evidence in some aspects of brain tumour research. Analysis was guided by the underlying premise introduced by Pawson and Tilley (1997) – that mechanisms which work or do not work are determined by context and mechanisms are a combination of the resources and the stakeholder's responses to these. The consideration of resource and reasoning as disaggregated components of mechanisms (Dalkin et al., 2015) also guided this process. Finally, missing components (of CMOs) relevant to each programme theory were considered. If they appeared valid, they are discussed below. Support of the existing theory, refinement or new theories were searched for and considered within each element of the programme theories. This analysis is detailed in the following sections.

4.9.1.1 Analysis of theory area 1 – Disease-specific HNA as a mechanism to legitimise needs and support shared decision making/problem-solving

Patients with brain tumours have needs specific to this condition and this context is complex and multifaceted. The contributing factors are likely to include a high symptom burden, increased levels of distress, poor prognosis (in a large proportion of patients), neurocognitive deficits and relative rarity and heterogenic symptom profile based on the tumour location (Ford et al., 2012). This combination of factors presents quite different contexts to other cancers. Support for using a disease-specific HNA, rather than a generic HNA, is linked to this as, in this context, a disease-specific HNA could reveal the most troublesome problems and help realise more positive outcomes. One area identified as a problem for HNA is when it does not ask the right questions – distressing problems are not detected if there is a lack of systemic questioning (Ahmed et al., 2014). In studies of long-term survivors of brain tumours, up to 75% report needs related to cognitive changes (Sloane et al., 2016). Previous chapters have provided support for this but, to summarise, the needs and symptoms of brain tumours are often not represented on generic cancer HNA tools. Simplicity and/or adaptability may be needed to support completion.

These contexts also often appear in combination. A study examining the correlation between distress, unmet meets and HRQOL, measured by the distress thermometer, Supportive Care Needs Survey (SCNS) and a brain-specific HRQOL (EORTC-BN20) (Hickmann et al., 2017) found that the presence of even low levels of cognitive impairment (based on the HRQOL) was the most highly correlated factor for distress. Unfortunately, the HNA did not utilise the brain tumour SCNS, which might have provided even more information about this relationship with unmet cognitive needs (Janda et al., 2006). The authors stress that these cognitive changes do not necessarily correlate with measurable cognitive deficits but rather are related to the patient's perception and impact of these on their life, such as their emotional or social functioning. They highlight that this group of patients should be carefully screened for their subjective perception of cognitive problems to deliver supportive interventions and mitigate the psychological impact. The authors feel that the generic HNA does not identify neurocognitive problems and the brain SCNS was not mentioned, so it may have been that it was not validated for this population (German). Another study which used the same tool – but with the addition of the brain tumour-specific questions – found that at both baseline (within three months of diagnosis) and then three months later, one of the top five concerns of patients was 'not being the same person' (T1 = 50%, T2 = 47%) (Langbecker & Yates, 2016). An HNA that identifies these can be a platform to direct discussion towards these issues.

High levels of need may also present challenges to the HCP in supporting this group. PBT patients have many significant needs across all domains. In one study, 23% of patients did not feel adequately supported (Renovanz, Maurer et al., 2018). Even among those patients who did say they were supported, many felt they would like more support from various healthcare professionals for specific problems, often neurological. This finding was not surprising as many patients reported needs that potentially could have been helped through that referral. However, without the identification and care planning for these needs, it does not appear to have occurred.

The process of seeking the patient's subjective assessment of their needs does help in supporting them, directing discussion and legitimising their concerns. The evidence across many cancers seems to support use of the HNA to give patients a voice or platform to raise their problems (Clarke et al., 2019; Green et al., 2017; Heyn et al., 2013; Johnston & Campbell, 2018c). There is also evidence that this allows greater exploration of emotional concerns and also gives more focus when the patient raises concerns in a consultation (Heyn et al., 2013). Providers may feel that this is of benefit to patients, with nurses in one study reporting that it empowers patients and allows a focused consultation on issues or problems which might not have been covered (Rogers & Lowe, 2014). This was also supported in a study of breast and colorectal patients, in which 20% in a normal care arm (with HNA and other patient reported outcome measures (PROM) completion) said that the process of completion helped communication with their clinician (Girgis et al., 2009).

In the view of nurses, use of an HNA might 'normalise' problems for the patient’s condition and the process itself might be beneficial and ease distress (Børøsund et al., 2014). The nurses also reported that, especially in 'quiet' patients, this allowed disclosure of problems they would not have suspected in that person. The provision of disclosure was described as an important benefit of HNA. A crucial beneficial area highlighted was the opportunity for reflection through completing a questionnaire focused on potential issues before seeing a healthcare professional. This allowed time to consider what actions might help potential issues and then the patient could be more involved in the discussions with on how to solve that problem. This analysis revealed chains of inference that could be considered, such as a disease-specific HNA providing a platform for reflection and an invitation for those who might struggle to disclose their problems.

A disease-specific HNA seemed to help identify related problems and seeing a symptom listed in the HNA confirmed to patients that their symptoms could be ‘normal’ for their condition (Børøsund et al., 2014; Johnston & Campbell, 2018c). The nurses described how this seemed to make the problems less frightening and that the patient sometimes felt relieved, even without a follow-up conversation (Børøsund et al., 2014). However, negative feedback from patients was related to a lack of a follow-up conversation after the assessment – as a bare minimum, nurses needed to acknowledge the information provided by the patient. This could also be beneficial for more sensitive issues –patients may (due to cultural considerations) be reluctant to highlight sensitive or private issues (such as sexual problems). The structured format of a questionnaire allows these to be broached (Rotenstein et al., 2017; Thewes et al., 2016).

The lack of a follow-up conversation could also result in feelings of betrayal (or breakdown of trust) and this can lead to increased distress (Biddle et al., 2016). A quote from this paper illustrates how one individual would feel, concerning non-engagement of the HCP:

> 'I definitely would expect someone to address the issue because otherwise I would feel betrayed… people have to be quite careful about asking questions if they don't want to deal with the answers' (p.10).

The literature had some interesting insights into how this process might support shared decision making. One study focused on HNA in patients with a variety of cancers. It found that, once the patients had identified their symptoms and had an opportunity to reflect on them in advance, nurses reported that they were more engaged in the conversation and time was put to better use to determine how troublesome the problems were and what to do about them (Børøsund et al., 2014). Alternatively, Clarke et al. (2019) reported that many patients in their study of prostate cancer preferred a paternalistic model of care rather than jointly decided actions. UK policy advocates shared decision making (NHS England, 2015; Wojcik, 2018) to replace paternalistic models of care. However, we do need to help patients in how to have these discussions and use provided information to empower them to make informed choices about their health. One perspective comes from a realist evaluation of Transforming Care after Treatment (TCAT), that looked at 25 local projects across Scotland which encompassed elements of the recovery package, 19 of them incorporating HNA (Johnston & Campbell, 2018a). One of the theories postulated was that HNA could help cancer patients 'see a way forward' in managing their problems and reduce dependency on the 'system'. It appears that this did improve the ability of some patients to manage, but the evidence for decreased dependency was not clear. This may support the idea that across all cancers, there will be challenges with self-management – and this might possibly be due to seeking a paternalistic model of care.

There have been some studies examining this in patients with brain tumours. In a study of glioma patients and their caregivers, which focused on rehabilitation and self-management, Piil et al. (2015b) reported that the participants undertook self-management actions that they hoped would optimise treatments, particularly in the early phases. Support from HCP was vital to them but they often felt it was lacking. The reason could be that what they had considered as a solution lacked evidence of efficacy – but from the perspective of the caregiver and patient, the role of these in 'giving hope' was important (the use of complementary therapies was the most common strategy). This possibly does present a challenge to HCP when they are evidence-based professions. The conflict of supporting something without this evidence, and that may also have costs associated with it, may be perceived as unethical or outwith their knowledge base. However, early assessment through HNA, dialogue and engagement can support a more joint approach, even to provide information about what is known or not known about various self-management strategies.

Another critical consideration is that patients with a brain tumour often have diminished cognitive function and there can be challenges in retaining information and advice (Cavers et al., 2013). This was also found in a study of glioma patients in which 39% required assistance to complete their questionnaire (Renovanz et al., 2017).. This author advocated that questionnaires also need to be designed (or services designed) that take account of these significant cognitive problems These cognitive difficulties were also noted in a study of supportive care needs in patients diagnosed with a glioblastoma or brain metastasis.(Seekatz et al., 2017). A large proportion of patients had significant cognitive deficits that restricted their ability to use an HNA (Seekatz et al., 2017) and the authors recommend that caregiver proxy needs to be considered. Although some research does demonstrate high congruence on some aspects, such as symptoms (Armstrong et al., 2012a), this may not be reflected across all of the holistic needs (Sloane et al., 2016). However, the alternative view that in this situation there may be an opportunity to take the caregiver’s viewpoint of their perceptions (and challenges) will be discussed in the next section.

The literature supports HNA as a mechanism to legitimise needs and focus on them in discussions and consultations, helping not only the patients and caregivers, but also the providers of care. However, there does seem to be a lack of evidence to support this as a means to shared decision making. Improved support for individuals to participate in decision making needs to be developed. This does remain a challenging area, as demonstrated by a Cochrane review in 2018, which was unable to recommend any specific interventions to support shared decision making (Légaré et al., 2018).

4.9.1.2 Theory 1 – Refinement

This theory remains important (See Table 12 for original candidate theory). In the context of complex neurocognitive needs, a disease-specific HNA that captures the perceived needs of patients with neurocognitive problems to legitimise these, direct discussion, and support a problem-solving approach, remains an important aspect of this programme theory. However, contexts and mechanisms that could lead to negative outcomes are one aspect that emerged as an area for refinement. With PBT patients, the impact that neurocognitive problems may have on many aspects of this programme also need to be considered, such as challenges with the assessment process and how these problems can affect engagement, confidence and problem-solving. Finally, when supporting patients with multiple complex problems who may also have a poor prognosis, maintaining hope and trust can be crucial in minimising distress. The refined theory is represented by Figure 9 below and changes to candidate theory are denoted by the red text.

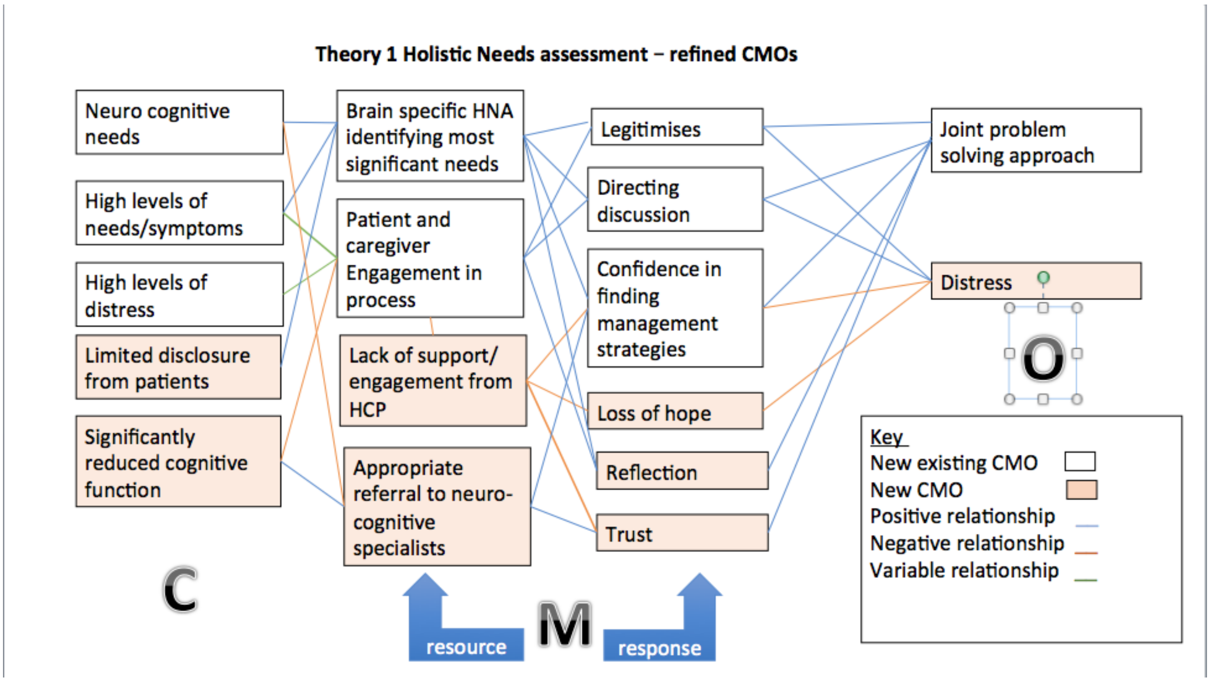


Figure 9– Refined Programme Theory 1

Patients with brain tumours have needs specific to this condition (C). When practitioners use a brain-specific HNA that supports individuals in the identification of their most significant unmet needs (M - resource), this provides a subjective assessment (M - resource) of the perceived problems, particularly cognitive impairments (C). This can lead to a feeling of being legitimised (M - response) and help the individuals and their families to direct discussions towards their most significant problems (M - response). Discussion prompted through an HNA (M - resource) may also support disclosure in patients who would otherwise be reluctant to do so (C) or, in other patients who seek to be involved in problem-solving (C), allow them to reflect (M - response), becoming more able to engage. The presence of significant neurological problems (C) is likely to benefit from referral to appropriate neurocognitive specialists. This can improve the problem-solving strategies (M) and build trust (M). This can also increase the willingness of patients and caregivers to engage (M -response) and develop confidence (M - response) in finding strategies to alleviate problems through a joint approach in solving them (O). Conversely, if an HNA is used (M - resource) and the practitioner does not acknowledge the problems (M -response), this can lead to a loss of trust (M - response) and increase distress (O).

4.9.1.3 Analysis of theory area 2: Caregiver distress and problems – unique challenges of brain tumour caregivers in supporting self-management

Caregivers can have significant unmet needs and levels of distress that are often higher than the patients themselves and there is widespread evidence to support this. This contextual factor is likely to be a significant determinant of individual responses to HNA and supporting self-management for the patient and themselves. A study which examined the unmet needs of caregivers of high-grade glioma patients over a six-month period – commencing during their concomitant chemo-radiotherapy – found that these were at significant levels, with 59% having at least five unmet needs (Halkett et al., 2018). In another study using the same needs assessment, that looked at caregivers for all cancers, comparatively fewer at only 29% had five elevated needs at six months (Girgis et al., 2013). In addition, this study found that the highest number of unmet needs correlated with the most distress and psychological morbidity (Halkett et al., 2018). However, these authors did highlight that the presence of distress may predicate having more needs, as opposed to more needs causing distress, and, potentially, characteristics such the presence of resilience may be a moderating factor. Finally, within the specific needs – which were collected at three time-points over the six-month period – the most common was related to the impact of caring on their working life or normal activities. The most common brain tumour-specific supportive care need was information on adjusting to cognitive changes, which increased over time from 33% at baseline to 38% at six months. This high level of distress and unmet needs does seem to indicate that a focus on these issues is important to support caregivers in continuing to function in their role. In the same study – but reported in a different paper (Halkett et al., 2015) – it was found that patients with a partner who was also their caregiver had significantly lower emotional wellbeing. This was an unexpected finding – however, the authors contribute two hypotheses to be considered. The first is the patient's awareness of the impact of their illness on their partner. However, the authors also considered that when a partner was caring for an individual, there was less formal support from HCP. The authors suggest that an assessment of needs and support for caregivers could improve the wellbeing of both caregiver and patient in these contexts. Similarly, another study examining the supportive care needs of PBT patients assessed needs within three months of diagnosis and then three months later (Langbecker & Yates, 2016). At baseline, the highest level of need was ‘concerns about the worries of those close to you’ in 69% of patients. This was not in the top five at the second time point. This supports the importance of assessing and addressing the needs of caregivers, even as a potential means to reduce distress in patients.

In another study, Trad et al. (2015) examined distress and problems in PBT patients at diagnosis and recurrence and found that distress was higher in the caregiver than the patient. The authors hypothesise that the contributing factors for this increase in distress may be due to the additional decisions to be made regarding palliative care options, increasing symptom burden (physical, cognitive) and financial stress. This supports the importance of also assessing the caregivers’ needs and capacity for caring, which could help to mitigate distress but also provide the resources and information that match the carer’s needs. This could also support the suggestion that early involvement of the caregiver in the process is important, as it could result in early dialogue about future needs that have patient involvement when they are able to have these discussions in the event of a patient’s neurocognitive decline.

Seekatz et al. (2017) report that caregivers of patients with PBT or brain metastasis who have higher levels of distress in the early stages after diagnosis are more likely to be in contact with the specialist palliative care team. However, this contact did not seem to have an impact on the symptom burden of patients. However, this may have been due to the disease trajectory causing worsening symptoms and the study was not set up to measure if this was influenced by the palliative care support. This study also showed that caregivers (76%) were more distressed than the patients themselves (62%). Caregivers with high levels of distress correlated with being a carer for a patient with high levels of cognitive changes. Interestingly, this study also showed that marital status was not a predictor of the need for additional specialist palliative care support. It could be that, while the authors were seeking knowledge of the living situation, they did not ask about other support or if the spouse supported their care and that may be more important than marital status. Additionally, socio-economic status or nationality did not play a role in the need for support either. This, however, may be highly context specific. This study was in the German healthcare system and would not necessarily translate to other health care systems or culture groups.

In a study which undertook longitudinal interviews with carers, one spoke of being ‘bound’ to the disease and having no break from it or opportunity to pursue any of their interests (Piil et al., 2015b). Others discussed the constant caring and increasing burden of responsibility often related to the cognitive changes and deterioration that the patient experienced. One study (Cavers et al., 2013, p.1301) has a particularly affecting quote:

> “It’s made it sort of unbearable sometimes. [..] It’s hard to imagine being able to live the rest of my life with somebody with that kind of temperament” (Sharon, wife of Ian, 46-year-old male, GBM patient).

This discussion of the impact on this caregiver about changes in personality were very significant. ‘Not being the same person’ in some cases could be the most important context for a variety of reasons. In this study, the caregiver highlighted that, at times, they needed to be the witness to what was happening with the patient. For example, the patient would say that everything was fine and they were coping well with normal activities but this did not reflect what was happening (Piil et al., 2015b). The caregiver had to manage the situation but also ensure that they were the history keeper of the care, to make sure their loved one got the best. Another challenge highlighted by caregivers which caused stress was the loss of the ability to share concerns or exchange views on the situation, which was often linked to memory loss. This potential isolation and emotional burden highlight the need to care for the caregiver – ‘rising’ to the challenge is considerable and the breakdown of their ability to care could have considerable impacts for the patient and the care system.

In a review of caregivers’ needs for supporting brain tumour patients, one of the most striking findings was the description of social isolation experienced by the caregiver and patient caused by personality changes and a ‘withering’ of their interpersonal relationship and roles (Madsen & Poulsen, 2011). This withdrawal from normal social interactions and support was described in one study as the patient’s ‘social death’ and this was another stressor for the caregiver.

A decline in the cognitive function, HRQOL and communication abilities of patients was demonstrated to have a detrimental impact on the psychological health of caregivers (Boele et al., 2013). This was despite intensive provision of psychological interventions for the caregiver and raises the question of what can be done to support them in these circumstances.

However, not all studies focused on the negative aspects of caregiving. One study in particular examined what might predict how caregivers perceive positive aspects of their role (Newberry et al., 2012). In patients with decreased reasoning abilities, caregivers had a higher score on positive aspects of their care. While this study was quantitative and examined correlations, the authors hypothesise that having something tangible to do may be the reason for this.

Traits such as a positive outlook and positive self-affirmation are fixed (so possibly contextual factors) in caregivers, which can help predict negative or positive responses to this role and the burdens/stressors that come with it. Those with positive outlooks may be more likely to engage in self-management, as they believe through their actions that more positive outcomes are possible. This study also highlighted the importance of caregiver assessment, as this could ultimately improve the quality of care administered at home.

The lack of focus on caregivers’ needs and capacity for caring has been discussed by a number of authors. Cavers et al. (2013), in a series of longitudinal interviews with glioma patients and their caregivers, highlighted the issue of discordance in needs, especially around information and prognosis. Caregivers spoke about not having enough information and that they could possibly have been better able to cope if they had had what they needed. These authors recommend that there could be provision for consultations for the caregiver if ethical issues are considered, for example, consent. This would seem to support that, through the provision of an HNA for the caregiver that is focused on their caring needs, their wellbeing and capacity for supporting self-management can be maximised.

In a review of caregiver needs by Madsen and Poulsen (2011) they suggest that caregivers are considered by HCPs in this role only and that their supportive care needs are not usually considered. However, they also found that caregivers would highly value the ability to contact a specialist source for advice (such as a nurse specialist) when they need help to self-manage or self-care when at home. One area of information identified as problematic was how to manage neurocognitive changes – this was a significant source of stress. However, some studies highlighted that informal channels such as support groups could be very beneficial for some people in helping to troubleshoot issues and vent frustration.

The desired outcome of HNA programmes in supporting self-management is often congruent with what families are striving for. Caregivers often seek to support the patient, manage care to keep strangers out of the home and allow the patient to stay at home. In a study that examined why PBT patients do not accept referral for support for their unmet needs, Langbecker, Ekberg and Yates (2017a) report that caregivers were often more dissatisfied with the care than the patients. They also found that one of the reasons patients cited for not using services was that they preferred using informal support, such as family or a partner. These statements were not correlated as this was not a quantitative study, but it is possible to consider that caregivers are not getting the support they need, while patients are possibly refusing more formal support, leaving the caregiver overburdened.

4.9.1.4 Theory 2 – refinement

One of the unconsidered areas in the context of caregivers was their self-efficacy. In a study examining the variables of caregiver distress, mastery, anxiety and depression, and its impact on patient survival from GBM patients recruited within three months of diagnosis, it found that mastery was correlated with survival (Boele et al., 2017). Caregiver anxiety, depression or the patient's symptom burden was not correlated in this study. The authors speculate on the reasons for this may be due to better communication between the patient and caregiver or, alternatively, that reactions to physical manifestations of the disease were proactively managed and acted on more quickly. Although this was a small study, the role of 'mastery' that the authors feel relates closely to self-efficacy seems to be a factor, which may be an important context. In caregivers who display mastery, providing information to support management of the patient can help to realise positive outcomes.

A number of studies demonstrated that caregivers have high levels of distress when caring for someone with a brain tumour, so their wellbeing should be supported. Specialist support, for example, from the palliative care team, may be accessed when the caregivers have higher levels of distress. This may mean that their capacity for self-care can only be realised if caregivers' distress is assessed and supportive measure introduced to reduce this. Boele et al. (2013) looked at a supportive intervention in caregivers of high-grade brain tumour patients, which was based on cognitive behavioural therapy and psychoeducational principles. The hypothesis was that this would improve their 'mastery' of coping as a caregiver, as well as their HRQOL. This was a small study but it demonstrated that mastery was improved (which the authors related to self-efficacy) and HRQOL did not deteriorate in contrast to the control arm. This lends some support to the part of the theory that meeting caregivers' needs, particularly psychological needs, can improve wellbeing. However, this study did have high attrition and while a good proportion was related to the death or deterioration of the patient, there was a higher drop out in the intervention arm that could have meant the intervention was too intensive. This may highlight the importance of a personalised assessment and a plan that delivers on this.

As reported previously, the impact on working life and normal activity can also worsen these effects as it can contribute to a loss of some support networks as well as increasing financial pressures. In addition, cognitive changes can be profound and correlate with distress, as caregivers are often left to manage without the support or information they feel is necessary. This is very likely to affect their ability to support self-management. There does not appear to be any evidence that this is an outcome referenced in the literature, although self-efficacy has been linked to decreased healthcare resource usage in other conditions with neurocognitive decline, such as dementia (Boele et al., 2017). This may be an area for exploration – however, there may be other outcomes relevant to caregivers which warrant further exploration, notably survival.

While there appears to be evidence to support the CMO configurations to some extent, models of care which address caregivers' needs and help to provide that emotional support need to be developed. The ability to care and manage could help to reduce use of health services but also have the capacity to prevent psychological morbidity in caregivers. The refined theory is represented by Figure 10 below and changes to candidate theory are denoted by the red text (See Table 12 for original candidate theory).

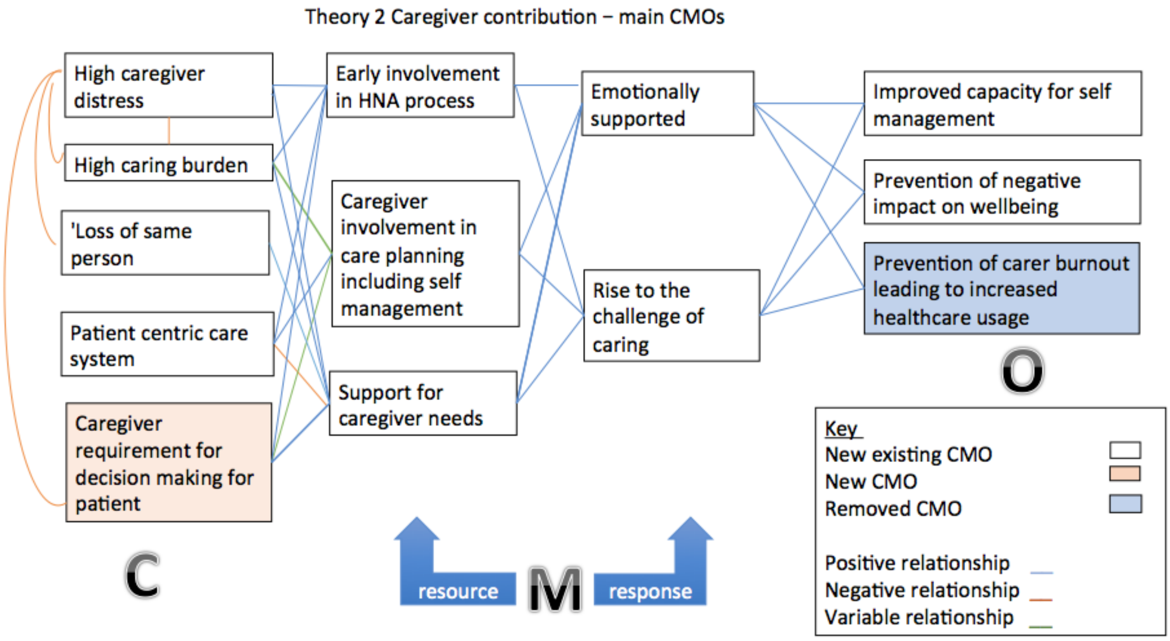


Figure 10– Refined Programme Theory 2

Family members or caregivers often feel a greater sense of distress and anxiety than individuals with the diagnosis (C). This is partly due to the impact of a significant caring burden (C) but they may also feel distress at the neurocognitive changes, in the patient, such as personality changes, memory loss or communication challenges. This may make the caregiver feel that the individual diagnosed with a PBT is 'not the same person' (C). Caregivers may also be distressed by a significant decision-making role in patients with a poor prognosis (C).

A focus on the patient within the context of a patient-centred healthcare system, may mean that a caregiver's needs or capacity for caring are not assessed (C). Caregivers may feel emotionally supported (M-response) through early and continued involvement of caregivers with the patient in a brain-specific HNA, followed by care planning and support for self-care (M - resource) that is also supportive of their personal or caring needs (M - resource). This inclusion may help them feel emotionally supported (M - response). This helps caregivers to rise to the challenge (M - response) and develop their capacity to support self-management (O), helping to prevent detrimental impacts on their wellbeing (O).

4.9.1.5 Analysis of theory area 3 – The role of self-efficacy and activation in positive outcomes with HNA programmes

This theory suggests that patients' and caregivers' abilities and readiness for shared decision making, problem-solving and self-management are key considerations. These can influence the ability to self-manage and need to be assessed in order to make plans for care, including what level of self-management might be possible for the patient/caregiver.

The only contextual factor from the candidate theory was related to self-efficacy and readiness for change. Clarke et al. (2019) looked at readiness for change among prostate patients in a study of an electronic HNA, followed by the patient making supported choices to help in the management of any problems. This seemed to be a challenge for many patients who wanted to avoid information to support decision making and preferred paternalism as opposed to empowerment. In addition, many patients were unable to harness the benefits of digital health and make informed decisions because they lacked interpretation and knowledge skills in using these tools. While this population might have significant differences to a PBT population related to their cancer, single gender, and older demographic, it raises important issues to be considered in terms of moving towards self-management. There may be a group of patients who prefer to defer decisions about their care and self-management to others where possible.

A study conducting longitudinal interviews with brain tumour patients and carers (done separately) found that this group of patients were motivated to undertake self-care or management activities, often with the purpose of optimising their treatment or increasing survival (Piil et al. 2015b). However, many of these were not discussed or recommended by healthcare professionals and came from friends or the internet. This was confusing in some instances for the participants. Some relatives expressed frustration with their HCPs when self-management activities such as complementary therapies were brought up and the HCP indicated that these were unlikely to influence survival. One caregiver felt that the healthcare team should encourage and support them to undertake self-management activities like these, as they felt this was linked to hope and quality of life. Some of the strategies used may have had no evidence of efficacy – or even a concern that they could be harmful –therefore there appears to be a guidance role for the healthcare professional here. However, it can be a challenge to balance discussion of self-management activity as an important strategy for ‘hope’ with appropriate consideration of the evidence base (and no proven efficacy). Accurate information is important. Despite using these self-management strategies when the disease had progressed, this study found that the participants often recognised the ineffectiveness of some strategies and seemed to regret invested costs or time. This would seem to indicate that there is a lack of communication around these issues where support for hope and accurate information is needed. In addition, it should be noted that the participants in this study did not use an HNA and this could have been a mechanism to support disclosure and discussion.

Other studies show that success or failure in managing problems would also seem to have an impact on self-efficacy. One study that provided a perspective on the area of confidence in self-management examined the strategies of prostate cancer patients in relation to some of their identified problems, such as urinary, bowel, and sexual dysfunction symptoms (Paterson et al., 2015). While there was evidence of self-management for these symptoms, the participants did not achieve symptom relief at baseline or at six months. Their self-efficacy score was also measured, and this demonstrated a decline. The authors feel this represents a decline in their confidence to perform self-management. The success of self-management requires a consideration of appropriate support but also managing expectations and reasonable aims. If complete symptom relief is unlikely, clinicians should include clear guidance on expectations when providing information about strategies. This underpins the importance of using evidence and expertise in supporting self-management activity.

However, while policy inclusive of HNAs often advocates supporting self-management and self-efficacy, some authors have revealed negative impacts of higher self-efficacy. Renovanz and Maurer et al. (2018) postulate that caregivers’ self-efficacy, or the perception that they should be independently managing, might be a barrier to seeking help. In their study of glioma patients and caregivers, the carers demonstrated significant needs and distress. Despite this, in most cases, specialist support was not sought. Although not formally assessed, the authors feel that this might be due to the perception that caregivers should be managing. While self-sufficiency and management may have been happening, this could have had detrimental psychological and physical effects which, with support, might have been alleviated. In particular, this study, in common with others, demonstrated high levels of carer distress and patient needs. It would seem to indicate the need for assessment of patient and carer distress, even if self-management capabilities seem to be high.

Another qualitative study looked at why PBT patients did not access support services, despite many needs. They came up with three themes – "don't need help", "don't want help" and "can't get help" (Langbecker et al., 2017). This has some very interesting observations – many with relevance to cognitive challenges. One direct quote resonated strongly:

> "Well, in some cases you possibly get told [about support services available], cause to start with, after the operation, you know, it takes a bit of remembering everything, in fact, I probably didn't" (p.1747).

This study also noted, along with memory problems, that reasoning skills may be impaired and have an impact on problem-solving. For example, one of their participants was struggling with work and making mistakes and they did not know what type of help might work or where to go. Finally, many seemed to have the impression that the problems they were having were just part of having a brain tumour and no matter how severe, they just needed to live with it.

For individuals diagnosed with a brain tumour, there can be an impact on cognitive functions such as memory, reasoning ability or problem-solving skills. This is a significant concern for up to 75% of patients, based on an internet questionnaire (Sloane et al., 2016). While in other cancers pre-existing levels of self-efficacy might have a clear relationship with problem-solving, this patient group may have an inherent change due to cognition. Even if they demonstrate high levels of self-efficacy, their coping and problem-solving may be impaired. Healthcare professionals not only need to assess the readiness for change, but they must also be able to understand the impact of background cognitive deficits and how this might affect the patient's ability to solve problems or undertake new learning.

The impact that cognitive changes may have on the ability to self-manage seems to be clear. One possible consideration could be the role of cognitive rehabilitation. A review found that, although this can be resource-intensive for caregivers, patients and providers, it was beneficial if they could manage to participate (Bergo et al., 2016). Importantly, although there were methodological weaknesses in most studies, improvements in cognition were seen, such as improved learning ability or memory based on subjective and objective measures. More importantly, this seemed to have an impact on autonomy in everyday life and the burden on caregivers.

Then again, as demonstrated in a study of glioma patients referred for rehabilitation, the demands of this were too great, with challenges around interpreting and following instructions and co-ordinating movements (Piil et al., 2015a). Yet, despite this, the opportunity could be considered for more patients. A more personalised approach which recognises the capacity of the patient, their coping styles, symptom burden, stage of illness and preferences for self-care needs to be considered when undertaking an HNA and subsequent care planning. There needs to be recognition that, in some cases, the extent to which a patient or caregiver is able or willing to be involved in problem-solving is extremely variable. Misjudging this may hinder success and adherence to any strategies.

However, giving caregivers confidence to solve problems, as measured by ‘mastery’ (Boele et al., 2017), could potentially be a key context. The authors of this study define ‘mastery’ as a perception of control over the situation. Caregivers with high levels of ‘mastery’ had a significant impact on their patients’ survival. They hypothesise that the caregivers’ proactive approach in managing problems resulted in these survival advantages. This outcome is potentially one of the most significant and needs to be considered further. However, a second study by this author examined an intervention of psycho-educational and cognitive behavioural therapy on improving mastery in a RCT. This did show an impact, but the burden of the intervention was quite high – the dropout rate in the intervention arm was 50% compared to 32% in the control. This may also highlight the need for individualised support of interventions to maximise the contribution caregivers can make.

In patients who did not want support, some had clear preferences to self-manage with existing informal support such as family. Self-management is a key goal of chronic disease management – however, in some cases, a lack of contact with HCPs can be detrimental. For example, if suffering can be alleviated through advice or interventions from specialist HCPs, it may be beneficial to have access to these professionals. In addition, others may shun support to demonstrate their continued independence and it is suggested that this may help with coping. However, the authors suggest that information and support should be tailored to individuals – and when there are high levels of unmet needs that communication skills are used to see how certain resources or referrals might be used to harness the patients' and caregivers' goals of self-managing, possibly with better outcomes.

Hickmann et al. (2017) highlight a very important point – in other cancers, distress and mood changes, such as depression and anxiety, may often correlate to how an individual copes with the diagnosis and management of a life-threatening disease. In brain tumour patients, particularly those who may have one in the frontal lobes, psychological changes can be due to the tumour. The implications are that studies examining interventions for supporting psychological changes in non-CNS cancers may have limitations due to the underlying cause of mood disturbance or coping mechanism. For example, avoidance coping in PBT patients may be due to brain injury influencing reasoning, so problem-solving may not be as straightforward.

Halkett et al. (2015) highlight that the level of information which individuals find adequate can be related to education. This study found that those with tertiary education did not feel they had adequate involvement in decision making. The authors hypothesise that higher education empowers and with information this empowerment can link to decreasing distress. The authors found that tertiary education had the strongest correlation with lower distress. Interestingly, Renovanz and Maurer et al. (2018) found that patients with university degrees had a wish for higher levels of support from HCPs. The study did not investigate the reasons, but it could reflect that they were seeking support to maximise their knowledge and improve the management of their condition.

A study of 84 primary high-grade glioma patients focusing on QOL, psychological outcomes and needs highlighted the importance of information needs being met in supporting shared decision making (Lucchiari et al., 2010). This study used the Needs Evaluation Questionnaire, which has eight questions focused on information management needs. This included considerations of the adequacy of information, communication and involvement in decisions. This research found that patients who had their information management needs met had higher QOL. The authors conclude that a misjudgement in information needs and how shared decision making is implemented may interfere with the production of a context of trust and satisfaction with the therapeutic journey, holding back the adjustment process (Lucchiari et al., 2010). These authors also highlight that assessment of need for information and decision making is often based around individual clinician skills rather than any systemic approach. This may introduce variability which could be negative, and this should be an important consideration of developing shared decision making.

4.9.1.6 Theory 3 - Refinement

To support self-efficacy and activation leading to self-management in PBT patients is complex. The impact of cognitive impairments on ability to self-manage and solve problems was one of the important areas of refinement. The specific impacts an individual is having need to be assessed as these can have a wide variety of influences on the ability to undertake activities or remember instructions.

Distress can be present even with seemingly high levels of self-management. It is therefore an important context to assess in both patients and their families. The very poor prognosis of a significant proportion of those diagnosed with PBT (particularly GBMs), limited treatment options and symptoms of the disease (particularly cognitive impairments) can understandably affect psychological distress and self-management. Assessing distress could allow the practitioner to improve this through interventions to help alleviate it, improving the ability to optimally self-manage.

An important consideration brought out by a study of prostate patients warrants further consideration in brain tumour patients and that is preference for paternalistic models of care. While this group may have significant demographic differences, the option for not sharing decision making may be a preference for some patients. As demonstrated in one study, shared decision making may be more of an expectation and a higher education level might influence this. However, some the mechanisms of this theory (such as empowerment) need to be explored further, as the research is limited. The use of communication skills and meaningful discussion, which invites individuals and caregivers to participate at a level that is appropriate to their readiness, activation and skills, is an important consideration for practitioners. The refined theory is represented by

Figure 11 below and changes to candidate theory are denoted by the red text. (See Table 12 for original candidate theory).

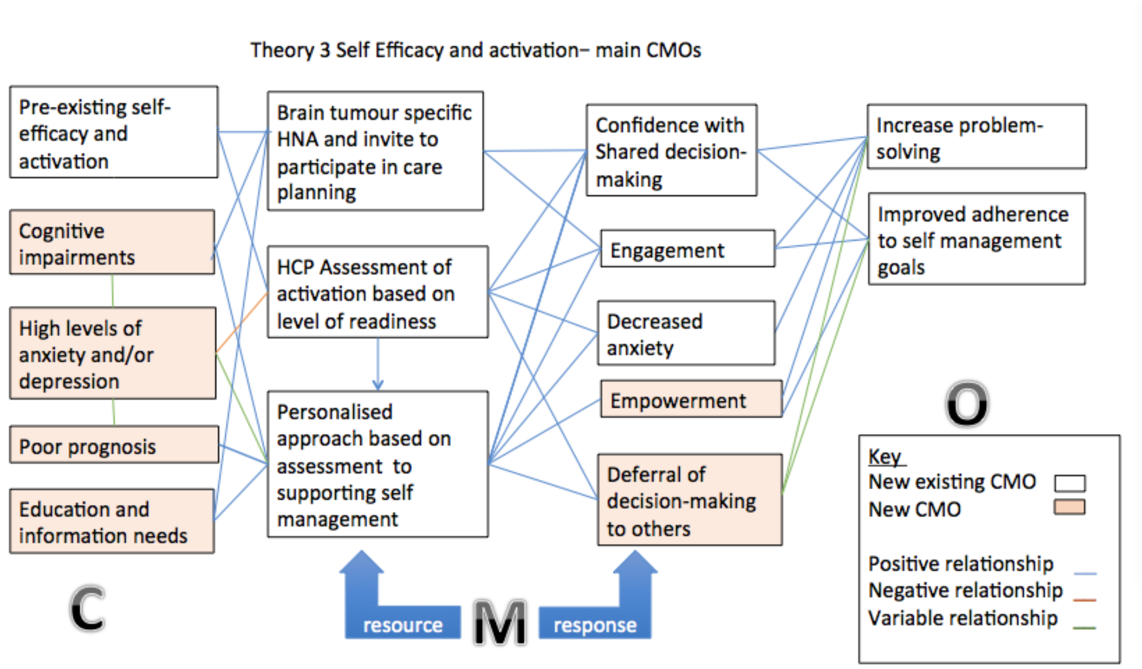


Figure 11– Refined Programme Theory 3

Patients and their caregivers will have pre-existing levels of self-efficacy and of activation (readiness for change) (C), which can influence confidence in shared decision making (M - response) and facilitate problem-solving to self-manage (O). When undertaking a disease-specific HNA, healthcare professionals invite patients and caregivers to participate in related planning and discussion (M - resource). Poor prognosis (and lack of life-extending options, cognitive impairments, and high levels of distress) are important factors (C) and should be part of the practitioner's assessment of the activation as part of the HNA discussion, based on their perception of the individual/caregiver's current situation (M - resource). They should then provide a personalised approach depending on level of readiness (M - resource) – this supports the engagement of the patient/caregiver (R - response). The level of input invited or sought over decisions around care and self-management can be influenced by education (C). What levels of support are needed can be gauged to an appropriate level to increase empowerment (M - response), reduce feelings of anxiety and/or build confidence (M - response). It can also increase problem-solving by patients and caregivers (O) and adherence to self-management strategies (O).

4.9.1.7 Analysis of theory 4 – Practitioners as a barrier or facilitator to HNA and self-management

Commonly in healthcare, policy can ‘dictate’ or recommend interventions that should be implemented. Several strategies, such as incentives (or punishment), education or training can impact if and how this might happen. With complex interventions, implementation can have many steps and each of these may have variable success, dependent on the practitioner.

Preconceptions or attitudes towards HNA

The attitude of the individual and the organisational perception of HNA are important contextual factors suggested by a number of studies. The practice culture, workplace priorities (such as organisational perceptions of priorities for care, for example, the biomedical focus) and embedded culture also need to be considered as important contexts which influence engagement and willingness to integrate needs assessment into care (Handberg et al., 2018). In a study which looked at both primary care and inpatient cancer wards, the hospital culture seemed to influence the perception of HNA and survivorship care planning as an additional and less urgent aspect of care, which could often be missed. A number of practitioners questioned whether the timing of the needs assessment (for example, during diagnosis) was valuable as information and resources should be focused elsewhere, such as the need to prioritise active treatment (Handberg et al., 2018). Another study examined nurses’ views a year after implementation of an HNA programme. They felt that their positive attitude would influence their positive actions as well as the engagement of the patient (Børøsund et al., 2014). They seemed to know that their acceptance was important and advised that, to achieve this, it was important to spend time supporting and educating staff about the value of the process. The integration and acceptance of HNA into routine care has been cited as an important consideration in successful programmes. This may be because the integration places an importance on the activity that reinforces its importance to the individuals (Børøsund et al., 2014; Handberg, Thorne & Maribo, 2018).

Role in supporting HNA (C2)

The acceptance of undertaking an HNA seemed to be strongly linked to the practitioner’s perception of their professional role and their underlying existing competence. There seemed to be a strong indication that HNA programmes were within the domain of practice for nurses, and this influenced the review and response to an HNA. Based on a survey of UK head and neck oncology nurses, only 33% of the nurse respondents felt that doctors made a strong attempt to find out about unmet needs – and 78% of nurses who responded felt that they made a strong attempt (Rogers, et al., 2011). However, only 9% of the nurses routinely used a structured questionnaire. Another study reported that nurses were reluctant to give questionnaires to patients if they did not think the doctor would review them – but it worked much better if nurses could review the patients themselves (Rotenstein et al., 2017). These papers examined the views of nurses but studies which surveyed across the professions reported different completion and adherence for different professions. One study found that the majority of nurses (89%), physicians (55%), and other providers (57%) reported referring to the symptom scores in clinic either ‘always’ or ‘most of the time’ (Bainbridge et al., 2011). In this study, medical staff said their reason for not using it was that they preferred their own assessment strategies, or they did not have time.

Other sources suggest that this fits with nurses’ scope of practice. Blum et al. (2014) highlight qualitative feedback that oncologists may not want to deal with symptoms and issues, and it can be helpful if nurses deal with it. Nurses do seem to see this as part of their scope of practice. One study reported that nurses saw symptom management as clearly part of their role (90%) and 84% agreed that a standardised assessment of symptoms was ‘best practice’ (Green et al., 2017). However, nurses with more than 10 years’ experience were slightly less positive about the routine usage of questionnaires in screening for symptoms or management of symptoms. The authors have postulated that this may be due to more recent graduates having more of an appreciation of evidence-based practice, or it could be that more experienced nurses perceive that they adequately assess and manage patients within their practice. This is despite extensive evidence demonstrating that nurses and doctors can miss symptoms and underrate their severity.

However, some of the role-related considerations might also pertain to resources for services. In a study which examined HNA and advised interventions, doctors did not complete forms on interventions suggested as result of unmet needs in cancer patients in 47.7% of contacts. This compared to 99.7% of telephone support workers who were nurses (Girgis et al., 2009). If a nurse undertook the consultation it was focused on needs, whereas the oncologists were intent on integrating this into their normal clinical consultation, so it is not surprising to see this difference.

It appears that organisational recognition and support of HNA programmes is needed, regardless of the professional group undertaking them. In a number of studies, nurses had the perception that HNA takes additional time (Biddle et al., 2016; Thewes et al., 2016) and did not appear to be part of a normal treatment or follow-up pathway (Thewes et al., 2016). Without accepting this as a priority for care and adequate resourcing, practitioners are resistant to undertaking the process and resentful of the additional role (Biddle et al., 2016). Patients felt frustration when practitioners did not review their responses or address their concerns because of time. However, there can be a concern that if you can't actually do anything about a problem (like finance), is talking about it any use, does it raise expectations that something can be done, and could it be detrimental? (Biddle et al., 2016). Ahmed et al. (2014) voiced a stronger argument, that without support or services to help with needs, it was unethical to ask about them.

However, in terms of organisations, it is important to remember that it is not only the workload or resources – other considerations, such as space, are important. In a study of head and neck cancers, lack of time was the highest-ranked answer for non-completion of the HNA, followed by no private space (Wells, Cunningham et al., 2015). Asking patients to discuss unmet needs without affording privacy can often provide conflict for any professional undertaking care, which may comprise confidentiality and dignity.

The belief or perception of responsibility for the actual assessment, and subsequent actions such as providing information, referral or other supportive care related to the identified problems, is an important factor in the implementation. Certainly, in the context of specialist care, there seems to be some evidence that this might be integrated and accepted more into the remit of specialist cancer nurses rather than other professionals – although it does need to be acknowledged that this may not always match the patient pathway. However, there was some support that primary care saw this as part of their remit, but access to specialist knowledge was very important.

Knowledge in responding to HNA

Ahmed et al. (2014) theorise that healthcare professionals may not enquire about potential symptoms and needs because they are uncommon, considered unimportant, they do not know how to treat it, or know what can be done to help within their time constraints. Many studies report deficits in knowledge as a barrier or challenge in managing supportive care needs. In a study of HNA with head and neck cancers, one of the barriers identified by nurses and a few allied health professionals was lack of knowledge, skills or confidence in providing supportive care (Wells, Semple et al., 2015). This was echoed in a study examining perceptions of the challenges of using HNA. It found that participants reported anxiety in dealing with non-physical problems (these were nurses and therapeutic radiographers), particularly psychological problems where they felt that training was inadequate (Biddle et al., 2016). This paper did say that they had training on what do for each problem, but this did not seem to be adequate. This does seem to highlight the need for reflection on what is actually needed.

This was shared in an evaluation of the Transforming Care after Treatment programme in Scotland, in which HCPs highlighted that they 'felt inadequate' when faced with problems without clear solutions. Patients in this same study also said that dealing with problems of a non-physical nature – such as relationships – were often not easily solvable and they seemed to question the benefits of this. However, others indicated that the process of being listened to could be therapeutic in itself, described as 'a release' (p.22), and this process helped them not to feel alone and depressed (Johnston & Campbell, 2018c). In this same report, patients highlighted that the HNA process helped guide them to help and 'get back to normal'.

In addition, it was voiced that in several cases patients did not feel they were speaking to the right professional to deal with their problems. For example, social, psychological or financial problems were not perceived as a problem for doctors. This also was a concern for some groups of professionals when identified problems were not their areas of expertise and there was not a clear referral option (Biddle et al., 2016). In addition, it can be quite satisfying if patients raise physical issues that are easily dealt with but assessment and support for psychological distress can be more challenging and not easily addressed (Biddle et al., 2016). However, Børøsund et al. (2014) present the view that, while many of the needs were not in the scope of practice, if nurses or other practitioners viewed themselves as facilitators for self-care or referral, this barrier could be overcome.

Confidence and competence of practitioners for HNA

The role of training and education has already been mentioned above as one of the key mechanisms in the development of knowledge leading to confidence and competence. While good experiences can build confidence, a sub-optimal experience in which the practitioner feels unable to provide care or advice can lead to feelings of inadequacy and anxiety (Biddle et al., 2016) and a reluctance to undertake or fully engage in HNA. Training in using an HNA tool and options for support and referral are important, but as Børøsund et al. (2014) found, communication training to support patient-centred and sensitive conversations was the most important training needed.

In a study of glioma patients and caregivers (Renovanz, Maurer et al., 2018) patients seemed to be poorly informed about support and had significant unmet needs. They linked this to the communication skills of HCPs as they relate to this specific group, with two important considerations that need to be addressed in terms of supporting unmet needs. Treatment options may be limited in this group, so the focus of discussion may be on palliation and maximising QOL. Also, the comprehension and memory of glioma patients may be impaired specific skills and strategies are required to support individuals in managing their needs.

Improved practitioner knowledge supports self-management

Key areas of knowledge have been identified as important elements to help practitioners in undertaking HNA and supporting patients, such as how to use the HNA and decision support aids, which might include local guidance on referral pathways, electronic decision trees or quick reference cards (Biddle et al., 2016; Santana et al., 2015). Nurses are aware of the detrimental impact that a lack of response can have. However, the study by Børøsund et al., (2014) identified a lack of advanced communication skills to support extreme reactions or sensitive subjects (such as dying or sexuality) and nurses said that they may avoid the situation or respond inadequately. The role of communication skills training was highlighted, and it is likely that this is a key mechanism to support successful implementation. The role of practitioners as a potential gatekeeper for information or referral to help individuals – and their specialist knowledge – is key to imparting this. Knowledge of interventions and services are needed, but some research raised concerns that there may be reluctance to refer, even with evidence of efficacy. Piil et al. (2015b) highlight that healthcare professionals had a reluctance to refer this group of patients to rehabilitation services (for example, a programme for individuals with other neurocognitive deficits) despite evidence of benefit. The reasons were not clear, but this could be a barrier to supporting self-management.

Johnston and Campbell, (2018c) in their realist evaluation of HNA, emphasise the importance of the assessor being embedded with or having strong links to the cancer patients' locality or community assets, but also with communication skills in listening and eliciting discussion about their concerns. This supports that who the assessor is needs to be carefully considered in terms of an intervention and they proposed that, "this would result in [patients and caregivers] feeling more reassured, more supported and confident to use the knowledge and contacts provided to increase self-management" (Johnston & Campbell, 2018c, p.34). This seems to reiterate that the linkage with local knowledge and an individualised approach is a key consideration.

The actual process of HNA was seen as an opportunity for patients to reflect and know they could get better support through primary care by voicing their problems using HNA in that setting (Clarke et al., 2019). Additionally, in this study the practitioners who were based in primary care felt they developed their knowledge for this group of cancer patients when holistically assessing needs to allow them to provide better supportive care and advice. This study did highlight that, in some cases, many patients still seek a more 'paternalistic' model of care, but that strategies such as an electronic HNA could be part of developing a platform to move patients towards more self-management.

There are some clear areas of recommendation that knowledge and training in the process, communication skills and how to support unmet needs are crucial elements of an HNA programme. Those designing services need to be clear on the aims of any HNA programme and all its elements, the roles and responsibilities, and the resources allocated, need to be clarified.

Disengagement or distress as an outcome of suboptimal HNA

In an examination of patients' views of HCPs not dealing with a problem, they voiced that they would feel 'betrayed' (Biddle et al., 2016). The implications of this might be disengagement but it would seem to indicate a potential breakdown of trust or the relationship. The lack of a follow-up after an HNA where the HCP gave support or advice for problems was also viewed as less than ideal, with patients reporting instances of advice for self-management which then did not progress beyond the first action (for example, buying a relaxation tape that was never used). The lack of continued engagement on the problems by the HCP seemed to also be linked to disengagement by the patients in their self-management strategies. Other literature highlighted that practitioners had concerns that they might not have the skills or knowledge to deal with all of the problems raised in the HNA, which could result in ambivalence as a response (Biddle et al., 2016; Børøsund et al., 2014).

However, even when support is offered, disengagement might still occur. In a study examining unmet needs in brain tumour patients within three months of diagnosis, there was lack of uptake of offered referrals for psychological care (Langbecker & Yates, 2016). The authors say that the reasons are not known but theorise that this may be linked to other research, which reports HCPs negative perceptions of psychological care in cancer care (Dilworth et al., 2014), lack of knowledge, or organisational challenges in referring patients. However, these authors feel that education and development of communication skills in discussing psychological issues is needed. The involvement of end users in developing appropriate services in this area is important, to design what might work best for this group within the capacities of the service and better match the needs and capacities of those who are referred.

Building acceptance

Those who are using HNA tools need to believe that they are useful, while also being easy to interpret and access. However, to do this, there needs to be agreement from everyone on what the goals of the programme are, and to agree priorities and compromises. One of the areas of disagreement may come from the choice of HNA tool and the reasons why. Many providers seem to share the perception that cancer-specific tools, rather than more generic cancer HNAs, may add additional benefit as they are then more specific to certain cancer populations (Pereira et al., 2016; Wells, Semple et al., 2015). In the routine implementation of a prostate-specific questionnaire – after they had already been using a generic cancer assessment tool – medical staff felt it was far more relevant as it focused on the specific issues, such as incontinence (Korzeniowski et al., 2016). However, there are alternative views. In one study, nurses commented that standardised validated tools should be used and this should be considered ‘best practice’. Others felt that some variation was needed due to symptoms that might be missed in specific cancers (Green et al., 2017). One of the recommendations of a recent Cochrane review on screening for psychological wellbeing and care needs in cancer – which found the results of studies were weak and did not demonstrate efficacy for this intervention – advocated that validated internationally recognised tools should be used, or core agreed outcome data sets. The implications are that, without this, we will continue to lack good quality studies to examine the efficacy of needs assessment. While both arguments have merit, as discussed in previous chapters, if the disease-specific needs that cause significant concern are not assessed, the studies will have limited usefulness to those populations.

Other considerations concern brevity and accessibility. It was considered important that questionnaires were not too lengthy and there should not be redundancy in asking questions twice – for example, routine questions in consultations, then also on the questionnaire (Rotenstein et al., 2017). Additional steps such as logging into separate systems can be cumbersome and seamless interfaces may make practitioners more accepting of this technology (Børøsund et al., 2014). The ability to see aggregated data was also considered a benefit that could improve adoption, as it could track changes over time which capture the patients’ perspective (Basch & Abernethy, 2011; Korzeniowski et al., 2016; Pereira et al., 2016; Rotenstein et al., 2017).

Studies which looked at PROM data across cancer types found that 63% of oncologists did not refer to the provided PROM data before the clinic appointment, despite having it. While HNA is only one type of PROM, the reasons postulated for this are lack of communication skill, no solution to the problem, or no time to deal with it (Greenhalgh et al. 2013). When actual transcripts of consultations using PROMs for patient symptoms were examined in oncology, doctors often closed down (minimise or don’t enquire further) conversations where an issue was identified in which there was no clear medical treatment, such as fatigue (Greenhalgh et al. 2013). This is not an issue unique to medical staff – in one study examining acceptance of an HNA programme, nurses expressed that unless they felt comfortable entering into communication with the patient about the HNA report, it would most likely not happen. Unconsciously, communication about sensitive topics was put at the bottom of the list of daily tasks if nurses did not feel confident about their communication skills and it was easier to focus on practical tasks (Børøsund et al., 2014). Acceptance is clearly linked to knowledge and training to undertake HNA, as well as putting in place the resources and plans for this service, including who and when this should be done.

4.9.1.8 Theory 4 – Refinement

While there may be an indication that the process of HNA may have a positive emotional impact for patients when completed by an engaged and knowledgeable practitioner, the link with improving self-management remains unsubstantiated. However, mechanisms likely to support self-management, such as shared decision making and improved engagement that were influenced by practitioner-related contexts and actions, did have support. This would be a key area to investigate in the future.

Organisational culture and support is a clear mechanism, as this can help put many contexts in place, such as role establishment, referral pathways and development of skills and knowledge to support this activity. Without this it would seem that the challenges of implementation are considerable.

The loss of trust when needs are ignored could cause harm. The impact of ignoring needs seems to be clear, with both patients and nurses being aware of the detrimental impact that non-responses have. This supports how important it is for assessors to have communication skills and knowledge to respond to voiced needs. The refined theory is represented by Figure 12 below and changes to candidate theory are denoted by the red text. (See Table 12 for original candidate theory).

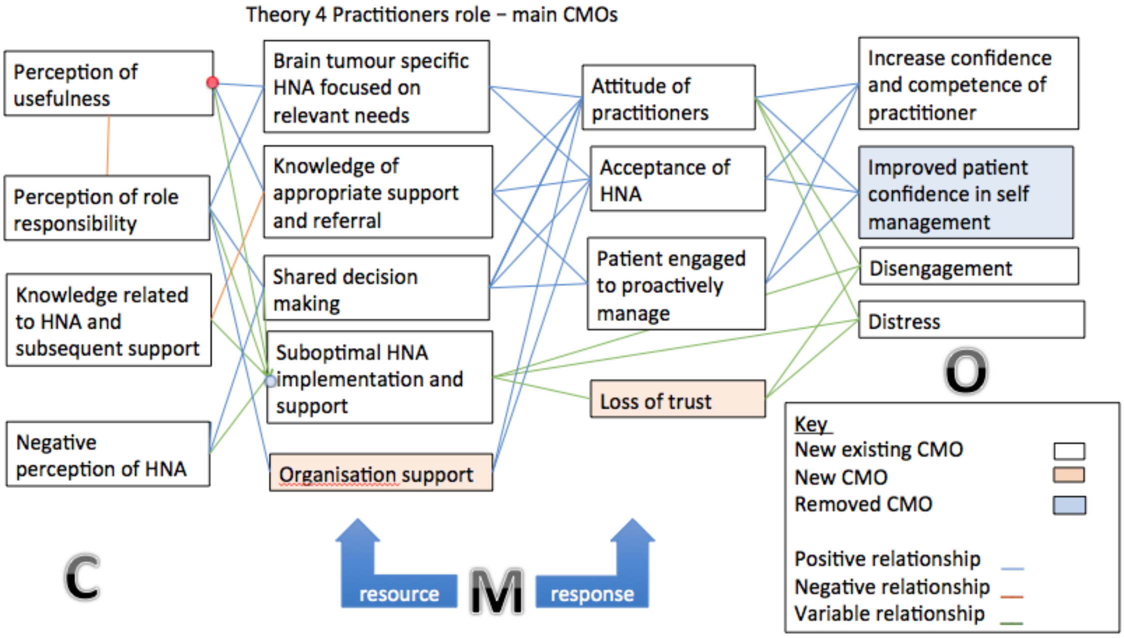


Figure 12– Refined Programme Theory 4

Practitioners may have preconceptions about HNA related to the usefulness of the intervention (C), their role in supporting the process (C), and their knowledge in responding appropriately to patients' holistic problems (C). This will influence their engagement, implementation and their attitude to undertaking HNA (M - response). A brain-specific HNA focused on the relevant problems of this patient group, that supported a holistic assessment of relevant needs (M - resource), can build acceptance of the benefits (M - response). If practitioners are provided with organisational backing, knowledge of appropriate support and referral options (M - resource) and as well as training in shared decision making to respond to problems (M - resource), this can improve the confidence and competence of the practitioner in undertaking HNA (O). For the patient and their caregivers, this can result in feelings of engagement in proactively managing their identified problems (M - response) when they are involved in decisions with practitioners about solutions to alleviate their needs (M - resource) which can improve their confidence in self-management (O). Alternatively, if the practitioner has negative perceptions about the HNA process (C) and suboptimal delivery results in needs being ignored, or mismanaged (for example, with solutions that do not work for the patient/caregiver) (R - resource), this can result in feelings of loss of trust (R - response), disengagement (O) and distress (O).

4.10 Discussion

4.10.1 Summary of findings

This realist review of 30 studies has provided some important insights into HNA in a PBT population. Unsurprisingly, the impact of cognitive changes is relevant, either directly for the individual as a contextual factor, or as a mechanism in how others deliver interactions or need to respond to optimise outcomes. Some of these were not unexpected, such as the importance of having an HNA which captures these cognitive issues, while others, such as self-efficacy, may need to be assessed and considered differently in this population due to cognitive impairments. For example, those who might previously have had high self-efficacy could have memory impairments which impair their responses.

Another significant area relevant across all the themes was the importance of the practitioner's skills and knowledge. These can be quite wide ranging and include knowledge of PBT, assessment of psychological issues and capacity to self-manage, and knowledge of disease, referral and support options. Skill in communication, shared decision making and supporting psychological needs was also highlighted. This is vitally important to help the assessor engage in this process fully – but also key for establishing trust and working with patients and families to build their confidence in this area.

Finally, one of the other areas of importance is the role of the organisation or service and developing and resourcing this programme. This needs clear acknowledgement and planning of who is undertaking it and then guidance on what options might be possible to support individuals with their identified needs within and outside that service. While the literature has mentioned that nurses, or more specifically, clinical nurse specialists, might be best placed to deliver this, resources such as time and space need to be considered.

4.10.2 Comparison with other literature

A realistic evaluation report examined HNA programmes at Transforming Care After Treatment, based in Scotland. One of the theories that emerged was focused on increasing self-management across all cancers (Johnston & Campbell, 2018c). This report proposed that the HNA programme and the responses to unmet needs (information, referral/signposting, care plans etc.) can help clarify the role of the person affected by cancer in recovery and this can increase self-management (and longer-term improvement benefit from this type of programme). These authors say that is it important to individualise this approach – however, what individualise means is not fully expanded upon – and consider the personalisation around, not only the problems, but also the *capacities* of the *individual and family*. The phrase 'a person affected by cancer' is quite relevant, however, consideration of the proxies (family) and the individual with cancer as a unit, at times may be more important due to cognitive difficulties.

Reducing distress which has not been a major focus may be a very important consideration in terms of potentially improving responses to HNA which are related to self-management for both patients and carers. There is limited evidence of what psychological support might be helpful, but Hutchison et al., (2006) provide a framework for stratified psychological support based on level of distress that could be a starting point. The assessment of psychological distress as an important component, similar to the North American models, could easily be done while still keeping the primary focus on support and self-management.

4.10.3 Limitations

The evidence used to support this realist review was not limited to any specific methodology. However, there were a number of limitations related to the available literature. RCTs that help test whether an intervention works or not – in this case HNA – were not available in PBT patients. This limits evidence demonstrating outcomes related to HNA programmes and these were often based on research into other cancers or from qualitative research. Qualitative studies are often more useful to explore the triggering mechanisms for why an intervention may or may not work in certain contexts. For some of the final programme theories demi-regularities were clearly evident, such as, the role of HNA in legitimising needs had a great deal of support. In other areas, such as assessment of activation, the reduction in anxiety had less support. Arguably there may be complexity that needs to be explored through focused research on these theories.

Due to this deficit, it is likely that a full consideration of the complexity of HNA programmes in brain tumour patients has not emerged. Although it is likely the components of this complex *intervention* can be considered, all aspects of complex adaptive system in which the programme is implemented may require further study. A realist evaluation focused on the delivery of an HNA programme can help to test the proposed programme theories from this research, while also seeking clarity of any emerging contexts, mechanisms or outcomes.

There were also limitations on the review based on time and resources. This review has focused on specific areas, such as self-management, but some areas which could have illuminated this further were not theorised, as the scope needed to be focused to be manageable. For example, the role of electronic tools in both assessment and management is likely to have an influence on self-management, but this area was not a focus due to these limitations.

Numerous publications on realist methodology have been cited in this thesis and guidance for undertaking a realist synthesis has been produced by the RAMESES quality standards (Greenhalgh et al., 2017). However, there is very limited explicit methodological guidance in the approach to undertaking data analysis and synthesis through each stage of theory generation and refinement (Gilmore et al., 2019). This iterative process requires a great deal of skill and is generally undertaken by larger teams. It is recognised within this methodology that a researcher's judgement and knowledge is utilised to refine the programme theories (Gilmore et al., 2019). While input was sought as described in the methodology, one researcher undertook the majority of this iterative process and this could have resulted in the lack of focus on an important demi-regularity or underlying mechanism. For this reason, realist reviews can benefit from a team approach as the decision-making could have provided further refinement and views of some aspects.

There were challenges in searching for evidence, with some chains of inference having only limited evidence, particularly in a PBT population. However, this does provide the basis for further refinement through a primary evaluation in this population.

4.10.4 Conclusion and recommendations

This review has provided some theories of what might work, for whom and in what circumstances when implementing an HNA programme for brain tumour patients. The theories provided the basis to see what might work in some circumstances and alternatively in what might not work. The evidence for some aspects of the programme theories was robust enough to help interrogate these and in others there was limited evidence and/or a lack of evidence in PBT population. Where possible, relevant other related research helped to provide some useful insights. One of the main considerations when using this data was the multifaceted impact and implications of cognitive impairment and considering when research from other cancers might not be relevant.

This review may also offer some aspects of these theories which are likely to be relevant in other cancers, for example, how practitioners can be a facilitator or barrier. However, it is important that the contextual differences of brain tumour patients are considered. The Macmillan recovery package and variations of this throughout the UK provide the structure for HNA to support and improve the QOL of individuals living with and beyond cancer (Macmillan Cancer Support, 2015). However, the emphasis on recovery may not provide the optimum support to those affected by a brain tumour who may have a poor prognosis or see recovery options as limited. There may be different pathways or additional considerations for support for PBT.

The process of HNA needs to include the assessment of not only their needs in isolation, but in combination with their self-management potential. Shared decision making to a level that is appropriate should be used and the identified needs should always be acknowledged. Care planning, information, support and referrals should be made with a strong evidence base that is shared in an understandable and compassionate manner. What is likely to be achieved through self-management should be clear. False expectations may ultimately result in the individual losing confidence.

Improving the capability for self-management requires assessment of needs and capabilities. Knowledge of what can be done to mitigate problems is possible, with clear appropriate communication being important. Consideration of interventions such as cognitive rehabilitation, which might improve the independence of individuals affected by a brain tumour, may be an important area to consider in supporting self-care. This may directly help some of the challenges with cognitive deficits but also improve reasoning and the ability to solve problems for challenges in other areas. This could also reduce the burden on caregivers, as individuals may be able to take a larger role in self-management.

There does seem to be evidence that taking care of the carer and involving them more holistically in care planning might link to improved outcomes for them and their patients. These benefits might be greatest for the carer’s psychological health to help them rise to the often considerable responsibilities they have. Currently much of this care is delivered by the third sector, but this is not routine and arguably many that need it most may not be accessing it.

Despite widespread adoption into routine practice in the UK, there does seem to be limited evidence of benefit for HNA programmes in cancer. For example, in the TCAT evaluation of HNA programmes in Scotland, the local and/or national projects failed to demonstrate an improvement of the effect on self-management (Johnston & Campbell, 2018a). While they indicate that there is a shift towards this, the authors note that this is challenging and complex to measure. It could be that seeking a quantitative evaluation of the effectiveness has lacked positive outcomes as studies have focused on the wrong outcome measurements, not taking into account the complexity of interventions, small sample sizes, and trial design. These problems have also been seen in trials examining interventions to assess self-management. While there might be slightly more consensus that PROMs focused on patient activation may be useful surrogate markers for self-management, what self-management can achieve in terms of improving outcomes that are measurable is also problematic. However, this realist evaluation does offer a focus and, through the ability to use multiple data sources in the future, additional evaluation can allow these programme theories to be refined further.

5 Chapter 5 - Discussion

5.1 Introduction

Throughout this thesis, the outcomes of each section of the research guided the next phase. Careful reflection on the results of the three studies provided a natural progression using this iterative process. The discussion below draws on the key components of each of the studies and examines the key outcomes from each stage and the related literature. These include the systematic review of HNA tools (study 1); the qualitative study of patients' and carers' experiences and perceptions of needs assessment and having their needs met (study 2); and the realist review of needs assessment in brain tumour patients as they relate to these questions (study 3). The discussion will focus on the progressive nature of the findings and how each stage of the study was built on the findings of the previous one.

This thesis set out to examine the following question: How can HNA, followed by appropriate support and care, improve outcomes for brain tumour patients? This chapter will revisit the main policy and practice framework for HNA in the UK to support discussion of this question. The Recovery Package was instigated by Macmillan Cancer Support and has been recommended or required as part of cancer policy for each of the countries within the UK (Macmillan Cancer Support, 2015). This programme comprises several key components – HNA and care planning, treatment summaries for the patient and GP, a cancer care review with primary care, and education and support for patients. These components ensure that the patient's unmet needs are detected and care is planned to respond to them. The programme should include support and information to help patients self-manage their conditions as much as possible and maximise their wellbeing.

There has been extensive literature examining needs assessment in good prognosis or common cancers (for example, breast or colorectal cancers) to support the development and use of HNA and the Recovery Package in these patients (Aranda et al., 2006; Clarke et al., 2019; Morrison et al., 2012; Nanton et al., 2017). However, in other complex, rare and poor prognostic cancers, such as brain tumours, research has been limited. This identified gap has been met through the outcomes of this research, which provides some evidence to support development and implementation of HNA in brain tumour patients.

This lack of research on HNA specifically in brain tumour patients was evident at the start of this study in 2014. Subsequently, there were many questions at the outset, including what should be used for assessment of HNA. This formed the main focus of the systematic literature review. Feedback from an informal scoping study in practice confirmed that there was nothing routinely used in Scotland, although the Brain PCI had been developed and used in a small research study in Edinburgh (Rooney et al., 2014). While this study helped to demonstrate the need for a brain-specific HNA, as many of the unmet needs were related to neurocognitive impact, the tool used had not undergone psychometric testing. Other authors advocated the need for HNA to be a tool with good psychometric properties (Bonevski et al., 2000; Richardson et al., 2007; Wen & Gustafson, 2004). Possibilities for what could be used as a brain tumour-specific HNA tool had not previously been evaluated and this led to the decision to conduct a systematic review, identified as study 1 in this thesis. The aim of this review was to explore what tools there are to assess needs or concerns in brain tumour patients then, when identified, based on an evaluation of the psychometric properties and clinical utility of these tools, seek to recommend what could be used in this population. As reported in Chapter 2, there was no tool that could be clearly recommended in its current form.

Another key outcome of study 1 was that there was a lack of patient input during the development of some of the assessment tools. Similarly, there was no patient or carer-related research that explored their perception of the usability and feasibility of the HNA when given the opportunity to look at different designs. This clear gap was explored in study 2 of this thesis, where patients and carers were invited to review the four potential tools (reviewed in study 1). This involved focus groups and interviews with patients and carers. Study 2 also examined patients' and carers' experiences and perceptions of unmet needs, how these were responded to, and any impact the response had. Although there are some studies focused on the unmet needs or symptoms of brain tumour patients, these do not examine care received in response, nor the related positive or negative impact (Ford, Catt, Chalmers & Fallowfield, 2012; Janda et al., 2008; Rooney et al., 2014). Study 2 also provided some extremely valuable findings revealing what patients and carers reported as their needs or concerns, and the subsequent responses. Their insights into what worked and what did not provide details of what interventions or responses might be useful (or not beneficial). The final questions and study 3 evolved from a consideration of the outcomes of both study 1 and 2 and was developed to propose how an HNA programme could work in practice, while taking into account the complexity and variability connected with patients and associated interventions and responses.

Consequently, the main findings of this thesis address the following questions:

1. What tools are there to assess needs or concerns in brain tumour patients? Based on an evaluation of the psychometric properties and clinical utility of such tools as an HNA, what could be recommended for use in this population?
2. What are patients' and caregivers' experiences and perceptions of unmet needs and strategies which may address these (or avert problems before they become unmet needs)? This includes their perceptions of the tools identified from study 1.
3. How and in what circumstances might an HNA programme improve self-management in brain tumour patients?

The first question was examined with support from study 1 and with some input from study 2, which focused on patients’ and carers’ perception of the tools. Question 2 was addressed in study 2 and the insights of this study provided the basis for the development of a conceptual model of unmet needs in brain tumour patients. Question 3 was examined through the realist review (study 3). However, it should be noted that this study integrated findings of study 1 and 2, therefore integrating findings from all stages of research in this thesis. This discussion brings together the contribution of the research contained within this thesis as a whole. Using the findings from this thesis, a practice-based model is presented in this chapter to propose, ‘How can HNA, followed by appropriate support and care, improve outcomes for brain tumour patients? This model provided the structure to outline the contribution and consideration of this research in the context of existing practice and policy, demonstrating the unique input of this thesis. Finally, recommendations for further research are outlined.

5.2 What tools exist to assess needs or concerns in brain tumour patients?

Four potential tools were identified from the systematic review in study 1. Yet, based on the proposed criteria (good psychometric properties and good HNA design), no tool as it was currently designed was successful in this endeavour. The options were to undertake psychometric testing and refinement of a 'good' tool designed for HNA or to expand one of the symptom tools (although this would also require additional psychometric testing for additional aspects). In study 2, the participants provided insight into these four tools during their review, observing that increasing length or complexity of questions in the questionnaires they reviewed presented more challenges for completion. Some patients reported that no matter how 'simple' a tool was, they would still need help with completing a questionnaire, regardless of format. Together, study 1 and 2 raised serious questions about the challenge in developing a tool with good psychometric properties and, ultimately, the usefulness of HNA as a measurement tool for clinical practice (with psychometric validity). This is a significant contribution as it provides evidence to show that HNA tools may not be a good outcome measure for clinical or research purposes in this population. The next section discusses why there are challenges with developing a psychometrically valid tool for this population. However, there are benefits of HNA, so the options will be discussed with support of the wider literature.

5.2.1 Why are there challenges in developing an HNA tool for brain tumour patients?

Developing a user-friendly yet psychometrically-valid and reliable tool has specific challenges, not least due to the complex presentation in this population. To establish content validity, 'need' should be comprehensively represented. 'Need' in itself is not a construct, but there are a wide variety of individual needs and arguably this is quite an extensive list. One of the tools, the Brain PCI, lists 50 individual areas of need (Appendix 6). This brings the challenge of establishing reliability through internal consistency by examining the relationships between items that measure the same concept. In Chapter 2, it was demonstrated that, while some needs or symptoms may have some theoretical or biological relationships, many are unrelated and individual. Although the symptom questionnaire MDASI-BT (Armstrong et al., 2006) was able to demonstrate internal consistency, it would require additional questions to ensure each construct of need is fully represented. This would increase the overall length and complexity, which Study 2 confirmed as being particularly problematic for brain tumour patients.

In addition, it could be questioned whether a psychometrically valid HNA questionnaire is required, as there appear to be limitations in its use as a measurement tool. Previous reviews of HNAs in other cancers have failed to demonstrate that needs can be reduced using psychometrically valid HNA tools and the authors raised questions about the sensitivity and suitability of a reduction in unmet needs as an aim of interventions to assess the benefits of HNA (Carey et al., 2012; Schouten, et al., 2019). More recently, a review of generic cancer HNA tools highlighted that none of them had 'good' psychometric properties, or indeed covered all domains, with unmet sexual and cognitive needs often not addressed. This may highlight both the challenges in having a psychometrically valid tool and a ‘good’ HNA (Jiao et al., 2018).

The struggles in developing these tools for any cancer patient are apparent. A large proportion of brain tumour patients, possibly those with more significant needs, might not be able to participate in developmental trials. Some of the participants in study 2 highlighted their inability to complete any questionnaire independently. This was mostly due to related cognitive issues such as trouble in seeing, reading or understanding. It is therefore to be expected that to establish validity in any new tool, those with less serious neurocognitive problems (who could complete it themselves) would need to be the target population. Nevertheless, this still leaves those that need it most (the current sample group) without a suitable tool that is either valid or user-friendly. In fact, focusing on feasibility for usage by the target population was supported as early as 2008 by Garssen and de Kok. They suggested that rather than focusing on psychometric testing to develop the perfect tool, being able to use this practically and easily in a real-world setting is far more important. Given the complexity of most assessment tools and the individuals who need to use them, the recommendation resulting from the analysis of study 2 is to focus subsequent research on the use of this as a communication aid for supporting assessment, rather than a measurement.

Accordingly, there are options which could be recommended if HNA is accepted as an accessory for assessment and communication with an experienced clinician. As part of an assessment process, an HNA tool could help to detect unmet needs better than clinician assessment alone. This can direct support, information, referral and communication about the most significant needs that could ultimately improve outcomes for patients and carers. There are potential options to support the use of these tools for those with cognitive problems. These include ensuring that tools are designed in line with the cognitive capabilities of potential respondents in item content, administration procedures and assessment contexts (Kramer & Schwartz, 2017). For example, with item content, developers should consider what cognitive demands are being placed on the patients. Are they asking them to remember their experience over a time period, or consider how others might perceive them? If so, this might add unnecessary cognitive demands. HNA for brain tumour patients' needs to be usable and accessible, simple in structure and have the ability to identify a wide variety of unmet needs.

5.2.2 Might a selected HNA tool be usable?

In study 2 of this research, where the participants reviewed the four tools identified in study 1, they reported that a longer or more complex tool was more challenging to complete. Although quantitative feedback such as a formal survey was not sought in study 2, the majority favoured the most uncomplicated design – the tick box of the Brain PCI (Rooney et al., 2014). Challenges with completing PROs have also been reflected in other research. A recent study established that the majority of brain tumour patients needed assistance with completion across a variety of PROs (Renovanz et al., 2016). In this study, 71% of patients required help to complete the HNA called the Supportive Care Needs Survey - Short Form 34 - German version (SCNS SF34-G). The EORTC questionnaires (EORTC QLQ-C30 + EORTC QLQ-BN20) and distress thermometer/problems checklist were completed with help by 49% and 58% of the patients respectively, due to disease impairment which was mainly neurocognitive. Interestingly, the short form of the Supportive Care Needs Survey which had only nine questions, albeit with Likert scales, was used in the second cohort of the study – and yet 68% of patients still required support to complete it correctly. This study raises even more questions about what is required in questionnaire design to allow these to be completed independently by brain tumour patients.

The outcomes of study 2 and the published research above highlights that a self-completed questionnaire might not be feasible for the majority of brain tumour patients,. Other authors have also shared the view that most patients with cognitive impairments cannot meet the demands of these questionnaires and respond to PROMs in a meaningful and valid manner (Kramer & Schwartz, 2017). However, in study 2 HNA was viewed as a potential benefit for supporting discussion and communication around need and there is merit in using HNA for this. While both carers and patients in study 2 did not think that the patient would be able to complete an HNA, the carers suggested that they could do it. The use of a tool that can be self-completed (or by significant others) and covers a potentially wide range of the unmet needs experienced by this patient group could save time in an overstretched health service and focus the discussion. Yet, balancing the breadth of potential needs for a comprehensive 'good' holistic assessment with ease of completion for capable patients – or one that is accurate if others complete it for them – remains challenging.

When others such as carers complete the questionnaire, it must represent the needs of the patient. This is imperative to ensure that patients' needs are captured as correct information, which is essential to help mitigate their unmet needs. It may be beneficial to consider proxy completion of HNA as an option. There is some evidence that completion by family members is accurate for some brain tumour-specific questionnaires. With a brain-specific symptom PRO, the MDASI-BT demonstrated good inter-rater reliability between patients and carers (Armstrong et al., 2012a). Interestingly however, another study demonstrated that carers may give a more accurate account of some symptoms (when compared to the clinician's assessment). In a study using Patient Health Questionnaire-9 (PHQ-9), which is used for depressive symptoms, carers achieved higher concordance than patients with the objective manifestations of depression when an in-depth assessment was undertaken by a specialist psychiatrist (Rooney et al., 2013). This was also seen in another study which demonstrated higher agreement between relatives and the clinician than the patients on scales which focused on behavioural changes (Simpson et al., 2015). These studies are important to consider as, although the reasons for patients' 'less accurate' reporting were not explored, such behaviours may also be explained by memory problems or aspects of these which did not cause the patient any concern. However, areas such as behavioural problems can be very challenging or distressing for the family or carers as highlighted in Study 2. Having knowledge of these to help support the carers is important.

A strong case is now emerging from this thesis for the inclusion of carers in the HNA assessment. While concordance with the patient experience requires additional research to test accuracy with HNA, the inclusion of significant others or carers is likely to add information and is essential to fully understand the impacts of the patient's disease. It may prove to be more accurate in the assessment of objective manifestations associated with behavioural changes caused by the brain tumour, but it could also help to reflect the experience and their impact on the carer and/or wider family.

It is essential to have a detailed reflection of unmet needs and the evidence above supports the view that there would be improved accuracy if a carer or relative helped in completing the assessment. Other strategies to consider are electronic HNAs, with some participants in study 2 suggesting that this is a benefit. In many centres where an electronic HNA has been used (Ipsos MORI Social Research Institute, 2015; Jiao et al., 2015; Nanton et al., 2017) there appears to be some clear benefits, such as providing channels for improved communication between health or social care professionals (Ipsos MORI Social Research Institute, 2015). This will be discussed in greater detail at the end of this chapter in Section 5.7.

Although the review did not identify a clear tool, there are potential options from the identified tools with the analysis from study 2. The Brain PCI provided the widest coverage of unmet needs so this could possibly be a tool ready to use, or a hybrid of this with any of the two symptom tools (MDASI-BT or NFbrSI-24) could be a starting point.

5.3 What are brain tumour patients' and caregivers' experiences and perceptions of unmet needs and needs assessment?

The second research question was answered by study 2, the primary research study. This study explored patients' and carers’ experiences and perceptions of unmet needs or concerns and strategies to address them. As highlighted in Chapter 3, four themes emerged from the analysis of this study – ‘Altered Self’, ‘Impacts of Others – Responses and Actions to Unmet Needs’, ‘Impacts on Patients and Carers – Perceptions of Coping and Self-management’ and ‘The Role of HNA Tools and Strategies’. These contributed to the conceptual model of need developed from the analysis of the findings, which provided a basis for theoretical linkages between these concepts and described the relationship between them. For example, the conceptual model helped to illustrate how a brain-specific HNA can facilitate a focus on the patient’s agenda and how this in turn can support the development of trust with specialist practitioners. However, this was a theoretical model and many questions still remained on how HNA could be implemented within the current practice and policy environment.

One option would have been to progress further studies to develop and refine the psychometric properties and/or HNA properties on one of the tools. However, the testing and design requirements for good psychometric properties with a clinical application which covered the required holistic needs, and to develop this in the brain tumour population, would present significant challenges. However, despite this, HNA was almost universally agreed as something that could 'help' in their discussions and communications with care providers.

At this point, discussions were held with the supervisory team to consider the next stage. The driver for a change of direction was focused on providing some evidence to guide practice. There was almost unanimous agreement among the participants in study 2 that HNA could improve discussions with health professionals and it was important to examine how it would work to improve their wellbeing. The findings of the first two studies, as well as the informal scoping study with practitioners, helped in the consideration of this. A number of other reviews on HNA had highlighted a lack of evidence over the benefits of HNA. There was also a debate about how efficacy should be assessed (outcomes) (Carey et al., 2012; Carlson et al., 2012b). Another consideration was that HNA packages are a very complex intervention. Although the systematic review focused on the HNA tool, this could not be viewed in isolation as it was the responses to the identified unmet needs (such as intervention or referral) outlined in the Recovery Package which were arguably more important. After considering the evidence and options for investigating complex interventions, realist methods were selected as they assess how a complex 'programme' may work and allow the author to deconstruct the components of what makes something work or not (Pawson & Tilley, 1997). One of the realist methodology experts, Gill Westhorp, put forward the following as the most appropriate context for this methodology:

> “Realist impact evaluation is most appropriate for evaluating new initiatives that 'seem' to work but where 'how and for whom' is not yet understood; initiatives that have previously demonstrated mixed patterns of outcomes; and those that will be scaled up, to understand how to adapt the intervention to new contexts” (Westhorp, 2014, p.7)

These criteria resonated with implemented HNA programmes and the research to date. Therefore, this methodology was chosen with a focus on a realist synthesis and the third question evolved and was developed. As this considered relevant practice and policy, it provided a bridge from theory to practice.

5.4 How and in what circumstances might an HNA programme be significant in improving self-management in brain tumour patients?

The results of study 1 and 2 led to the realist review and the primary question of, How and in what circumstances might an HNA programme improve self-management in brain tumour patients? This study was focused on interrogating the literature through a realist lens to propose (based on evidence) how and when HNA might work to improve outcomes in brain tumour patients.

The realist programme theories proposed and interrogated in Chapter 4 built on the concepts of how the actions and responses of patients, carers and healthcare professionals could influence positive or negative outcomes of HNA programmes. The interplay of these three roles became even clearer during the realist review and supports the view that to achieve improved outcomes for the brain tumour patient, there needs to be a focus on how this could work – not only for the patient, but also their family or carer, and the professionals supporting their care. The realist approach was particularly valuable as these methods recognise the interplay of different individuals, each with their own interests and objectives, who are within certain structures and institutions (Marchal et al., 2018). This helps to explain how contextual factors relevant to those individuals, and their choices and responses, influence how a programme may or may not work when interventions are introduced. This provides theories based within a social context and may limit some generalisability – but, in turn, provide theories that can be tested or used within a certain context to guide implementation (Rycroft-Malone et al., 2012).

One of the other significant areas to emerge through the iterative approach to analysis was distress. The choice of outcome proposed at the start of Chapter 4 was self-management, one of the important aims of The Recovery Package in the UK (Macmillan Cancer Support, 2020a). The other option considered was distress, as its reduction was the rationale for using HNA in North America (Bultz et al., 2011; National Comprehensive Cancer Network, 2018). However, as this research is based in the UK, self-management was deemed more relevant. As highlighted in Chapter 4, it became very clear that distress plays a central role as both a context (that influences self-management) and as an outcome on its own. Therefore, this thesis became focused on the outcomes of reducing distress and enabling self-management.

This resulted in a shift in focus from the primary outcome of self-management to a consideration of both self-management and distress. These outcomes are evident in the four refined programme theory areas indicated in the table below, reproduced here for easy reference.

Theory area	Refined Programme Theory
1. Disease-specific HNA as a mechanism to legitimise needs and support shared decision	Patients with brain tumours have needs specific to this condition (C). When practitioners use a brain-specific HNA that supports individuals in the identification of their most significant unmet needs (M - resource), this provides a subjective assessment (M - resource) of the perceived problems, particularly cognitive impairments (C). This can lead to a feeling of being legitimised (M - response) and help the

making/problem-solving	individuals and their families to direct discussions towards their most significant problems (M - response). Discussion prompted through an HNA (M - resource) may also support disclosure in patients who would otherwise be reluctant to do so (C) or, in other patients who seek to be involved in problem solving (C), allow them to reflect (M - response), becoming more able to engage. The presence of significant neurological problems (C) is likely to benefit from referral to appropriate neurocognitive specialists. This can improve the problem-solving strategies (M) and build trust (M). This can also increase the willingness of patients and caregivers to engage (M -response) and develop confidence (M - response) in finding strategies to alleviate problems through a joint approach in solving them (O). Conversely, if an HNA is used (M - resource) and the practitioner does not acknowledge the problems (M - response), this can lead to a loss of trust (M - response) and increase distress (O).
2. Caregiver distress and problems – unique challenges of brain tumour caregivers in supporting self-management	Family members or caregivers often feel a greater sense of distress and anxiety than individuals with the diagnosis (C). This is partly due to the impact of a significant caring burden (C) but they may also feel distress at the neurocognitive changes, in the patient, such as personality changes, memory loss or communication challenges. This may make the caregiver feel that the individual diagnosed with a PBT is ‘not the same person’ (C). Caregivers may also be distressed by a significant decision-making role in patients with a poor prognosis (C). A focus on the patient within the context of a patient-centred healthcare system, may mean that a caregiver’s needs or capacity for caring are not assessed (C).

	Caregivers may feel emotionally supported (M-response) through early and continued involvement of caregivers with the patient in a brain-specific HNA, followed by care planning and support for self-care (M - resource) that is also supportive of their personal or caring needs (M - resource). This inclusion may help them feel emotionally supported (M - response). This helps caregivers to rise to the challenge (M - response) and develop their capacity to support self-management (O), helping to prevent detrimental impacts on their wellbeing (O).
3. The role of self-efficacy and activation leading to positive outcomes with HNA programmes	Patients and their caregivers will have pre-existing levels of self-efficacy and of activation (readiness for change) (C), which can influence confidence in shared decision making (M - response) and facilitate problem-solving to self-manage (O). When undertaking a disease-specific HNA, healthcare professionals invite patients and caregivers to participate in related planning and discussion (M - resource). Poor prognosis (and lack of life-extending options, cognitive impairments, and high levels of distress) are important factors (C) and should be part of the practitioner's assessment of the activation as part of the HNA discussion, based on their perception of the individual/caregiver's current situation (M - resource). They should then provide a personalised approach depending on level of readiness (M - resource) – this supports the engagement of the patient/caregiver (R - response). The level of input invited or sought over decisions around care and self-management can be influenced by education (C). What levels of support are needed can be gauged to an appropriate level to increase empowerment (M - response), reduce feelings of

	anxiety and/or build confidence (M - response). It can also increase problem-solving by patients and caregivers (O) and adherence to self-management strategies (O).
4. Practitioners as a barrier or facilitator to HNA and self-management	Practitioners may have preconceptions about HNA related to the usefulness of the intervention (C), their role in supporting the process (C), and their knowledge in responding appropriately to patients' holistic problems (C). This will influence their engagement, implementation and their attitude to undertaking HNA (M - response). A brain-specific HNA focused on the relevant problems of this patient group, that supported a holistic assessment of relevant needs (M - resource), can build acceptance of the benefits (M - response). If practitioners are provided with organisational backing, knowledge of appropriate support and referral options (M - resource) and as well as training in shared decision making to respond to problems (M - resource), this can improve the confidence and competence of the practitioner in undertaking HNA (O). For the patient and their caregivers, this can result in feelings of engagement in proactively managing their identified problems (M - response) when they are involved in decisions with practitioners about solutions to alleviate their needs (M - resource) which can improve their confidence in self-management (O). Alternatively, if the practitioner has negative perceptions about the HNA process (C) and suboptimal delivery results in needs being ignored, or mismanaged (for example, with solutions that do not work for the patient/caregiver) (R - resource), this can result in feelings of loss of trust (R - response), disengagement (O) and distress (O).

Table 13 - Study 3, Refined Programme Theories

These theories provide an evidence base to take HNA forward in brain tumour patients, focusing on what might work (or might not work), for whom and in what circumstances. From Study 2, a conceptual model of ‘Needs in Brain Tumour Patients’ was developed and elements of it informed the realist review. This research provided the basis for a practice-based model which will be established and discussed in the next section. This practice-based model was developed for a brain tumour HNA programme modelled around the UK Recovery package to contextualise the contribution of this research. In line with the principles of realist enquiry, it will provide heavily contextualised guidance to inform the implementation of a brain tumour HNA programme. It outlines multiple outcomes that are influenced by a number of processes and in certain contexts (Astbury, 2018). While it will not provide an exact blueprint, it will help alert policymakers to the difficulties that they might expect and help provide causal explanations of how something might work or where it might not work in other circumstances.

5.5 How can HNA, followed by appropriate support and care, improve outcomes for brain tumour patients?

The primary research question in this thesis is ‘How can HNA, followed by appropriate support and care, improve outcomes for brain tumour patients?’. To answer this, the Recovery package(Macmillan Cancer Support, 2020a) was utilised as a starting point and built upon to illustrate and propose a practice model for this patient group. It would be useful at this point to recap the core elements, which are HNA and care planning, treatment summaries for the patient and GP, a cancer care review with primary care, and education and support for patients. Consequently, the practice-based model for a brain tumour HNA programme will be proposed around the following components – the HNA and assessment process, followed by care planning, and then support and communication interventions.

Some of the components of UK Macmillan Recovery Package (see Figure 2, Chapter 1) will be discussed in broad terms as a starting point that provides a basis for proposing a new practice model for those affected by a brain tumour. In terms of the Recovery Package, the discussion will have some relevance to treatment summaries and the cancer care review, but rather than explicitly examine these it will focus more generally on the communication, support and education needed for primary care, patients and carers. This practice-based model will focus on how these elements of HNA programmes can reduce distress and enable self-management in brain tumour patients.

This practice-based model will be described in the next section. This model reflects the outcomes of Study 1 and 2, particularly the themes and conceptual model but primarily emerged from the analysis of Study 3. As this final study considers the prior results in this thesis, the proposal of the practice model is largely informed by this, but certain aspects of these studies are highlighted when needed. The realist review highlights important contextual factors and chains of inference through many of the programme theories, but it is important to note that not all of these will be explicitly mentioned. However, recognising the complexity is a core concept of a realist study. Therefore, the full programme theories and chains of inference fully discussed in Chapter 4 should be considered in relation to any planned implementation and delivery of an HNA programme for brain tumours. The next section will present the contribution of the research in this proposal of a practice-based model. Links to aspects of the research in this thesis will be outlined as each part of the model is justified.

5.5.1 Assessment

Study 3 confirms that the assessment of unmet needs in brain tumour patients must be improved to enhance wellbeing. HCPs often do not accurately assess unmet needs and this may be related to detection, or the level of impact that the unmet need is having on the patient. It also confirms the requirement for a brain-specific HNA that allows the assessment of specific additional neurocognitive impacts, as experienced by a patient and carer, to be included in this process. These two elements would allow for a patient-centred approach in assessment and subsequent planning care.

These findings support theory 1 and provide evidence that there is a role for HNA in allowing permission to discuss unmet needs and for patients and carers to introduce these in consultations. In addition, the design and delivery should consider cognitive or other impairments that influence completion. There are potential benefits for the patient from discussions alone in terms of 'normalising' or 'legitimising' what they are experiencing. However, discussion without responses to their problems, such as referral, information and interventions can be detrimental, particularly in relation to the patient's trust in the practitioner. A lack of response has the potential to negatively impact the wellbeing of the patient, particularly through causing distress.

Study 3 also demonstrates that, in some patients, this can support shared decision making with healthcare professionals, possibly through an opportunity to reflect on their problems through completing the HNA. This helps patients to think of a way forward but might it not be addressed by healthcare professionals due to lack of knowledge, available resources or whether there is evidence of efficacy, as discussed in theory 4. These factors, along with a HCP's own perception of their role in HNA, establish healthcare professionals as gatekeepers to facilitate or block successful self-management. In addition, a patient's ability to engage in this can be very different based on several factors. Theory 3 explores a number of factors which negatively influence engagement and ability in problem-solving, such as a high burden of need and poor and higher levels of distress. This can lead to a negative impact on shared decision making capability. Another important factor to consider is the mental capacity and capability of the individual when asking the patient to be involved in shared decision making and problem-solving. Finally, if they are not able to do this, then what is the capacity and capability of carers to take on these roles?

Research examining the capacity for medical decision making related to reasoning and appreciation among glioma patients found that this was impaired in more than half the patients (Triebel et al., 2009). There is evidence that the ability to participate in shared decision making is affected by cognitive impairments and deteriorates with a higher tumour grade and psychological distress (Hewins et al., 2019). It can be important to involve the family who can support or assist with decision making and use multiple methods of imparting information. Additionally, the inclusion of caregivers can allow them to undertake a role in problem-solving and seek their perspective and needs in relation to this.

This thesis highlights carers' significant levels of distress and this is exacerbated when they are 'ignored' in consultations. Theory 2 confirms this finding, while also showing that assessing needs, information provision and support have been identified as a means to improve wellbeing and ability to support self-management. The distress of family or caregivers also causes concern for patients, so supporting this can help alleviate this impact. In study 3, there is also a suggestion that patients might prefer all support to come from their family or carer and they do not want external support. However, the carer may not share this view. Consequently, assessment of the carer's needs can improve outcomes for the caregiver. If they are struggling to cope with the demands of caring, an assessment and appropriate support can reduce their burden and distress.

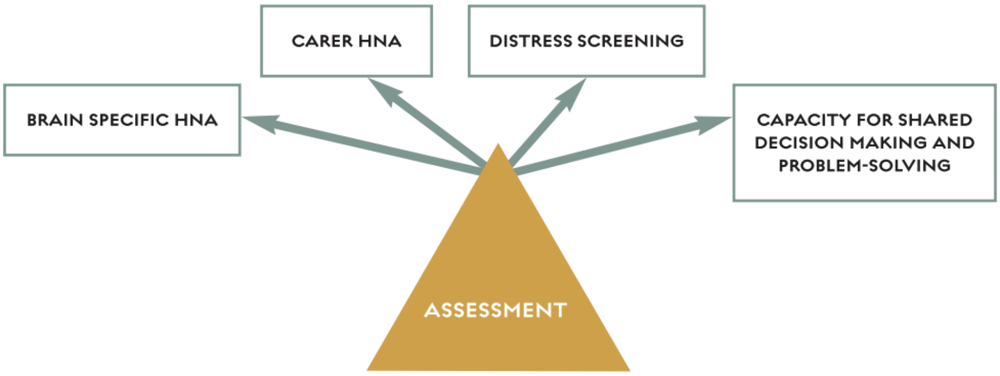


Figure 13 - Recommended Components of Assessment for Brain Tumour

Figure 13 outlines four different boxes which should make up the aspects of a comprehensive assessment. These are a brain-specific HNA, carer assessment, distress assessment or screening and an assessment of capacity for shared decision making and problem-solving. This ensures a comprehensive assessment, not only for the patient but also the carer. Neurocognitive changes have such significant impacts that assessing these and how they are experienced by both the carer and patient are important for several reasons, not least being able to offer appropriate support and referral. Additionally, some changes can have an influence on the ability to solve problems and make decisions or be a source of distress. Distress screening can be an extremely simple measure and help to highlight those who could benefit from a more in-depth assessment and possible referral. While distress has many detrimental impacts for both the carer and patient, it can also be useful to have this measure when considering abilities to self-manage. Finally, some assessment of ability for shared decision making and problem-solving could help in determining what level of support might be needed. Some patients who do not have the current capacity or capability to solve problems or self-manage may need more support. Alternatively, some might have greater capacity and capability but may require additional information to maximise their self-management abilities. These four components can all be useful as the first step in this process.

There are practice recommendations or suggestions from this review and the broader literature on how each assessment might be undertaken. The brain-specific HNA has been covered with one of two recommendations summarised in section 5.1 of this chapter. While this thesis has not undertaken a review of what tools or processes might be most appropriate for other assessments, there are potential options that could be considered for each aspect. There are at least two studies in neuro-oncology that could be consulted which examined the assessment of carers, the Carer Support Needs Assessment Tool (CSNAT) (Aoun et al., 2015) or the Caregiver Needs Assessment (CNA) (Scaratti et al., 2017). Distress screening has been done regularly with the NCCN distress thermometer and has been shown to have very good validity when assessed against longer and more complex measures, such as the Hospital Anxiety and Depression Score (HADS) (Goebel & Mehdorn, 2011). This has been widely used in brain tumour patients and is advocated as an important aspect of care to ensure that support is put in place to mitigate impacts which include low quality of life, poor therapeutic effects and satisfaction with care (F. Liu et al., 2018). Undertaking an assessment of patients' and carers' capacity for shared decision making and problem-solving is more complicated. As there is a possibility that mental capacity may be legally limited, this can be even more challenging for practitioners. The National Institute for Health and Care Excellence (2018) produced an evidence-based guideline 'Decision Making and Mental Capacity' to recommend best practice. Although the 444-page publication is useful, this lengthy document underscores the complexity and knowledge needed to assess and support this aspect (National Institute for Health and Care Excellence, 2018b). Although legal capacity is vitally important to assess, it is equally important to assess the patients' and carers' capacity, ability and desires to solve problems, as this may be variable. However, despite the complexity this is an area that requires consideration.

5.5.2 Care Planning

Study 3 is important as it helps to consolidate the findings that HNA on its own is of little benefit and could even be detrimental. Healthcare professionals and patients both seemed to have an awareness that a discussion of unmet needs without a plan to address these was of little value and possibly detrimental to the development of trust between them. It is responses with appropriate actions to a brain-specific HNA that have the potential to improve outcomes and some of these actions will be discussed in relation to care planning.

One area highlighted in theory 1 as important was the support offered in relation to neurocognitive problems and the option for neurorehabilitation. Among the patients and carers who contributed to study 2, only one mentions a referral to rehabilitation. This was valued but while they felt it was helping, once finished there was nothing else offered. Many others reported challenges which could have been considered, such as speech difficulties or mobility issues (Kushner & Amidei, 2015). The evidence supporting Theory 1 highlights a lack of referral for neurocognitive problems, which could have been improved through referral and intervention. This problem seems to be an issue in the UK, with the National Institute for Health and Care Excellence (NICE, 2018) also highlighting inconsistency in resources, referral and eligibility for neurorehabilitation resources for brain tumour patients. Currently, there are no nationally agreed standards for neurocognitive rehabilitation and support for brain tumour patients (Day, Gillespie, et al., 2016) and there is a scarcity of cancer services specifically addressing patient cognitive decline and reducing the impact of neurocognitive changes on carers. There could be an opportunity to learn from other teams involved in neurorehabilitation, such as those supporting services in dementia or head injury, and models for services could be considered.

However, these conditions often have more global impacts on the brain (Bergo et al., 2016). There has been linked research on neurorehabilitation for brain tumour patients, with some only appearing a few years ago, possibly due to the poor prognosis, but what was undertaken demonstrated some positive results (Bergo et al., 2016).

It is also challenging to ensure that managing and planning for other support needs is patient-centred and gives patients and carers choice and control. While evidence-based care pathways and guidelines are important in ensuring that quality and care is delivered efficiently, they may not consider 'what matters' to people and their capabilities and needs. As discussed in theory area 3, HCPs need to tailor their approaches for supportive care planning and supporting self-management based on the recipient's needs and expectations. Study 2 highlights how some patients and families seek to self-manage as much as they can. However, at times they lack information on how to do this. This lack of information can lead to needless suffering as beneficial interventions are not implemented or the patient and care have increased anxiety managing their needs. This could be exacerbated by avoiding care with their primary care teams as they may not have trust in the knowledge of their primary care team. For those patients, information shared from the specialists with both them and primary care team can help this, as well as timely access for specialist advice when additional information is needed.

One way of balancing these is considering stratifying care, including what can be self-managed based on the important factors or contexts. HNA and other assessments noted above can play an important role in determining these and theory 3 explores how a personalised approach based on capacity and capability could empower patients' carers to problem-solve and self-manage. However, others may require higher levels of support and direct care. Factors such as poor prognosis, distress or low self-efficacy may present barriers in their ability to manage. A stratified pathway could be effective in supporting this.

Stratification has been used with breast, colorectal, lung and prostate cancers within NHS England's cancer strategy (NHS England, 2016; NHS Improvement, 2016). HNA is important for considering suitability for this pathway. Other criteria includes the level of risk associated with cancer type, short and long-term effects of treatment, other co-morbidities, the patient's ability to manage, and the level of professional involvement required.

In a good prognostic group, a significant level of advised self-management pathways were used, with up to 75% on this pathway for breast, 45% for colorectal and 30% for prostate. Alternatively, within a poorer prognostic group, lung cancer, all would require a professionally managed pathway (NHS Improvement (Cancer), 2012, 2016). However, this has benefits for both the health service and patients, with tailored care packages reducing unplanned admissions by 6-8% at pilot sites. By having a more flexible and patient-focused service they were also able to change the face-to-face appointments to telephone consultations in 20-25% of cases, potentially reducing resource usage and preventing hospital visits. Additionally, 18% of appointments arranged at the request of the patient or another healthcare professional were asked for to avoid emergency admissions.

While full self-management pathways might have very limited suitability for brain tumour patients, stratification of more versus less direct care, and more versus less support information for self-management could reduce the negative impacts of cancer and treatments, not only for the individual but also significant others. This could ensure that those who need more direct care have a clear pathway, but those who are capable can be supported to self-manage whatever aspects they can. Implementing tailored packages of care, such as neurocognitive rehabilitation based around specific deficits would require a more responsive and flexible service. However, there may be savings elsewhere, such as preventing hospitalisations. This has the potential to empower patients and their carers to feel that they have some control over the impacts of the brain tumour.

Theory 4 highlights the considerable challenges for healthcare professionals in supporting individuals and carers with such a complex condition. One of the areas that healthcare professionals felt was a particular challenge was responding to unmet needs when they lacked the knowledge or resources to do so. Patients, carers and healthcare professionals were all aware of the negative impact of not reacting and this was a barrier to implementing HNA. However, signposting for self-care, knowledge of services and other health professionals' roles and skills, support of the third sector and interagency working can help with this. The two tools in the Macmillan Recovery Package which support the role of primary care are the treatment summary and cancer care review. The treatment summary goes to the patient and primary care team and is an important component, but the findings of this research highlight that additional information and access to a specialist for advice is essential. However, sharing earlier assessments can improve the continuity of care. Carrying out an HNA at this point can help the cancer care review meet the needs of the patient and carer at the end of treatment and better support needs that might be identified. However, other assessments, such as a neurocognitive assessment, remain important and a pathway for continued assessment and support should remain with the specialist neuro-oncology providers.

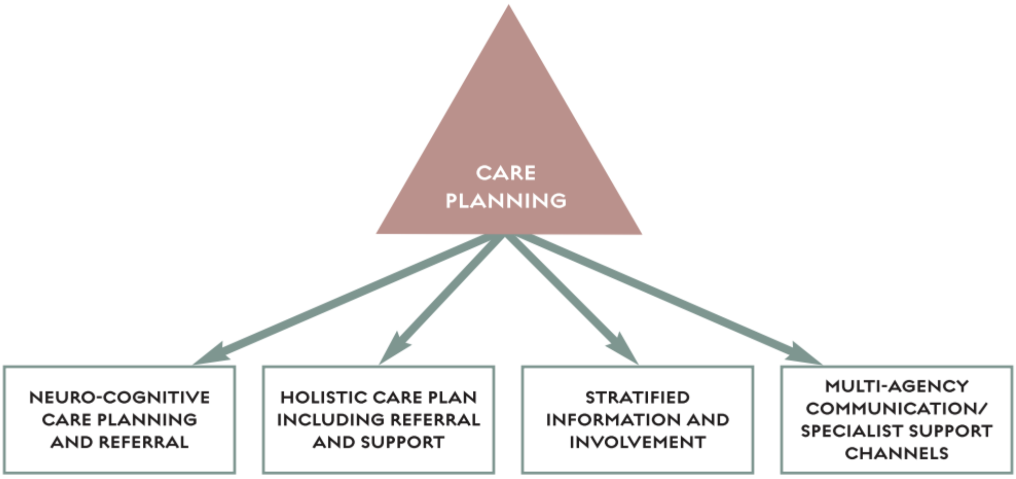


Figure 14 - Recommended Components of Care Planning for Brain Tumour Patients

Figure 14 shows four boxes that represent recommendations in care planning, neurocognitive care planning and referral and overall holistic care plan, stratified information and self-management plan, and pathway planning. Neurocognitive rehabilitation and support have the potential to provide improvement or even reduce the pace of deterioration.

5.5.3 Support and communication interventions

The execution of care planning is very important and there are broad considerations which study 3 shows as being crucial to acknowledge in the implementation of interventions to maximise the intended outcome of improving distress and self-management.

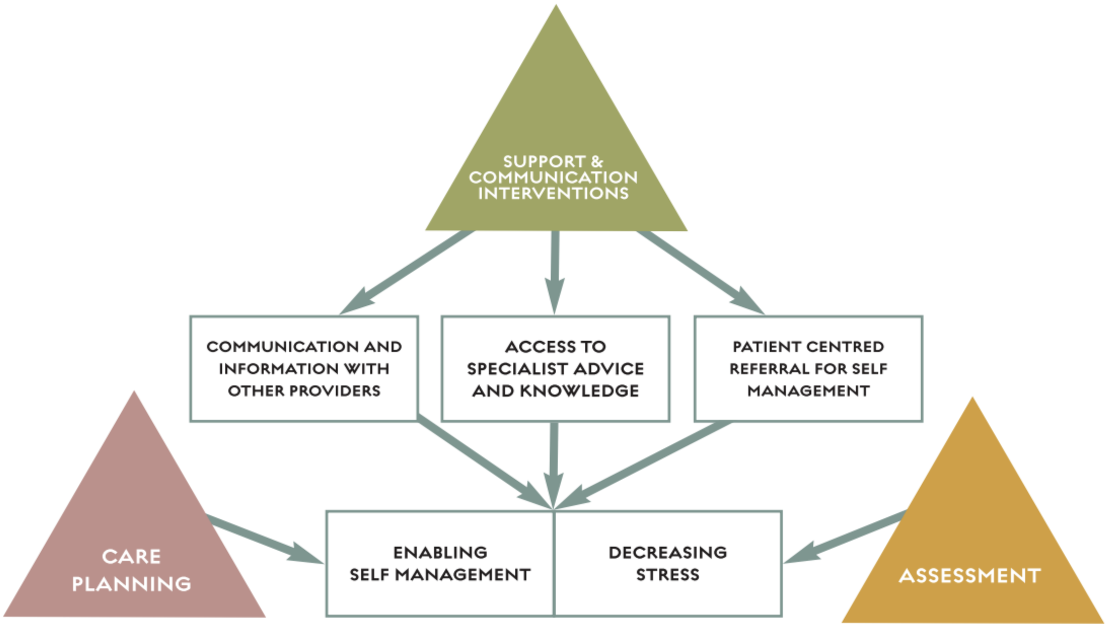


Figure 15 - Recommended Components of Support and Communication interventions for Brain Tumour Patients

In Figure 15, the green triangle at the top represents the final components of HNA package recommendations concerning support and communication interventions. The three middle boxes indicate the three areas of actions, which should be communication and information with and from other providers, access to specialist advice and knowledge, and patient-centred referral for self-management. These three actions, if implemented with the recommendations for care planning and assessment described above, can enable self-management and reduce distress, which are the desired outcomes represented by the two boxes at the bottom of the figure.

This research highlights the influence of communication on many levels. While HNA is a tool, the assessment can only be made with good communication. Theory 1 shows that good communication between the patient and/or carer and healthcare provider is a mechanism that can help build trust and deliver outcomes of reduced distress and self-management. As already emphasised in the discussion of assessment and care planning, a patient-centred approach is necessary. This patient-centred approach also needs to be considered in the patient and carers' expectations for self-management and provision of self-management advice. Theory 3 suggests that the wrong level of advice (too high or too low) could result in increased anxiety, but appropriate levels could build confidence. Additionally, theory 2 supports the importance of listening and assessing and then providing appropriate support, specifically for carers in this area. This can result in a feeling of emotional support and, if gauged appropriately, can help them rise to the challenge of caring.

The lack of knowledge of conditions or perceived mismanagement of symptoms outside of specialist oncology services was a common issue for many of those affected. This was reported to start in the diagnostic period, with some cases continuing throughout. Diagnosis of brain tumours in primary care is problematic as patients present with a lack of clear predictive symptoms (<1%) outside of seizure (1-2.3%) (Schmidt-Hansen et al., 2015). This delay means that many patients present as an emergency preceded by multiple primary care consultations. While this paper was not focused on this issue, it was experienced by a number of participants and had an effect on patients' and carers' trust and confidence in the knowledge and abilities of primary care (or other services). This diagnostic problem is not easily solved (nor within the scope of this thesis) but communication and information sharing between care providers helps to optimise the support from primary care.

There are no nationally agreed standards at present for how neurocognitive rehabilitation should be implemented among brain tumour patients (Day, Gillespie, et al., 2016) and there is a scarcity of cancer services specifically addressing patient cognitive decline and reducing neurocognitive changes on carers. There has been some limited research on neurorehabilitation for brain tumour patients, with this demonstrating some improvements (Bergo et al., 2016). However, while awaiting additional research, referral pathways for support should be set up as advised within national guidance so that healthcare practitioners can refer when patients would like to pursue this (National Institute for Health and Care Excellence, 2018).

5.6 Meeting the holistic needs of brain cancer tumour patients – situating the finding in the current evidence

This study focuses on developing evidence of how HNA could improve outcomes for brain tumour patients. HNAs are integrated into the Recovery Package and this is the primary use of these assessments for cancer within the UK (Macmillan Cancer Support, 2015). However, the focus of the UK Recovery Package is on survivorship, recovery after treatment, and supporting the development of self-management in patients to reduce demands on services through improved wellbeing. There is a clear focus on 'recovery after treatment' or survivorship, and linking with interventions to support recovery (Macmillan Cancer Support, 2018) but questions surround whether the UK model of HNA and its focus on 'recovery' or survivorship is appropriate for brain tumour patients. As a significant proportion of brain tumours have a very poor prognosis and recovery is unlikely for the majority of these patients, this focus on recovery may seem irrelevant for many patients. Then again, if we look at the components of survivorship care as defined by the US Institute of Medicine (Institute of Medicine and National Research Council, 2006), it still could be an appropriate model for even poor-prognosis brain tumours. They define survivorship as:

- Prevention of recurrent and new cancer.
- Surveillance for the spread of cancer and physical and psychological acute and late effects.
- Intervention for consequences and treatment of cancer (holistic impacts). This includes cancer survivors and their caregivers.
- Coordination between services to ensure all needs are met.

The components may all have some degree of relevance depending on the patient and prognosis. However, surveillance for effects, interventions and coordination of services are very relevant and possibly even more important in a poor prognostic group.

Another aim of the Recovery Package is the development of self-management in patients to reduce demands on services through improved wellbeing. While these aims may also be relevant for some brain tumour patients, this thesis supports a focus on reducing distress and a recognition that, in some cases, services may need to increase (rather than decrease) to achieve greater wellbeing. The benefits of self-management remain important and valid, although this patient group and their families may have more challenges.

Assessment of physical and psychological effects and detecting holistic impacts is also a key component. Research in other areas of cancer, as previously reported, indicates that HCPs do not accurately assess patients' unmet needs.

Research continues to support this as the case, with a more recent study demonstrating that nurse specialists were only able to identify between three and six of the top ten concerns of patients within specific cancer types (Mitchell, et al., 2018). Unmet needs and problems affect QOL and also correlate with increased psychological distress (Carlson et al., 2012). For these reasons, and to support improved self-management, cancer policy in the UK advocates that a holistic assessment of needs and care planning should be provided to all cancer patients (National Institute for Health and Care Excellence, 2018) with variations of this in the other three UK nations. Despite this, the Recovery Package appears not to be widely implemented, with potentially the poorest implementation in brain tumours.

This thesis provides evidence that would support the aims of self-management and reduction of distress as an achievable outcome resulting from the implementation of an HNA programme in this population. As Chapter 4 is a focused iterative realist synthesis which presents broad discussion of the HNA literature, the final consideration of the wider literature will focus more specifically on the outcomes of distress and self-management.

5.6.1 Self-management as an outcome of HNA in brain tumours – consideration of the literature

Self-management has been highlighted as a means to improve wellbeing in chronic disease for a number of decades (Corbin, 1998), and cancer is widely considered to be a chronic disease (World Health Organization, 2020). While the definitions of self-management are numerous, Barlow et al. (2002) present the view that it is a person's ability to manage symptoms and the consequences of living with a disease that includes physical, social, emotional and lifestyle changes. Corbin and Strauss (1991) highlight that multifaceted management includes managing the medical aspects of the illness, changing life roles, and the psychological consequences of the illness.

A healthcare professional’s role in supporting self-management is the provision of education and support to increase skill and confidence in managing problems, regular goal setting and problem-solving support (McCorkle et al., 2011). They also advance the view that the required skillset for self-management includes problem-solving, decision making, resource utilisation and working in partnership with HCPs while taking the required actions forward, all set within the context of the family (McCorkle et al., 2011). The list of activities that could be considered self-management is extensive and for brain tumours, like many chronic illnesses this can include undertaking advised follow-up, symptom monitoring and management, psychological wellbeing, healthy lifestyle, managing emotional wellbeing, etc.

These activities align to a more recent mid-range theory, Riegel's 'Theory of Self-Care of Chronic Illness' and this focuses on three strands of self-care or management which include; self-care maintenance, self-care monitoring, self-care management (Riegel et al., 2019). Activities in cancer clearly align under these headings and as highlighted in the editorial by Biagiolo et al., (2021) this has clear relevance to cancer with its focus on self-management exemplified by the UK Recovery Package (Macmillan, 2018) and Transforming Care After Treatment initiatives (Scottish Government, 2016b). This paper also emphases the importance of self-care confidence and this was supported by the programme theory and practice model proposed in this thesis that emphasise the importance of assessing capacity for self-management.

However, in brain tumour patients, this needs to be considered in the context of the family or carer. Due to the complex cognitive skills needed to fully participate in self-management, we need to consider these differently for this group. The definitions of self-management discussed in Chapter 4 focused on strategies and activities *individuals* undertake to deal with the physical and psychological consequences of cancer which promote survival, health and wellbeing (Foster et al., 2015 and McCorkle et al., 2011). However this research has helped to demonstrate that in brain tumour patients, this is an aim for not only for the brain tumour patients but also for their carers. In individuals who have experienced a brain tumour, self-management may be a shared and at times an increasing responsibility of the carer. The demands of this can be significant for the carer as they have to focus on the needs of the brain tumour patient as well the physical and psychological burden of caring. The involvement of carers and consideration of their role in supporting patients with brain tumours can improve their capacity to support the individual with a brain tumour. This could help them to achieve the aims of improving health behaviour, reducing demands on services, improving communication between patients and health care providers. The provision of information on coping with neuro-cognitive changes, where to get additional specialist advice when needed and supporting their psychological wellbeing can all maximise their ability to support self-management. This also should include strategies that can support the development of higher self-efficacy. This research supported that self-efficacy can be a moderator of behaviour that supports the development of self-management (Baydoun et al., 2018). In family groups, perceptions of the level of self efficacy were often shared by the carer and patient. However, it is possible this could be developed through having access to specialist information and support that is readily available to help them cope successfully. Through having this more holistic view of inclusion of the carers and developing the perception that they can cope and manage, higher levels of self-management are potentially achievable.

Study 2 and 3 highlighted that there are often many unmet needs and that information or referral for supporting these needs can be inadequate, including when patients are seeking to manage their symptoms. Research to support self-management in brain tumours is very limited and a review of self-management interventions in cancer with experimental or quasi-experimental designs published in 2019 (Cuthbert et al., 2019) found only one study (Khan et al., 2014). This was a cohort study which compared patients diagnosed with a glioma who were allocated a multidisciplinary rehabilitation programme to those allocated standard treatment (no rehabilitation). There were improvements in the Functional Independence Measure in the motor outcomes of ‘self-care’, ‘sphincter’, ‘locomotion’, ‘mobility’ ($p < 0.01$ for all); and ‘communication’ ($p < 0.01$) and ‘psychosocial’ subscales ($p < 0.05$), with small to moderate effect size ($r = 0.2–0.4$). While there were statistically significant and clinically relevant improvements, the authors highlight that delivery is expensive, fragmented and lacks protocols. However, despite this, the National Institute for Health and Care Excellence (2018a) supports the consideration of neurorehabilitation for all brain tumour patients. Although there may be some increase in demand on resources, this group would only make up a small proportion of overall referrals, so the increase would not be excessive.

This research provides unique and important evidence that can help when healthcare professionals implement interventions or provide support and education to maximise self-management for patients and their carers who have experienced a brain tumour.

5.6.2 Reducing distress as an outcome of HNA in brain tumours – consideration of the literature

The impact on carers or families when an individual is diagnosed with a brain tumour is significant and detrimental. As highlighted in Chapter 1, the impacts go far beyond physical manifestations and the psychological or social impacts can be equally or more devastating. The challenges and experiences of patients in this research are supported by wide-scale surveys in the UK. The Cancer Patient Experience Survey demonstrated that brain cancer had the worst quality of care outcomes of any tumour type (NHS England, 2017). The multifaceted impact of cognitive changes in particular contributed to this and this thesis demonstrates that inadequate assessment or support of needs can also exacerbate distress.

The patients and carers in study 2 reported a 'loss of self', and loss of activities, roles and interactions as a result of their brain tumour. This resulted in distress and is supported by study 3. Distress was often in response to symptoms, many related to neurocognitive deficits such as balance, behavioural changes and memory or communication difficulties. Other symptoms such as fatigue also contributed. These changes often involved a social withdrawal, while some were related to role changes (work). Others withdrew from social contacts due to a lack of confidence exacerbated by cognitive changes, or because previous contacts withdrew their involvement in the brain tumour patient's life. This decrease in social contacts was also noted in a meta-synthesis by Cubis, Ownsworth, Pinkham and Chambers (2018). This was strongly linked to functional impairment but also compounded by practical issues, such as a loss of the ability to drive. Their review demonstrated how distressing and detrimental this was to their wellbeing. However, those who were able to draw on social networks or create new social contacts experienced greater social participation.

Subsequently, this research group undertook a study in brain tumour patients which examined the correlation between perceived cognitive and physical impairments, anxiety and depression, and confidence in social support (Cubis et al., 2019). This demonstrated (similar to many studies) that there is a clear correlation between perceived higher levels of impairment and depression and anxiety. However, for those with high levels of social support, anxiety and depression can be moderated, even with levels of high impairment. These authors conclude that, while social isolation occurs regularly, facilitators could help to guide interventions to improve social networks and through this support the individual's wellbeing. This viewpoint is important to consider in terms of unmet needs and distress, as supporting the development of social interaction could be an important aspect of helping patients to cope with and manage needs.

This thesis also draws attention to the fact that there is often a significant caring role due to disabilities imposed by the tumour and neurocognitive impact. Several studies have explored the psychological impacts and experiences on caregivers of caring for a person with a diagnosis of glioma. One study of glioma patient carers demonstrated rates of 58-61% of moderate to high distress over the first six months of diagnosis (Halkett et al., 2018). Another study of high-grade glioma reported that 78.1% of carers met the criteria for distress and referral for psychological support at diagnosis (in contrast to 37.5% of patients) (Trad et al., 2015). Elsewhere, a review examined the lived experience of patients and caregivers with high-grade glioma. Although this represents the group with the worst prognosis, many of their findings in relation to the causes were similar, such as isolation, loss of relationship, and the burden of responsibility (Sterckx et al., 2013). In a subsequent study examining caregivers, one of the main areas of distress was the loss of the 'patient's old self' but also the loss of their 'old life' prior to diagnosis (Coolbrandt et al., 2015).

Additionally, managing the changing roles for the individual with a brain tumour, as well as the increased roles and stressors for significant others such as decision making, can be unique when we compare this to other cancers. A patient support charity advises of the benefits of an HNA to support discussions around advanced care planning. This can be used for legal purposes, such as a power of attorney, but also to help set out decisions that take into account priorities or wishes when others have to make decisions for the patient (The Brain Tumour Charity, 2020). Yet, in a review of end-of-life care in PBT patients, only 37% had advanced care plans in place in Britain (compared with 46% in Dutch patients and 6% in Austrian patients) (Von Bueren et al., 2016). A qualitative study of 15 neuro-oncology specialists highlighted there was an avoidance by the HCP of discussions of advanced care planning. The reasons for this included a culture of avoidance of death and dying, avoidance of emotive discussions, lack of time, perception of professional role and understanding of advanced care planning (Llewellyn et al., 2018). Advanced care planning could be an important mechanism to help with important decisions at the end of life to reduce the carer's burden but also help provide guidance for other aspects of managing the disease as patients would wish when their capacity to make decisions is lessened.

Distress for both brain tumour patients and carers is greater than in other cancers because of complex and varied needs due to the presence of neurocognitive deficits. These deficits often result in changes in roles, social isolation and other psychological impacts. The impact of these is experienced differently compared to people diagnosed with other cancers, so support needs to be different for brain tumour patients. This thesis provides an evidence-based model of how practice can be taken forward, with considerations for clinicians and researchers on how interventions could be designed and delivered for certain patients and carers in certain contexts. The next section will outline the implications for practice and policy more fully.

5.7 Implications and recommendations for practice and policy

This thesis focuses on investigating HNA packages in brain tumour patients, a practice-based issue which is recommended for all cancers in the UK. An informal look at the three neuro-oncology units in Scotland to examine implementation was conducted at the beginning of this study and the results indicated that it is not routine and is undertaken only in the context of previous research (Rooney et al., 2014). While statistics for Scotland are not available, a baseline report from NHS England for 2017 (NHS England National Cancer Programme, 2018) involving 109 trusts reported that brain and CNS tumours had the highest completion rates of HNA at 92% – but then the care planning completion drops to approximately 40%. Treatment summaries (for primary care and the patient) were at 0% and the provision of health and wellbeing support was 42% – the second-lowest level – compared to 92% for breast cancer patients. It is also notable that, in this same period, the England-wide Cancer Patient Experience Survey found that only 25.5% of brain tumour patients reported getting a care plan (NHS England, 2017). The challenges of managing this complex disease have been discussed at length and this next section will propose a practice-based model that has emerged from this research.

5.7.1 How could HNA programmes be reimagined for brain tumour patients to support self-management?

Figure 16 below illustrates how an HNA package could be delivered to help support individuals affected by a brain tumour. This brings together all the elements of this thesis to propose how such a programme could be designed and implemented. While this has not yet been tested in practice, there is potential for this to be developed to support HNA in a practice-based setting and help ensure that outcomes to improve wellbeing for patients and carers are achieved. This will be pursued in post-graduate research to test and refine the practice-based model and programme theories, focused on examining the effectiveness of an HNA programme in reducing distress and improving appropriate support for self-management.

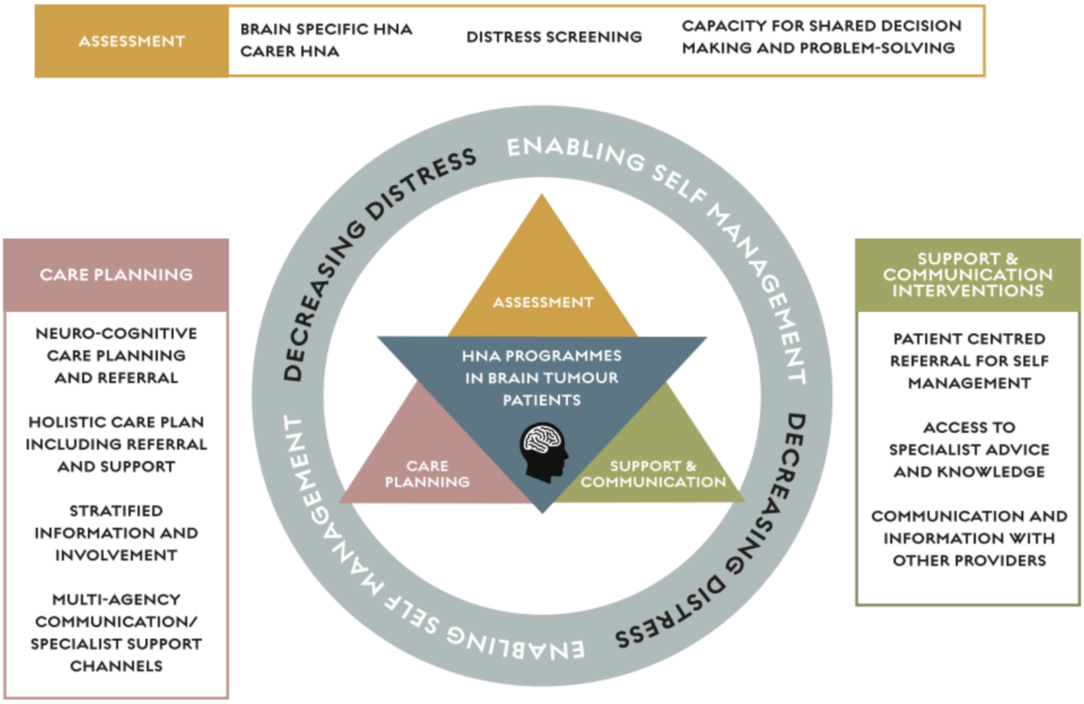


Figure 16 - Final Practice Based Model – HNA Programmes and Support Pathway for Brain Tumour Patients and Carers.

In Figure 16, we can see the final practice-based module developed from the analysis of this thesis. The boxes around the edges represent the individual components that feed into assessment, care planning and support and communication interventions, which are represented by the three triangles making up the components of the HNA programme. The two desired outcomes for this programme, reducing distress and enabling self-management, are in the outer circle.

The next section will summarise the components of each section, highlighting the strengths of this model but also identifying some areas which require further development.

Assessment

The proposed practice model has three main areas of assessment. These are patient and carer holistic needs assessment, distress screening and an assessment of capacity for shared decision making and problem solving. The starting point in undertaking an HNA remains utilising a brain-specific HNA which includes the significant and detrimental impacts of neurocognitive deficits. One reason why clinicians tend not to use PROs is because of their perception that the existing HNA is not fit for purpose (Yang et al., 2018) due to the mismatch between brain tumour symptoms and general cancer HNA tools. An HNA which includes brain tumour symptoms and needs could overcome this resistance by some clinicians. However, a holistic assessment of needs is required for planning care and support to minimise the detrimental impacts of the disease. This study also demonstrated that HNA needs flexibility in the approach to ensure, as much as possible, that neurocognitive impairments do not prevent completion. For example, some patients may need assistance, some may prefer to complete at home and others in clinic. Carers also may experience significant disruption to their lives and an assessment of their needs is also important to help support them in their role and minimise effects on their wellbeing. This model recognises the importance of assessment of both the patient and carer as important first steps. The form alone should not be viewed as the assessment in itself, but rather as a support to help identify needs during a consultation process.

Additionally, with both the patients and carers experiencing such high levels of distress, it would seem prudent to introduce distress screening as part of this process. This would also allow it to be monitored over time to evaluate the impact of any process. As demonstrated in Canada, the distress thermometer is easily integrated with HNA (Howell, Keshavarz et al., 2015) and is easily completed with good psychometric properties (Miller et al., 2013). To date, there is only one randomised study (Ownsworth et al., 2015) which has demonstrated the positive impact of a multimodal psychosocial intervention to improve depressive symptoms in patients with brain tumours, which indicates the need for additional research to focus on this area. Despite a recent Cochrane review, guidance for using pharmacological treatments for depression remains unclear due to significant adverse effects, such as the risk of seizures (Beevers et al., 2020). Nonetheless, this remains an important question due to the high prevalence of depression in brain tumour patients. This recommendation would allow assessment and then, for those that need it, a consideration of options for care or referral to help alleviate this psychological distress. This is also relevant for caregivers, although there is some different evidence that could be considered.

There has been a limited amount of research into what can help alleviate caregiver distress, based on a review published in 2016. Only one comparative study of cognitive behavioural therapy, which demonstrated that this intervention prevented deterioration of mental health, has been completed (Sherwood et al., 2016). Despite the significance of this issue, no further comparative studies evaluating interventions were found. While the cancer literature has some relevant studies on what might be done to help caregivers – the loss of the 'patient's old self' may be the most significant source of distress – perhaps there is merit in seeking guidance from studies in areas where caregivers might also experience distress caused by neurological changes experienced by the family member, and the subsequent caring burden. More extensive research has been conducted on interventions which focus on improving psychological outcomes for carers of people with dementia. CBT and psycho-educational intervention delivered cost-effectively with paraprofessionals, in groups or remotely, remain effective and have been shown to reduce psychological morbidity (Cheng et al., 2019). This service could be extended to support carers of patients with brain tumours.

An assessment of capacity for decision making and problem-solving which has links to self-management will provide a basis for planning what information, support and referral is needed. However, carer readiness and capability to support self-management should also be assessed. Assessing but also supporting caregiver 'mastery' or perception of control over the care situation has been demonstrated to correlate with increased survival in one study of glioma caregivers (16.1% risk reduction in patient death, 95% confidence interval, 0.771-0.913; P<.001) as well as reducing their distress. If further research demonstrates a survival advantage, this should include an assessment of information needs that can facilitate a level of shared decision making suitable for that patient and carer. After a full assessment is completed, care planning should then be undertaken with specific consideration of stratified care pathways.

However, there are some areas that this research does not examine. The time to complete a HNA is not advised within model presented in this thesis, but policy advises this should be personalised for each patient. However, it might not be idea to undertake when patients are seeking HCPs to discuss significant changes or updates on their condition (such as when they might receive diagnostic or progression information) (Macmillan Cancer Support, 2020b). The role of an electronic HNA is not explored in any great detail in this thesis, but there appears to be many potential benefits, such as linkage to care plans and support and communication between providers. However, the needs of patients with neurocognitive symptoms would require a customised HNA and have provision that allows others to support completion because of challenges that many of these patients might experience. Technology potentially has a great deal to offer, with options for 'spoken' HNA questionnaires and records that report trends and changes easily for patients and carers over time.

Care planning

In responding to needs there are a number of areas relevant to care planning that this model recommends. The care plans should be holistic, including a focus on neuro-cognitive support and referral as appropriate. The care planning also needs to include improved access for patients, carers and primary care to specialist neuro-oncology services with clear and responsive communication to better support shared care and self-management. Finally, the care plan needs to be stratified around the capacity and capabilities of the patient and carer to self-manage. The following paragraphs will elaborate on these areas.

The first recommendation is that care planning should be developed with the patient and family, then shared with others to deal with their holistic unmet needs. It can be detrimental to the wellbeing of the patient and their trust in the healthcare professional if the professional does not appear to react to identified needs. As highlighted earlier in this thesis, staff may not do this for a variety of reasons which include not seeing it as part of their remit or not knowing what to do about it. Recent surveys of cancer patients' experiences in England have shown that brain tumour patients have a poorer experience (Brainstrust, 2019; NHS England and NHS Improvement, 2018) and Brainstrust, a patient support charity, suggests may be a contributing factor is a lack of care plans (which are part of the HNA pathway), with only 26.5% of brain tumour patients reporting as having this in 2018 (Brainstrust, 2019), the lowest of all cancer types. Around one in ten people (9%) responded that they did not know or understand what a care plan was. In Scotland, only 30% of all cancer patients were given a care plan (Scottish Government, 2019), compared to 35% in England (Quality Health, 2018). In the 2018 Scottish survey, brain tumour patients' responses were not published separately from other cancers, (due to responses of less than 50), however, in the 2015 survey, only 7 out of 39 patients or 18% reported they had received a care plan (Scottish Government, 2016). The lack of provision of a care plan demonstrates a clear gap in care in Scotland, as well as England. The authors of the Scottish report highlighted that care planning was an important factor as those who did not have a care plan were significantly more negative on 45 out of the 47 questions (Scottish Government, 2019). This is a key area for improvement, not only in brain tumour patients, but in all tumours.

As part of care planning, neuro-rehabilitation should be a priority as this is often not integrated or considered despite the significant detrimental impacts of neurocognitive deficits on many areas of life. In addition, referral and support is inconsistent and inadequate. While the evidence base is limited, there are some indications of positive outcomes of neuro-rehabilitation. Guidance from NICE (National Institute for Health and Care Excellence, 2018a) supports this and therefore this recommendation is justified.

Stratified support and information based on the carer’s and patient’s capacity and capability for self-management can help to meet needs and empower but they can also ensure that expectations are matched by the wishes and capacity of the patient and family. By screening this population for those that may be struggling with managing their condition more independently, different pathways of care could be introduced and appropriate interventions that can help mitigate unmet needs should be offered, within the resources of the health service. One option suggested by Hutchison et al. (2006) is a stratified tiered approach with five levels of psychological care, from first level routine support to the highest level of specialist external mental health service referral.

All of this needs to be underpinned by a clearer communication strategy that involves the patient, family, specialist neuro-oncology clinicians and primary care. The role of primary care is vital to ensure management of unmet needs takes place in the community and with local support. However, this study demonstrates that, in many cases, the relationship with primary care is not ideal. This role can be optimised by sharing information in the care plans, but also providing responsive conduits from primary care to specialist help. The next section will provide some additional specific information about interventions and support that should be considered as plans are implemented.

Support and communication interventions

Due to the unique and complex side-effects, particularly the neurocognitive impacts of a brain tumour, support from the specialist neuro-oncology service is needed. As highlighted in 106 trusts, treatment summaries were not provided for any of the patients for their primary care providers (NHS England National Cancer Programme, 2018). This communication pathway between specialist care and primary care is needed to help primary care better support this group of patients. Treatment summaries and shared care planning have a strong potential to be a beneficial mechanism to meet the needs of patients with a rare cancer and neurological condition. The role of such an approach would increase the confidence of patient and carers that all providers have the requisite knowledge to give support where appropriate. That said, as the problems are so unique in providing a responsive service, seeking advice for primary care could be an important component of helping support patients and carers. This is also important when we want patients and carers to self-manage as much as possible.

Trusted healthcare professionals can be a key element in facilitation of self-management services. However, the level of information sought, desired or needed will vary depending on individual needs, educational levels and social support. Such information provision could lead to greater levels of self-management and may be a valuable addition to a model of supporting self-management and holistic wellness in brain tumour patients and their families.

5.7.2 Summary of practice and policy recommendations

HNA programmes have the potential to improve outcomes for brain tumour patients and their informal carers and implementation and evaluation of these should be a priority for this patient group. However as outlined above there are important modifications that should be considered to current UK guidance for cancer. Modifications in the assessment process for this patient group include using a brain tumour specific HNA, but the use of this as a measurement tool is not recommended, and this should be seen as an aid to assessment, complemented by robust communication skills. The assessment phase should also include assessment of the carer’s needs, distress screening, and an assessment of the patient’s ability to self-manage. Improved communication and access to the specialist neuro-oncology team, for patients, carers and primary care are important areas for development. Due to the neurocognitive issues and more substantial needs than other cancers, these areas should be priorities for enhancement in the post-treatment care pathways. To support self-management there should be increased referral and information for the management of neuro-cognitive symptoms as well as distress.

Implementation of the Recovery Package remains suboptimal, with a survey in England in 2017 showing that only 56% of Macmillan GPs and NHS leaders were confident of the Recovery Package continuing to progress in England (Macmillan Cancer Support, 2017). There are currently no UK-wide or devolved Scotland implementation statistics at the time of this research however, based on communications with the Scottish Adult Neuro-Oncology Network, HNA was not routinely implemented in any of the five Scottish neuro-oncology centres (Scottish

Adult Neuro-Oncology Network, personal communication, November 11, 2020). Although it has been implemented into policy and strategy, the lack of a strong evidence base for some aspects could affect uptake by professionals and their approach to implementation as this research proposes. Therefore, continued evaluations of aspects of this programme under a realist lens could help improve motivation and acceptance. In addition, as highlighted in this review, there needs to be an increased evaluation in groups with higher needs and poorer prognoses.

In the majority of the literature, it was emphasised that nurses are the main professional group who implement HNA programmes. Yet, in the UK (as in many other regions), nurse input into policy can be limited by numbers and/or confidence. There is only one representative from the nursing profession on the independent National Cancer Advisory group – which monitors the NHS England policy of implementation of Achieving World Class Cancer Outcomes, including monitoring of HNA programmes – while six members are from medical professional organisations (Cancer Research UK, 2018). Nurses at all levels, need to develop their confidence and voice to influence areas of cancer care policy to support the professional needs of the workforce to implement 'best practice' (Rasheed et al., 2020). As highlighted by Anders (2021), barriers for nurses include a lack of political sophistication, gender issues, work demands limiting time, and a lack of confidence. However, as subject experts, nurses need to overcome these barriers and they should be central to policy changes and improving patient care.

The last year (2020) has brought unprecedented challenges with the Covid-19 pandemic. The implications for cancer are significant, with the impact of delayed treatment and diagnosis likely to be profound with diagnoses at a more advanced stage, in many cases leading to higher mortality. Despite this, Macmillan has urged the UK Government to continue to offer HNA programmes (like the Recovery Package) as much as possible, with the use of electronic HNAs being promoted as one important strategy (Macmillan Cancer Support, 2020c). While there are many competing priorities, supporting self-management through this process has never been more important as the danger from Covid-19 continues for this vulnerable group of patients and carers.

5.8 Recommendations for further research

As this study has highlighted, healthcare policy supports increasing self-management and the increased role of shared decision making in this in providing support and choice for patients and carers. However, as highlighted, the readiness and capacity for this in patients and carers may be variable. This may be due to factors prior to the diagnosis such as education, a preference for a passive role or as an impact of the diagnosis and disease. Research is needed that can guide practitioners on how to implement shared decision at the right level for different patients and carers to maximise choice and autonomy. This includes not placing expectations on those that prefer a more passive role, that may end up increasing stress and anxiety. This research should seek to be inclusive of the carers or family and focus on how this can best be implemented in the presence of neuro-cognitive impairments.

This research showed that one of the barriers in undertaking and responding to HNA for healthcare professionals was knowing what do in response to identified unmet needs. It has been highlighted that brain tumours have numerous areas lacking a strong evidence base which creates challenges in the implementation of evidence-based practice with supportive care and supporting self-management. While some research into other cancers may give some insight, in others it will be of limited value as brain tumours are a neurological disease and responding the unmet need related to the neuro-cognitive impairments is a priority. Guidelines for brain tumours such as Evidence Reviews for Supporting People Living with a Brain Tumour (National Institute for Health and Care Excellence, 2018) or the European Association for Neuro-Oncology (EANO) Guidelines for Palliative Care in Adults with Glioma (Pace et al., 2017) draw attention to many areas of supportive care where limited or no evidence to guide practice exists. For example, the benefits of neurorehabilitation have limited and poor-quality evidence according to both of these guidance documents. And the EANO guidance has emphasised the low-quality evidence for guiding practice with undertaking needs assessment and the even lower levels supporting caregiver needs. In the course of this research the paucity of evidence for complex aspects of care was noted. The traditional hierarchy of evidence used in these guidelines that rank systematic reviews using meta-analysis and RCTs as the best sources of evidence, does not recognise the complexity of HNA programmes or similar areas of practice. Therefore, this hierarchy should be re-evaluated (Greenhalgh et al., 2016). Research that is focused on neurorehabilitation with methodology that recognises the complexity should be a priority.

This research raises questions similar to other reviews of HNA, such as; What are the outcome measures that should be considered in terms of HNA (Ahmed et al., 2014) and what is the clinical utility of using an HNA? Research is required to evaluate if a brain tumour HNA programme with assessment followed by actions results in benefits for patients and carers. This is an extremely complex area to examine but looking at one aspect in isolation is unlikely to have a beneficial impact within the context of brain tumours. If future research uses the programme theories presented here, these could be further refined and developed to facilitate better outcomes.

A notable area not examined in depth but also requiring investigation – particularly in this population – is the use of electronic HNA and care planning and communication tools. It could provide increased accessibility for some patients with communication limitations and, dependent on the software, provide information about how to meet needs for both the healthcare professionals and patients supported by the most up to date evidence.

5.9 Strengths

This thesis is the first study that has explored how HNA could be used in brain tumour patients and strategies are urgently needed to help improve the experience of this patient group and their significant others. This view is underpinned by large scale studies which have established that this group has the poorest care experiences of any cancer group in England. This is supported by the gold standard measure of the Cancer Patients Experience Survey (Brainstrust, 2019).

This thesis as a whole, and through the three individual studies, demonstrates a number of strengths. The systematic review used the COSMIN criteria to consider the psychometric properties. These criteria were relatively new at the time of this review and using it can help to ensure the reliability of systematic reviews focused on measurement tools compared to other criteria (Scholtes et al., 2011). This thesis also collated criteria from a number of different reviews focused on HNA to come up with a considered appraisal process for the quality of HNA tools which could be replicated by other studies. This has been published in a good quality cancer journal and has undergone peer review to disseminate the process used in this systematic review, so others could replicate this if required.

The second study used a pragmatic research approach. Early on in this study it was apparent that many patients with cognitive impairments (often those with what appeared to be more significant impairment) either would not be able to make a set time for a focus group due to dependency on others, or they did not feel comfortable in doing so. Also, although this was initially focused on patients, carers were also invited to attend for support and to provide more information if needed. The use of focus groups (with carers and patients), carer and patient interviews and individual interviews provided multiple insights that would not have been revealed through only one of the methodologies. The initial plan of focus groups allowed individuals to reflect on others' comments and aspects such as shared challenges of diagnosis or debate about how HNA tools could be used to provide additional insights. The inclusion of carers changed the outcomes and findings of the study as the integral needs of inclusion and care for carers would not have been explored without their views. Individual interviews allowed in some cases for additional disclosure, such as the impact of marriage breakdown that might not have been as easily revealed in a group setting. This triangulation of methods has helped to increase confidence in the findings presented.

The final research phase involved a realist review and this methodology permitted the exploration of a very complex intervention programme of HNA with subsequent care planning and support. This methodology is suitable when the intervention is complex and findings have been inconsistent. In this area of study, one of the main strengths recognised was the importance of 'context matters'. These aspects, such as an individual's self-efficacy, carer support or the characteristics of the practitioner, could be considered in proposing theories of what might work for whom in what circumstances. The theories provided through this are specific enough to help guide an implementation of pilot studies in clinical practice and the provision of a practice-based model will allow policy makers to consider this in the planned implementation of an HNA programme.

This research had a number of strengths that have helped to provide a basis for taking forward HNA in brain tumours in practice. Research has demonstrated that in knowledge transfer activities findings which have a sensitivity and recognition of the user's context are important to support implementation of research (Boyko et al., 2018). The specific focus on brain tumours and the focus of the research in the UK will be beneficial in supporting the findings of this thesis. In addition, the provision of action-oriented findings which recommend how and what could be done are more likely to be considered by the practitioners.

5.10 Limitations

Brain tumour-specific literature was limited both in terms of tools and the evaluation of programmes or interventions relevant to HNA. There were a limited number of tools that could be considered for an HNA in brain tumour patients. A limited number of researchers (Hickmann et al., 2016; Renovanz et al., 2016) used generic HNA's to examine needs in brain tumour patients, however one Renovanz et al., (2016) highlighted the lack of brain tumour specific needs as a limitation of their study. This choice made prior to the publication of these papers, remains justified due to the need to comprehensively assess the specific impacts of a brain tumour.

Through an investigation of approaches in regions where HNA is widely implemented, the decision was made to use validated brain tumour symptom assessment tools. This was done for cancer HNA and distress screening as a validated symptom assessment with additional questions is the approach used in Canada (Bultz et al., 2011). While this inherently limited their use as an HNA, and this was reflected in the systematic review, it did provide additional options for consideration.

In Study 2, there were revisions to the protocol due to recruitment challenges. The initial design only had focus groups (not interviews) and there was a refusal rate of approximately 95% due to severity of symptoms and poor prognosis, travel difficulties or other reasons. The exclusion of those with more severe symptoms or poor prognosis was mitigated to some extent through changing to an interview option (acceptance rate of 71%) but it is likely that there was a proportion of patients who did not feel able to participate. Therefore, this remains a purposive sample. The inclusion of carers as a pragmatic choice, in retrospect was beneficial, but this could have been considered in more depth as this provided a unique opportunity to potentially explore the patient and carer experiences of the same events (Morgan et al., 2013). However, it also needs to be recognised that individual views could have been suppressed to present the shared view when these dyads invariably had close relationships (Norlyk et al., 2016). Agreements such as ‘we cope within the family with challenges like this’ would be hard for an individual to refute and may have impacted the discussions.

Realist methods, as with many research methodologies, could benefit from researchers working together and they are challenging for a novice researcher. With an iterative approach to sampling the literature there are aspects which require additional decision making as the rules are not as fixed as in other methods, such as the Cochrane review. To mitigate this, an inclusion and exclusion criteria, data extraction forms and systematic approach in considering the contribution of literature was devised. However, in theory-driven approaches used in realist reviews it is not uncommon to be overwhelmed by the complexity of revealing all potential causal mechanisms (Jagosh, 2019). In the event, a prioritisation of the candidate theories, with a focus on self-management, was selected to take forward as a PhD project. However, in subsequent research, it would be interesting to consider other theories focused on other outcomes that were not the main focus and consider their relevance to HNA in brain tumour patients.

In undertaking study focused in an area of complexity (HNA programmes in brain tumour patients), there are significant challenges in transferring the findings into practice. Knowledge transfer of research findings, specifically those from realist reviews need to be presented to knowledge users in a way they find useable and beneficial (Boyko et al., 2018). The chains of inference presented in Chapter 4 that support each theory are numerous, so it was important to synthesise these into action-based recommendations useful for practice. To some extent, the practice-based model was able to use the theories to present a suggested framework for practice to take forward HNA in brain tumours. This is important as when seeking to maximise knowledge transfer, knowledge users in practice have been shown to favour actionable findings (Boyko et al., 2018). However, in some areas of the practice-based model, the lack of specific recommendations, such as capacity for shared decision making, could limit the translation of this research into practice.

6 Chapter 6 - Conclusion

This thesis focuses on assessing and addressing unmet needs in people affected by brain tumours. The aim of this study is to explore how HNA assessments followed by appropriate support and care could improve the outcomes for brain tumour patients. While there has been a great deal of research into HNA in cancer, there has been a lack of research into this topic in PBTs. This thesis has helped to address this gap.

The majority of brain tumour patients experience neurocognitive impairments, and their impact has an influence on their needs. But it also has implications for how HNA should be undertaken, and support delivered. Although there are similarities with HNA research in other cancer sites, or cancer more generally, this research highlights the limitations without considering these impairments. The main findings have also provided the basis for recommendations how the existing UK model should be adapted to support this patient group.

Due to neuro-cognitive impairments, an assessment of the patient's ability to self-manage combined with inclusion and support of carers in HNA programmes for brain tumour patients is needed to maximise the potential for self-management. As patients diagnosed with a brain tumour are relatively rare and they may have significantly different needs than other cancers, primary care practitioners may lack the knowledge and skills to meet these needs. However, improving the communication and the flow of information between neuro-oncology specialists and primary care practitioners may help address post-treatment needs and improve wellbeing. In addition, easily accessible support and communication for patients and carers from the specialist neuro-oncology team could improve their ability to self-manage. Distress can be a barrier to self-management and with clear recognition that this is often higher in brain tumour patients and their carers than other cancer, the assessment and management of this should be an integral part of HNA programmes in brain tumours.

The paragraph above summarises the main findings, but to develop HNA for brain tumour patients, the programme theories provided a proposal of how HNA could work for brain tumour patients. The identification of specific contexts and mechanisms in delivery for each step of the HNA process will allow those developing services to structure models of care for brain tumour patients and their significant others. This thesis ultimately proposes a practice model which can provide the basis for further study or even implementation of an HNA package which takes these complexities into account. The research questions addressed in this thesis have provided a platform to help shape further research and clinical practice for this group.

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Appendices

Appendix 1 - Methodological appraisal

	Score (0-3)									
Criteria	Paper: (Janda et al., 2006b)	Paper: (Janda et al., 2008a)	Paper: (Armstrong et al., 2005)	Paper: (Armstrong et al., 2006)	Paper: (Armstrong et al., 2012b)	Paper: (Armstrong et al., 2014)	Paper: (Rooney et al., 2014)	Paper: (Weitzner et al., 1995)	Paper: (Cella et al., 2003)	Paper: (Lai, Jensen, et al., 2014)
Explicit theoretical framework	0	0	3	2	2	0	0	3	3	3
Statement of aims/objectives	3	3	2	3	3	3	3	3	3	3

in main body of report										
Clear description of research setting	3	3	2	2	3	3	3	3	3	3
Evidence of sample size considered in terms of analysis	3	1	3	2	2	2	0	2	2	1
Representative sample of target group of a reasonable size	2	2	2	2	2	2	2	3	3	3
Description of procedure for data collection	2	2	3	3	3	3	3	3	3	3

Rationale for choice of data collection tool(s)	1	2	3	2	3	3	3	3	2	3
Detailed recruitment data	3	3	2	3	3	3	3	3	3	3
Statistical assessment of reliability and validity of measurement tool(s) (Quantitative only)	NA	0	2	3	3	3	1	3	2	3
Fit between stated research	NA	1	3	3	3	3	3	3	3	3

question and method of data collection (Quantitative only)										
Fit between stated research question and format and content of data collection tool e.g. interview schedule (Qualitative only)	3	NA	2	NA	NA	NA	2	2	NA	2

Fit between research question and method of analysis (Quantitative only)	NA	2	2	3	3	3	2	2	3	3
Good justification for analytic method selected	2	1	3	3	3	3	2	2	3	3
Assessment of reliability of analytic process (Qualitative only)	2	NA	1	NA	NA	NA	0	2	NA	1

Evidence of user involvement in design	0	0	1	1	0	0	0	2	3	2
Strengths and limitations critically discussed	2	1	0	0	2	2	2	2	2	2
SCORE AND PERCENTAGE	26/39 66.7%	21/42 50%	34/48 70.8%	31/42 73.8%	35/42 83.3%	33/45 73.3%	31/48 64.6%	39/48 81%	38/42 90%	39/48 81%

Appendix 2 – Holistic Needs Assessment in Brain Tumour Patients: A Systematic Review of Available Tools – published paper

Afseth, J., Neubeck, L., Karatzias, T., & Grant, R. (2019). Holistic needs assessment in brain cancer patients: A systematic review of available tools. *European Journal of Cancer Care*, *28*(3). https://doi.org/10.1111/ecc.12931

Appendix 3 Patient information sheet and consent

PATIENT INFORMATION SHEET

Version Number: **5.0** **Date: 17 April 2015**

Study title: An exploration of the concerns of patients who have been diagnosed with a brain tumour and their carers when they are attending outpatient clinics.

You are being invited to take part in a research study. Before you decide it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. Ask us if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part.

Thank you for reading this.

What is the purpose of the study?

The purpose of this study is to help define what are the concerns of patients and their significant others or carers when they have been diagnosed with a brain tumour. We are interested in exploring what the concerns are when attending outpatient consultations with health care professionals (for example with your doctor at the hospital).

To find this out we will be undertaking focus groups and interviews. Focus groups are group discussions with approximately 5 to 10 people in total that have been diagnosed with a brain tumour and their carers or significant others. The patient can nominate one carer or significant other for the study and both will attend the same focus group (although patients may also attend without a carer or significant other). Interviews are discussions where the researcher will ask questions to a person who has been diagnosed with a brain tumour and if their carer or significant other (if the patient would like them to be there)

The focus groups will be held at suitable public location, which will be advised by the researcher. Interviews will be held a suitable location agreeable to yourself and the interviewer (this can be a public location or your home). Full details and directions of the exact location will be provided in a letter to you after you agree to take part in the study.

We hope that the information that is gained through these discussions will help in the design of a simple tool that will highlight the individual concerns of patients, which can be easily used in outpatient appointments.

This study will also form part of the PhD for the lead researcher, Janyne Afseth.

Why have I been chosen?
You have been chosen because you are currently seeing health care professionals and have been diagnosed with brain tumour.

Up to 30 patients and carers in up to 3 focus groups and up to 8 interviews are required for the study and all must comply with several study entry criteria. These include being willing to provide informed consent and attend focus groups or interviews.

Do I have to take part?
No, you do not have to take part. It is up to you to decide whether or not to take part. If you do decide to take part, you will be given this information sheet to keep and be asked to sign a consent form before the focus group or interview. You can choose if you would like to take part in a focus group or an interview (as long as we are still needing participants for both). If you decide to take part you are still free to withdraw at any time. You can withdraw without giving any reason and your medical care or legal rights will not be affected.

What will happen to me if I take part?
If you enter the study a few steps need to be taken before the start of the study.

A member of the research team will:

- Confirm your consent to take part by signing the consent form
- Confirm that you meet all the criteria for the study.
- Collect a small amount of relevant information about yourself such as your recent outpatient attendances
- If we do not already have the details of your hospital consultant and GP we will collect these details prior to the start of the focus group or interview.

After this you will be given some dates to participate at a focus group. There is some information about this focus group or interview we would like to give you:

- Refreshments will be available such as tea, coffee, water and snacks (if not at your home)

- Travel expenses will be reimbursed at the standard Edinburgh Napier University rates

- You are free not to discuss certain areas if you would prefer not to for any reason during the discussion
- The focus groups and interviews will be audio recorded and transcribed (the recordings typed up). In the transcription your name will be replaced by a pseudonym (made up name) and it should not be possible to identify you in the analysis.
- All information you give during the discussion may be used in analysis (unless you request for it not to be) but will be made anonymous.
- You are free to ask questions at any time before, during or after the focus groups or interviews.

What are the possible disadvantages and risks of taking part?
Because we will be discussing concerns about brain tumours, there is the possibility that some of the discussion during the focus groups or interviews may be distressing. However, you would be free to choose not to take part in that bit of the discussion, or to leave the focus group or stop the interview if you preferred.

What are the possible benefits of taking part?
You may not receive any direct benefit, as this will provide information for the development of a tool to benefit patients at a later time. However, you may find the opportunity to discuss your experiences and concerns of some benefit, as well as the knowledge you will be helping others.

What happens when the research study stops?
When the research stops the results will be used to inform the research team about concerns and aid them in designing a tool that best meets the needs of patients attending outpatient clinics

What happens to the information collected during the study?

The audio recording will be transcribed and then checked it is correctly transcribed. Analysis of this transcription will then take place and then recording will be destroyed. All the transcribed data will be anonymised, and your name will be replaced with a pseudonym, and it will not be possible for you to be identified in any reporting of the data gathered. All anonymised data collected will be kept in a secure place (locked cabinet in locked room/stored on a pc that is password protected on a university server) to which only the research team has access. The transcriptions and analysis from the study will be kept for 7 years from the date of focus group or interview.

Any information about you used for analysis or published will have your name and address removed so that you cannot be recognised from it. A copy of your consent form will be locked securely in a cupboard in the university and a copy will be placed in your hospital notes where you are or have been treated to inform your health care team that you participated in the study. Your GP will also be informed of your participation in this study.

Your hospital records will be checked to confirm your diagnosis and obtain the address of your GP. Your medical or research records may be inspected by the research department of the NHS Lothian or NHS Greater Glasgow to check that the study is being carried out correctly. Your name, however, will not be disclosed outside the hospital through this process.

What will happen to the results of the research study?
Your anonymised data will be analysed by the research team, which includes researchers from Edinburgh Napier University, Lothian Health Board and Brainstrust UK. The results of the study may be used in reports and scientific presentations or publications. You will not be identified in these publications.

A summary of the results of the study will be provided to the Brainstrust which will be available on the website www.brainstrust.org.uk or details can be posted to you (the research team will ask at the focus groups or interview if you would like this to happen).

Should you decide to withdraw from the study, the information collected on you up to that point will be used in the results of the study unless you request for it not to be.

Who is organising and funding the research?
The Lothian Health Foundations Grants has put forward funding for this study.

Who has reviewed the study?
The study protocol has been reviewed by the South East Scotland Research Ethics Committee 01, Edinburgh Napier University Faculty of Life, Health and Social Sciences Ethics committee and the patient support charity Brainstrust (brainstrust.org.uk)

Contact for further information

If you would like to discuss the study in more detail with the researcher please contact:

Janyne Afseth, Lecturer, School of Nursing,
Midwifery and Social Care, Edinburgh Napier
University, EH11 4BN
0131 455 5703
email: j.afseth@napier.ac.uk

or Research Supervisor: Professor Thanos Karatzias, School of Nursing,
Midwifery and Social Care, Edinburgh Napier
University, EH11 4BN
0131 455 5345
Email: t.karatzias@napier.ac.uk

If you would like to discuss this information with an independent advisor please contact:

Barbara Neades, Senior Lecturer, Edinburgh
Napier University, 0131-455-5315

If you have any questions about your rights as a research participant, questions about taking part in the study, or complaints about the study you may contact someone independent from the study at;

The NHS Lothian Complaints Team on 0131 465 5708 or at complaints.team@nhslothian.scot.nhs.uk

Or

The NHS Greater Glasgow Complaints Team on 0141 201 4500 or at complaints@ggc.scot.nhs.uk

If you decide to take part in this study, you will be given a copy of this information sheet and a signed consent form to keep.

Thank you for taking the time to read this information leaflet

PATIENT CONSENT FORM
(Final Version 5.0, 17 April 2015)

Centre Name:
Study Identification Number:

Study title: An exploration of the concerns of patients who have been diagnosed with a brain tumour and their carers when they are attending out patient clinics.

Name of Researcher: Janyne Afseth — Initial below

I confirm that I have read and understood the information sheet for the above study and have had the opportunity to ask questions.	☐
I understand that my participation is voluntary and that I am free to withdraw at any time. I understand that I can withdraw without giving any reason, without my medical care or legal rights being affected.	☐
I understand that sections of any of my medical or research notes may be looked at by responsible individuals from research departments of health boards where it is relevant to my taking part in research and the research team. I give permission for these individuals to have access to my records.	☐
I understand that the focus groups or interviews will be audio recorded and transcribed. All the transcribed data will be anonymised and all names will be replaced with a pseudonym.	☐
I understand that my hospital consultant will be notified of my participation. I give permission for my hospital consultant to be informed	☐
I understand that my GP will be notified of my participation. I give permission for my GP to be informed	☐
I agree to take part in the above study.	☐

Name of Participant	Date	Signature
Name of Person Taking Consent	Date	Signature
Witness if required	Date	Signature

3 copies, 1 for patient, 1 for researcher and 1 to kept with hospital notes

Appendix 4 – Carer information sheet and consent

CARER INFORMATION SHEET

Version Number: **5.0** **Date: 17 April 2015**

Study title: An exploration of the concerns of patients who have been diagnosed with a brain tumour and their carers when they are attending out patient clinics.

You are being invited to take part in a research study. Before you decide it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. Ask us if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part.

Thank you for reading this.

What is the purpose of the study?
The purpose of this study is to help define what are the concerns of patients and their significant others or carers when they have been diagnosed with a type of brain tumour. We are interested in exploring what the concerns are when attending outpatient consultations with health care professionals (for example with your doctor at the hospital).

To find this out we will be undertaking focus groups and interviews. Focus groups are group discussions with approximately 5 to 10 people in total who have been diagnosed with a brain tumour and their carer or significant other. The patient can nominate one carer or significant other for the study and both will attend the same focus group (although patients may also attend without a carer or significant other). Interviews are discussions where the researcher will ask questions to a person who has been diagnosed with a brain tumour and if their carer or significant other (if the patient would like them to be there)

The focus groups will be held at suitable public location, which will be advised by the researcher. Interviews will be held a suitable location agreeable to yourself and the interviewer (this can be a public location or your home). Full details and directions of the exact location will be provided in a letter to you after you agree to take part in the study.

We hope that the information that is gained through these discussions will help in the design of a simple tool that will highlight the individual concerns of patients, which can be easily used in outpatient appointments.

This study will also form part of the PhD for the lead researcher, Janyne Afseth.

Why have I been chosen?
You have been chosen because your significant other or someone you care for is currently seeing health care professionals and has been diagnosed with brain tumour. They have nominated you as their significant other or carer.

Up to 30 patients and carers in up to 3 focus groups and up to 8 interviews are required for the study and all must comply with several study entry criteria. These include being willing to provide informed consent and attend focus groups or interview.

Do I have to take part?
No, you do not have to take part. It is up to you to decide whether or not to take part. If you do decide to take part you will be given this information sheet to keep and be asked to sign a consent form before the focus group or interview. You can choose (with the patient you are attending with) if you would like to take part in a focus group or an interview (as long as we are still needing participants for both). If you decide to take part you are still free to withdraw at any time. You can withdraw without giving any reason and your medical care or legal rights will not be affected.

What will happen to me if I take part?
If you enter the study a few steps need to be taken before the start of the study.

A member of the research team will:

- Confirm your consent to take part by signing the consent form
- Confirm that you meet all the criteria for the study.
- Collect a small amount of relevant information about yourself such as the outpatient appointments you have attended with your significant other or the person you care for

After this you will be given some dates to participate at a focus group. There is some information about this focus group or interview we would like to give you:

- Refreshments will be available such as tea, coffee, water and snacks (if not at your home).
- Travel expenses will be reimbursed at the standard Edinburgh Napier University rates
- You are free not to discuss certain areas if you would prefer not to for any reason during the discussion
- The focus groups and interviews will be audio recorded and transcribed (the recordings typed up). In the transcription your name will be replaced by a pseudonym (made up name) and it should not be possible to identify you in the analysis.

○

- All information you give during the discussion may be used in analysis (unless you request for it not to be) but will be made anonymous.
- You are free to ask questions at any time before, during or after the focus groups or interviews.

What are the possible disadvantages and risks of taking part?
Because we will be discussing concerns about brain tumours, there is the possibility that some of the discussion during the focus groups or interviews may be distressing. However, you would be free to choose not to take part in that bit of the discussion, or to leave the focus group or stop the interview if you preferred.

What are the possible benefits of taking part?
You may not receive any direct benefit, as this will provide information for the development of a tool to benefit patients at a later time. However, you may find the opportunity to discuss your experiences and concerns of some benefit, as well as the knowledge you will be helping others.

What happens when the research study stops?
When the research stops the results will be used to inform the research team about concerns and aid them in designing a tool that best meets the needs of patients attending outpatient clinics

What happens to the information collected during the study?

The audio recording will be transcribed and then checked it is correctly transcribed. Analysis of this transcription will then take place and then recording will be destroyed. All the transcribed data will be anonymised and your name will be replaced with a pseudonym, and it will not be possible for you to be identified in any reporting of the data gathered. All anonymised data collected will be kept in a secure place (locked cabinet in locked room/stored on a pc that is password protected on a university server) to which only the research team has access.
The transcriptions and analysis from the study will be kept for 7 years from the date of focus group or interview.

Any information about you used for analysis or published will have your name and address removed so that you cannot be recognised from it. A copy of your consent form will be locked securely in a cupboard in the university.

Your research records may be inspected by the research department of NHS Lothian or NHS Greater Glasgow to check that the study is being carried out correctly. Your name, however, will not be disclosed outside the hospital through this process.

What will happen to the results of the research study?
Your anonymised data will be analysed by the research team, which includes researchers from Edinburgh Napier University, Lothian Health Board and Brainstrust UK. The results of the study may be used in reports and scientific presentations or publications. You will not be identified in these publications.

A summary of the results of the study will be provided to the BrainsTrust which will be available on the website www.brainstrust.org.uk or details can be posted to you (the research team will ask at the focus groups or interviews if you would like this to happen).

Should you decide to withdraw from the study at any time, the information collected on you up to that point will be used in the results of the study unless you request for it not to be.

Who is organising and funding the research?
The Lothian Health Foundations Grants has put forward funding for this study.

Who has reviewed the study?
The study protocol has been reviewed by the South East Scotland Research Ethics Committee 01, Edinburgh Napier University Faculty of Life, Health and Social Sciences Ethics committee and the patient support charity Brainstrust (brainstrust.org.uk)

Contact for further information

If you would like to discuss the study in more detail with the researcher please contact:

Janyne Afseth, Lecturer, School of Nursing,
Midwifery and Social Care, Edinburgh Napier
University, EH11 4BN
0131 455 5703

Email: j.afseth@napier.ac.uk

or Research Supervisor: Professor Thanos Karatzias, School of Nursing,
Midwifery and Social Care, Edinburgh Napier
University, EH11 4BN phone: 0131 455 5345

Email: t.karatzias@napier.ac.uk

If you would like to discuss this information with an independent advisor please contact:

Barbara Neades, Senior Lecturer, Edinburgh
Napier University, 0131 455 5315

If you have any questions about your rights as a research participant, questions about taking part in the study, or complaints about the study, you may contact someone independent from the study at:

The NHS Lothian Complaints Team on 0131 465 5708 or at complaints.team@nhslothian.scot.nhs.uk

Or

The NHS Greater Glasgow Complaints Team on 0141 201 4500 or at complaints@ggc.scot.nhs.uk

If you decide to take part in this study you will be given a copy of this information sheet and a signed consent form to keep.

Thank you for taking the time to read this information leaflet

CARER CONSENT FORM
(Final Version 5.0, 17 April 2015)

Centre Name:
Study Identification Number:

Study title: An exploration of the concerns of patients who have been diagnosed with a brain tumour and their carers when they are attending out patient clinics.

Name of Researcher: Janyne Afseth

	Initial below
I confirm that I have read and understood the information sheet for the above study and have had the opportunity to ask questions.	☐
I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason, without my medical care or legal rights being affected	☐
I understand that the focus groups or interviews will be audio recorded and transcribed. All the transcribed data will be anonymised and all names will be replaced with a pseudonym.	☐
I understand that sections of any of my research notes may be looked at by responsible individuals from research departments of health boards where it is relevant to my taking part in research and the research team. I give permission for these individuals to have access to my records.	☐
I agree to take part in the above study.	☐

Name of Participant	Date	Signature
Name of Person Taking Consent	Date	Signature
Witness if required	Date	Signature

2 copies, 1 for carer and 1 for researcher

Appendix 5 – Interview schedule and topic guide

Interview Schedule

General information - to be undertaken at the start of the interviews

1. Welcome and introduction
2. Review of the purpose of the interview
3. Review of the participant rights (i.e can withdraw consent, stop interview or chose not to answer any question)
4. Remind of recording and data protection
5. Check for any outstanding questions

2.0 Interview Schedule

[The exact words maybe slightly different or the format may be modified dependent on the discussion from the participants, for example if a topic is already covered. The points in italics are prompts to be used if required]

The topic we are looking at is concerns that brain tumour patients may have during their outpatient appointments.

To start with I wondered if you could tell me what the word ‘concern’ means to you? How would you describe this term? I will give 30 to 60 seconds to allow you to have a think and gather your thoughts before answering.

> *Are there any other words that you might use instead of concern?*
>
> *How does this compare with the word ‘need’? or problem?*
>
> *What word to you think would be the best word to be used for highlighting issues?*

Do you feel that you; or patients and their carers in general, are usually able to discuss concerns during outpatient appointments?

> *As a patient or carer do you feel you have an opportunity to discuss all the concerns you have?*
>
> *Is there usually enough time?*

What areas are the health care staff not as good at discussing with you? Or *Are there some areas that are not discussed? (prompt for each domain if not brought up: physical, psychological, social, emotional, financial, sexual, functional and spiritual domains)*

Are there areas that you would feel you want help with but find difficult discussing?

Do the health care staff seem to understand what your concerns are when you tell them?

Researchers have previously designed four tools to help identify symptoms, problems or concerns for patients with Brain Cancer (Facilitator will give copies of the *Brain Patient Concern Inventory, MD Anderson Symptom Inventory with Brain Module, Comprehensive Cancer Network/Functional Assessment of Cancer Therapy-Brain Symptom Index (NFBrSI-24) and the Supportive Care Needs Survey with Brain Subscale* at this point). Take a moment to look at these tools, which give some examples of asking about needs or concerns before appointments

Do you think these would be useful to help identify concerns before an appointment? Why or why not?

(*support discussion to go through each and say they think is good and what they do not think is good or the limitations of each tool*)

There are some aspects of need that some researchers have suggested are important to examine. For example:

- Do you think it is important to rank (*say which concern is most important*)?
- or put the severity of concerns?
- Do you think it is important in the questionnaire to say if you want help or not with this need?
- Do you think it is important that you are able to say how much of a burden this is – or how much it interrupts your life?

A few general questions about design.

What do you think of the length of each of these?

Can you see any difficulties in completing any of these questionnaires?

When and where do you think would be the best time to fill this in?

For example in the waiting room?

Or sent out to you before your appointment?

Can you think of any (other) benefits of using a tool like this before consultations with the specialist nurse or doctor?

Can you think of any disadvantages of using a tool like this before consultations with the specialist nurse or doctor?

Are there certain appointments you think this tool might not be good to use?

For example, when a patient is first diagnosed? Or when starting a new treatment such as radiotherapy

Is there anyone anything would like to add about identifying and dealing with problems in appointments?

Does anyone have additional feedback about any of these tools?

Many thanks for your participation and support of this research

Topic Guide

General information - to be undertaken at the start of the focus group

1. Welcome and introduction
2. Review of the purpose of the interview
3. Review of the participant rights (i.e. can withdraw consent, stop interview or chose not to answer any question)
4. Remind of recording and data protection
5. Check for any outstanding questions

2.0 Focus group guide

Note - This is a topic guide and the exact words maybe slightly different or the format may be modified dependent on the discussion from the participants, for example if a topic is already covered. The points in italics are prompts to be used if required

The topic we are looking at is concerns that brain tumour patients may have during their outpatient appointments.

To start with I wondered if you could tell me what the word ‘concern’ means to you? How would you describe this term? I will give 30 to 60 seconds to allow you to have a think and gather your thoughts before answering.

Are there any other words that you might use instead of concern?

How does this compare with the word ‘need’? or problem?

What word to you think would be the best word to be used for highlighting issues?

Do you feel that you; or patients and their carers in general, are usually able to discuss concerns during outpatient appointments?

As a patient or carer do you feel you have an opportunity to discuss all the concerns you have?

Is there usually enough time?

What areas are the health care staff not as good at discussing with you? *Or Are there some areas that are not discussed? (prompt for each domain if not brought up: physical, psychological, social, emotional, financial, sexual, functional and spiritual domains)*

Are there areas that you would feel you want help with but find difficult discussing?

Do the health care staff seem to understand what your concerns are when you tell them?

Researchers have previously designed three tools to help identify problems or concerns for patients with Brain Cancer (Facilitator will give copies of Patient Concern Inventory, MD Anderson Symptom Inventory with Brain Module Comprehensive Cancer Network/Functional Assessment of Cancer Therapy-Brain Symptom Index (NFBrSI-24) and the Supportive Care Needs Survey with Brain Subscale at this point). Take a moment to look at these tools, which give some examples of asking about needs or concerns before appointments

- Do you think these would be useful to help identify concerns before an appointment? Why or why not?
- (support discussion to go through each and say they think is good and what they do not think is good or the limitations of each tool)
- There are some aspects of need that some researchers have suggested are important to examine. For example:
- Do you think it is important to rank (say which concern is most important)?
- or put the severity of concerns?
- Do you think it is important in the questionnaire to say if you want help or not with this need?
- Do you think it is important that you are able to say how much of a burden this is – or how much it interrupts your life?

A few general questions about design.

- What do you think of the length of each of these?
- Can you see any difficulties in completing any of these questionnaires?

When and where do you think would be the best time to fill this in?

- *For example in the waiting room?*
- *Or sent out to you before your appointment?*

Can you think of any (other) benefits of using a tool like this before consultations with the specialist nurse or doctor?

Can you think of any disadvantages of using a tool like this before consultations with the specialist nurse or doctor?

Are there certain appointments you think this tool might not be good to use?

For example when a patient is first diagnosed? Or when starting a new treatment such as radiotherapy

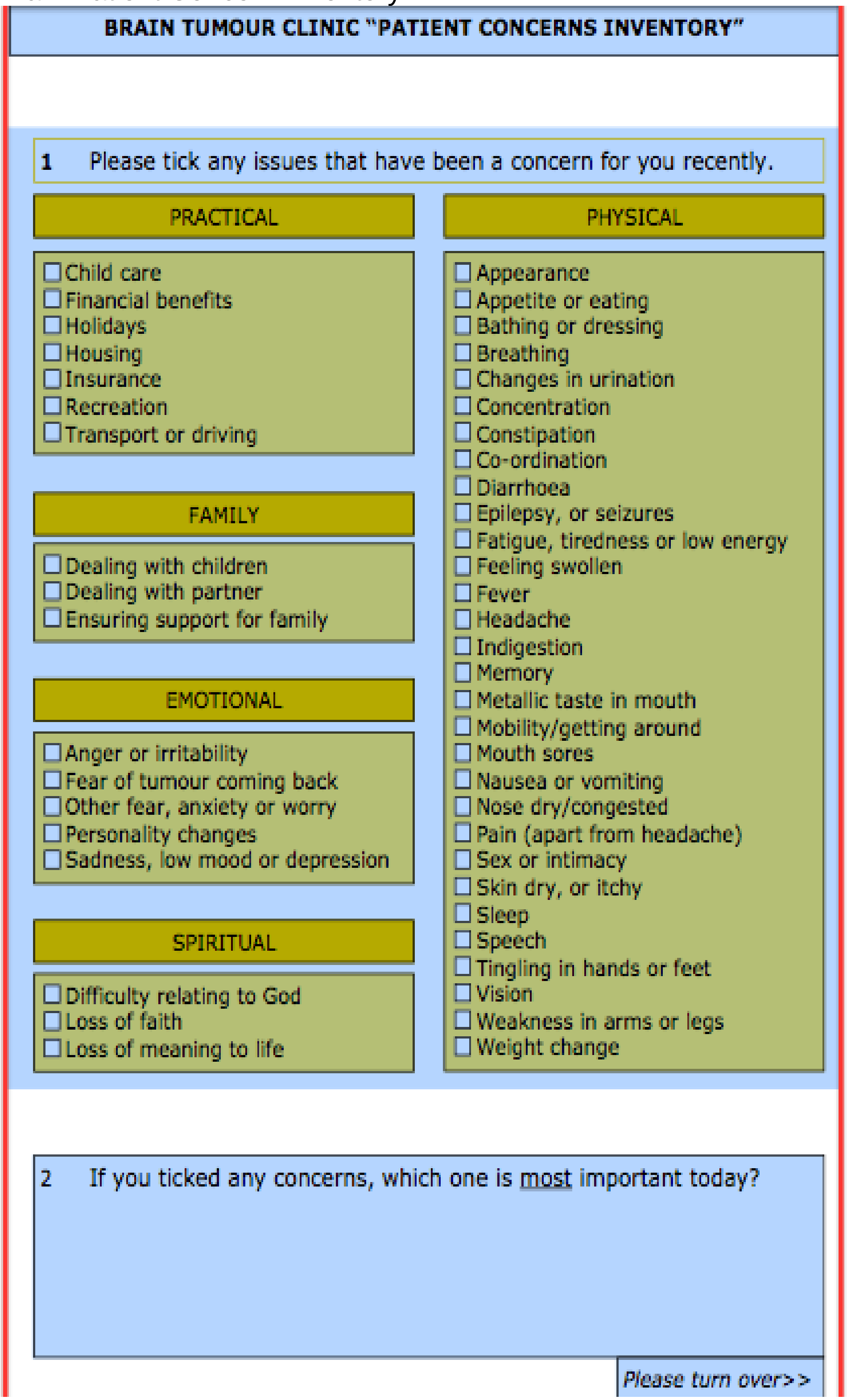
Is there anyone anything would like to add about identifying and dealing with problems in appointments?

Does anyone have additional feedback about any of these tools?

Many thanks for your participation and support of this focus group.

Appendix 6 - Copy of Patient HNA and symptom assessment tools for discussion at focus groups and interviews

Brain Patient Concern Inventory



BRAIN TUMOUR CLINIC "PATIENT CONCERNS INVENTORY"

1 Please tick any issues that have been a concern for you recently.

PRACTICAL

☐ Child care
☐ Financial benefits
☐ Holidays
☐ Housing
☐ Insurance
☐ Recreation
☐ Transport or driving

FAMILY

☐ Dealing with children
☐ Dealing with partner
☐ Ensuring support for family

EMOTIONAL

☐ Anger or irritability
☐ Fear of tumour coming back
☐ Other fear, anxiety or worry
☐ Personality changes
☐ Sadness, low mood or depression

SPIRITUAL

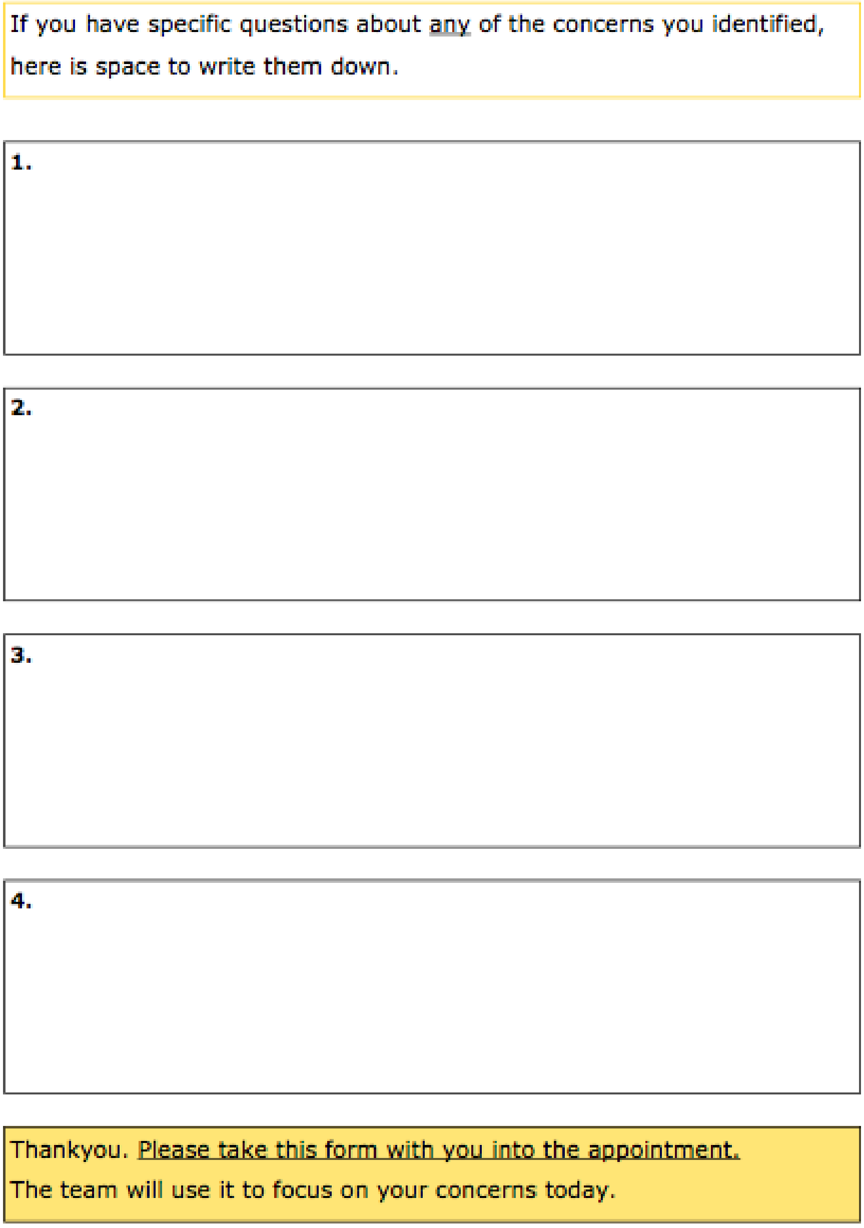
☐ Difficulty relating to God
☐ Loss of faith
☐ Loss of meaning to life

PHYSICAL

☐ Appearance
☐ Appetite or eating
☐ Bathing or dressing
☐ Breathing
☐ Changes in urination
☐ Concentration
☐ Constipation
☐ Co-ordination
☐ Diarrhoea
☐ Epilepsy, or seizures
☐ Fatigue, tiredness or low energy
☐ Feeling swollen
☐ Fever
☐ Headache
☐ Indigestion
☐ Memory
☐ Metallic taste in mouth
☐ Mobility/getting around
☐ Mouth sores
☐ Nausea or vomiting
☐ Nose dry/congested
☐ Pain (apart from headache)
☐ Sex or intimacy
☐ Skin dry, or itchy
☐ Sleep
☐ Speech
☐ Tingling in hands or feet
☐ Vision
☐ Weakness in arms or legs
☐ Weight change

2 If you ticked any concerns, which one is most important today?

Please turn over>>



If you have specific questions about any of the concerns you identified, here is space to write them down.

1.

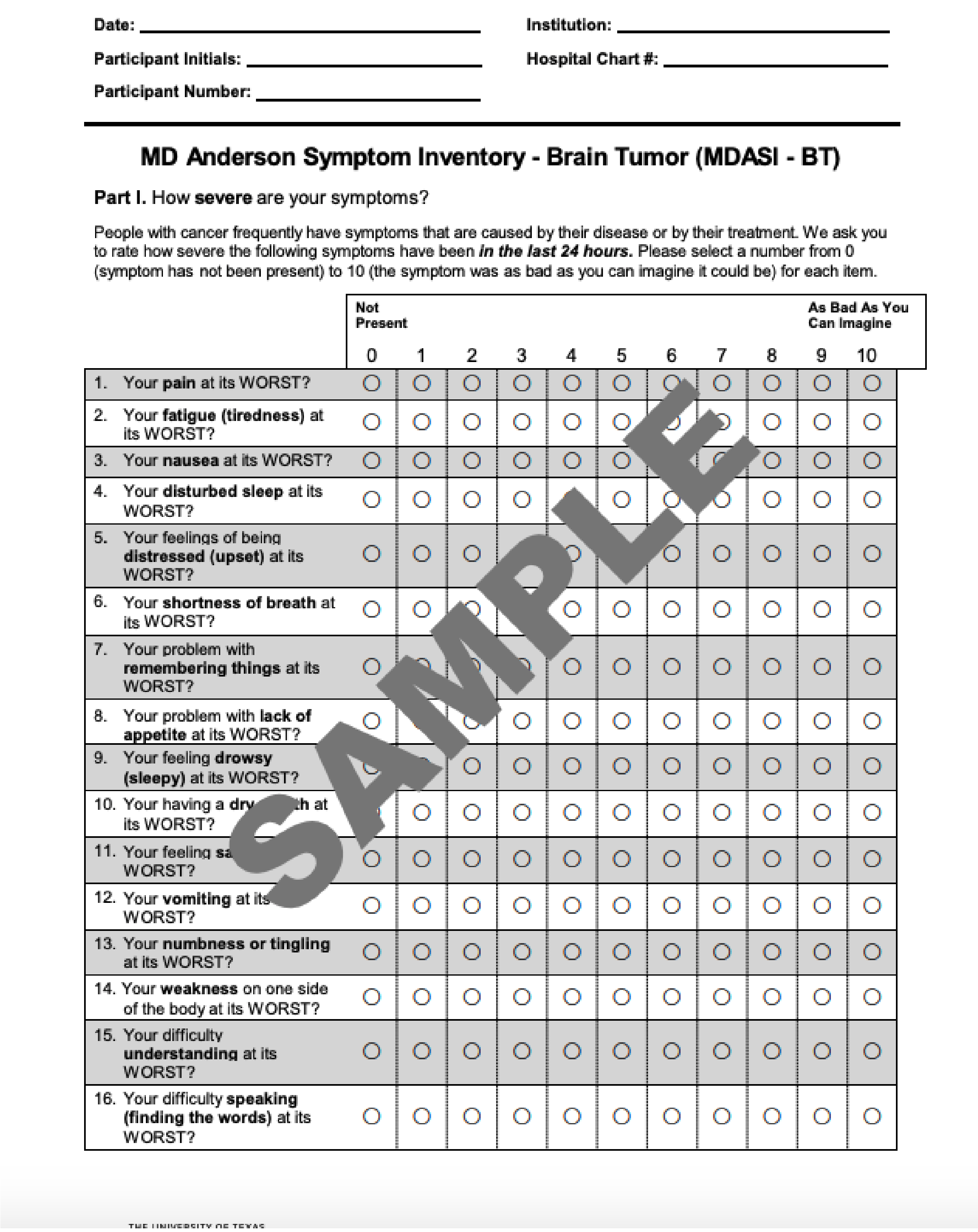
2.

3.

4.

Thankyou. Please take this form with you into the appointment.
The team will use it to focus on your concerns today.

MD Anderson Symptom Inventory with Brain Tumor Module



Date: ______________________ **Institution:** ______________________

Participant Initials: ______________________ **Hospital Chart #:** ______________________

Participant Number: ______________________

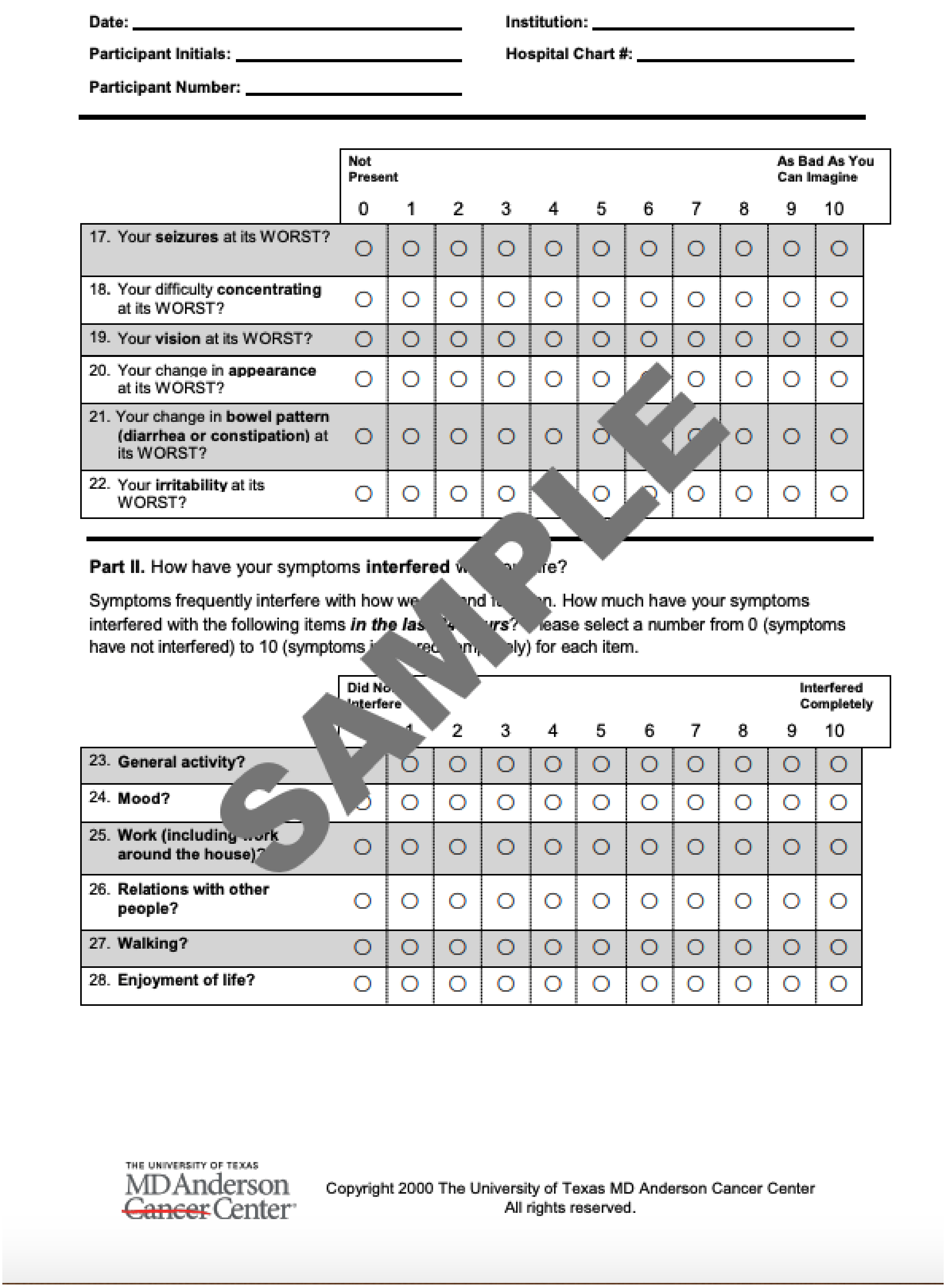
MD Anderson Symptom Inventory - Brain Tumor (MDASI - BT)

Part I. How **severe** are your symptoms?

People with cancer frequently have symptoms that are caused by their disease or by their treatment. We ask you to rate how severe the following symptoms have been ***in the last 24 hours.*** Please select a number from 0 (symptom has not been present) to 10 (the symptom was as bad as you can imagine it could be) for each item.

	Not Present 0	1	2	3	4	5	6	7	8	**As Bad As You Can Imagine** 9	10
1. Your **pain** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
2. Your **fatigue (tiredness)** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
3. Your **nausea** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
4. Your **disturbed sleep** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
5. Your feelings of being **distressed (upset)** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
6. Your **shortness of breath** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
7. Your problem with **remembering things** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
8. Your problem with **lack of appetite** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
9. Your feeling **drowsy (sleepy)** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
10. Your having a **dry**[illegible]**th** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
11. Your feeling **sa**[illegible] WORST?	○	○	○	○	○	○	○	○	○	○	○
12. Your **vomiting** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
13. Your **numbness or tingling** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
14. Your **weakness** on one side of the body at its WORST?	○	○	○	○	○	○	○	○	○	○	○
15. Your difficulty **understanding** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
16. Your difficulty **speaking (finding the words)** at its WORST?	○	○	○	○	○	○	○	○	○	○	○

THE UNIVERSITY OF TEXAS



Date: ____________________

Institution: ____________________

Participant Initials: ____________________

Hospital Chart #: ____________________

Participant Number: ____________________

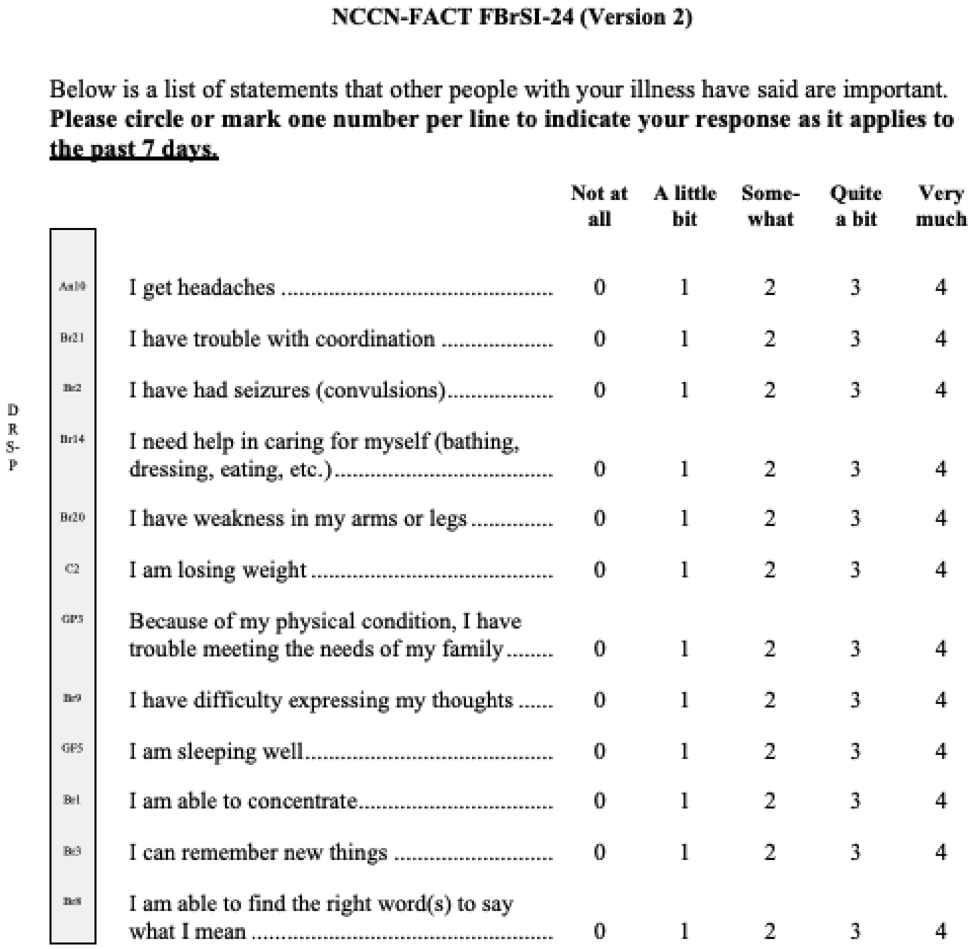
	Not Present									**As Bad As You Can Imagine**	
	0	1	2	3	4	5	6	7	8	9	10
17. Your **seizures** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
18. Your difficulty **concentrating** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
19. Your **vision** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
20. Your change in **appearance** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
21. Your change in **bowel pattern (diarrhea or constipation)** at its WORST?	○	○	○	○	○	○	○	○	○	○	○
22. Your **irritability** at its WORST?	○	○	○	○	○	○	○	○	○	○	○

Part II. How have your symptoms **interfered** with your life?

Symptoms frequently interfere with how we feel and function. How much have your symptoms interfered with the following items ***in the last 24 hours***? Please select a number from 0 (symptoms have not interfered) to 10 (symptoms interfered completely) for each item.

	Did Not Interfere									**Interfered Completely**	
	0	1	2	3	4	5	6	7	8	9	10
23. **General activity?**	○	○	○	○	○	○	○	○	○	○	○
24. **Mood?**	○	○	○	○	○	○	○	○	○	○	○
25. **Work (including work around the house)?**	○	○	○	○	○	○	○	○	○	○	○
26. **Relations with other people?**	○	○	○	○	○	○	○	○	○	○	○
27. **Walking?**	○	○	○	○	○	○	○	○	○	○	○
28. **Enjoyment of life?**	○	○	○	○	○	○	○	○	○	○	○

Comprehensive Cancer Network/Functional Assessment of Cancer Therapy-Brain Symptom Index (NFBrSI-24)

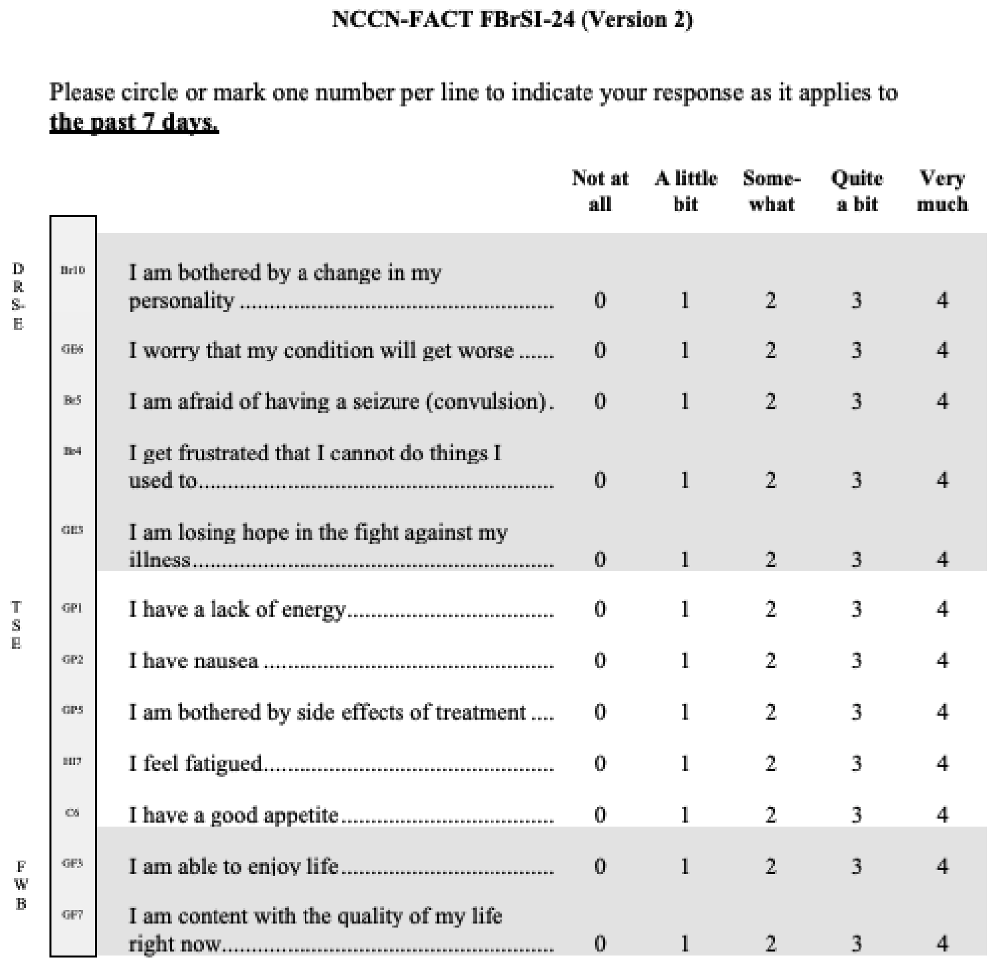


NCCN-FACT FBrSI-24 (Version 2)

Below is a list of statements that other people with your illness have said are important. **Please circle or mark one number per line to indicate your response as it applies to <u>the past 7 days.</u>**

DRS-P

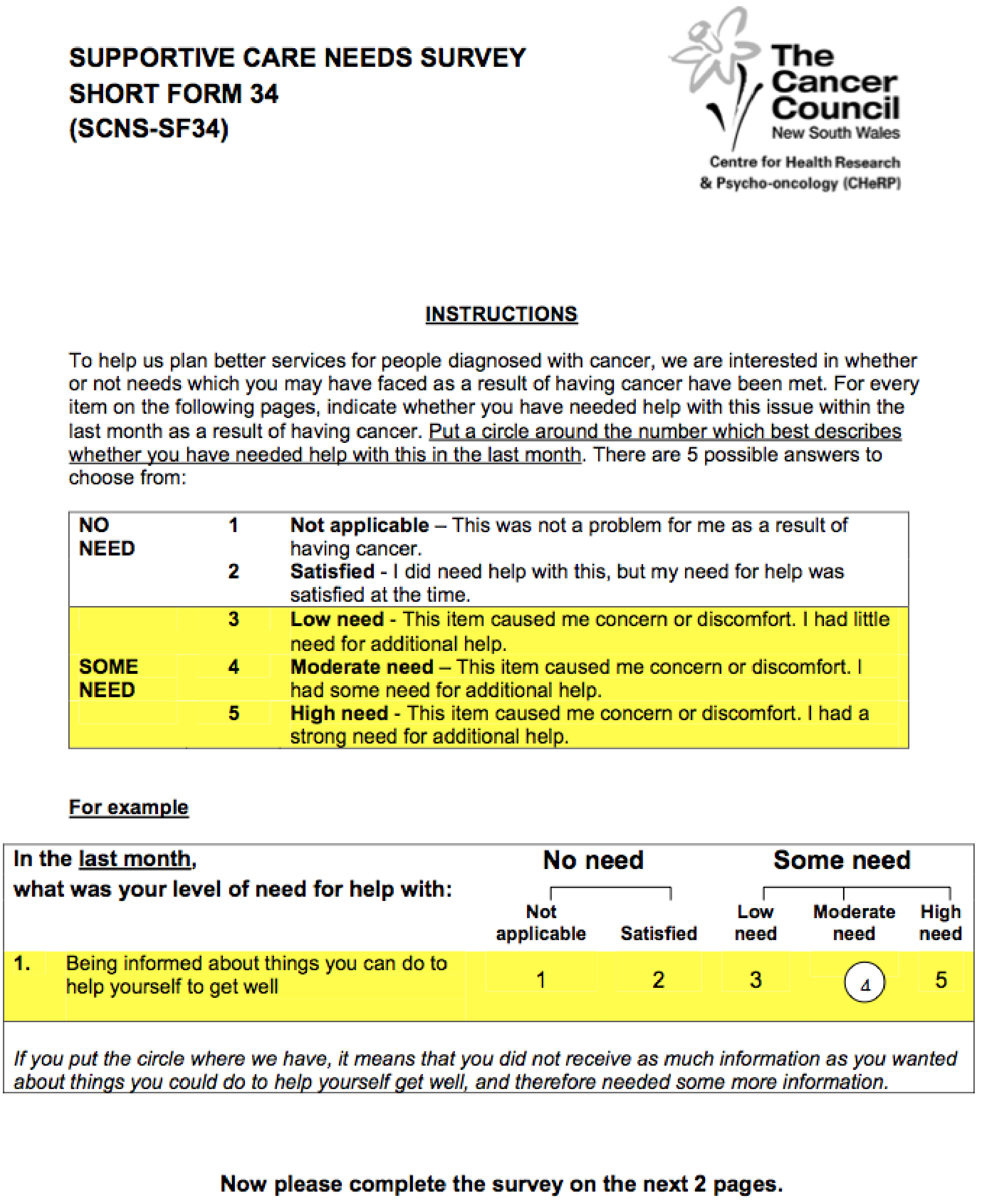
		Not at all	A little bit	Some-what	Quite a bit	Very much
An10	I get headaches	0	1	2	3	4
Br21	I have trouble with coordination	0	1	2	3	4
Br2	I have had seizures (convulsions)..................	0	1	2	3	4
Br14	I need help in caring for myself (bathing, dressing, eating, etc.)......................................	0	1	2	3	4
Br20	I have weakness in my arms or legs	0	1	2	3	4
C2	I am losing weight	0	1	2	3	4
GP3	Because of my physical condition, I have trouble meeting the needs of my family........	0	1	2	3	4
Br9	I have difficulty expressing my thoughts	0	1	2	3	4
GP5	I am sleeping well..	0	1	2	3	4
Br1	I am able to concentrate..................................	0	1	2	3	4
Br3	I can remember new things	0	1	2	3	4
Br8	I am able to find the right word(s) to say what I mean	0	1	2	3	4



NCCN-FACT FBrSI-24 (Version 2)

Please circle or mark one number per line to indicate your response as it applies to **the past 7 days.**

			Not at all	A little bit	Some-what	Quite a bit	Very much
DRS-E	Br10	I am bothered by a change in my personality	0	1	2	3	4
	GE6	I worry that my condition will get worse	0	1	2	3	4
	Br5	I am afraid of having a seizure (convulsion).	0	1	2	3	4
	Br4	I get frustrated that I cannot do things I used to...	0	1	2	3	4
	GE3	I am losing hope in the fight against my illness...	0	1	2	3	4
TSE	GP1	I have a lack of energy....................................	0	1	2	3	4
	GP2	I have nausea	0	1	2	3	4
	GP5	I am bothered by side effects of treatment	0	1	2	3	4
	HI7	I feel fatigued..	0	1	2	3	4
	C6	I have a good appetite....................................	0	1	2	3	4
FWB	GF3	I am able to enjoy life....................................	0	1	2	3	4
	GF7	I am content with the quality of my life right now...	0	1	2	3	4



SUPPORTIVE CARE NEEDS SURVEY
SHORT FORM 34
(SCNS-SF34)

The Cancer Council
New South Wales
Centre for Health Research
& Psycho-oncology (CHeRP)

INSTRUCTIONS

To help us plan better services for people diagnosed with cancer, we are interested in whether or not needs which you may have faced as a result of having cancer have been met. For every item on the following pages, indicate whether you have needed help with this issue within the last month as a result of having cancer. Put a circle around the number which best describes whether you have needed help with this in the last month. There are 5 possible answers to choose from:

NO NEED	1	**Not applicable** – This was not a problem for me as a result of having cancer.
	2	**Satisfied** - I did need help with this, but my need for help was satisfied at the time.
SOME NEED	3	**Low need** - This item caused me concern or discomfort. I had little need for additional help.
	4	**Moderate need** – This item caused me concern or discomfort. I had some need for additional help.
	5	**High need** - This item caused me concern or discomfort. I had a strong need for additional help.

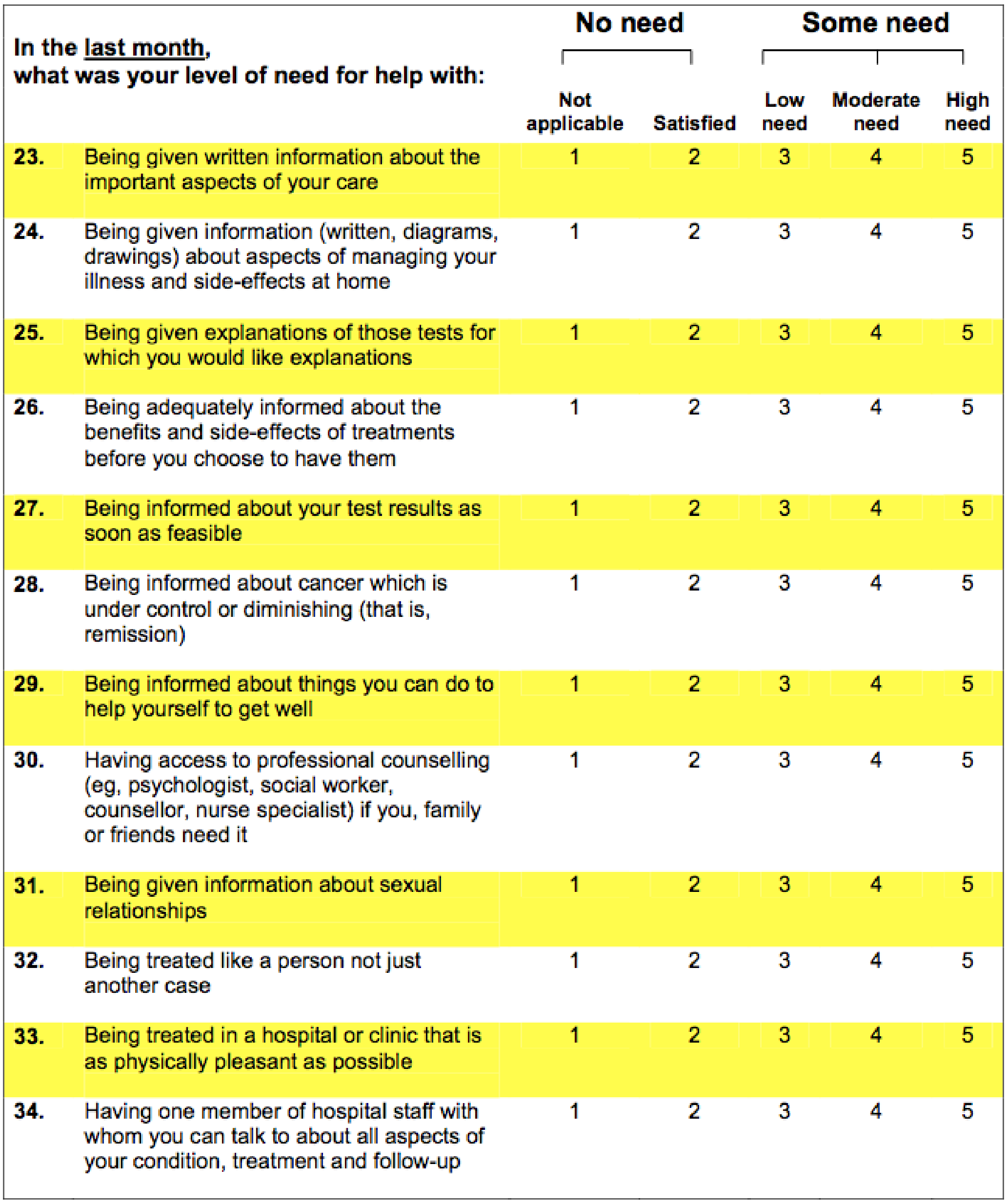
For example

In the last month, what was your level of need for help with:	**No need**		**Some need**		
	Not applicable	**Satisfied**	**Low need**	**Moderate need**	**High need**
1. Being informed about things you can do to help yourself to get well	1	2	3	[✓ 4]	5

If you put the circle where we have, it means that you did not receive as much information as you wanted about things you could do to help yourself get well, and therefore needed some more information.

Now please complete the survey on the next 2 pages.

In the last month, what was your level of need for help with:		No need		Some need		
		Not applicable	Satisfied	Low need	Moderate need	High need
1.	Pain	1	2	3	4	5
2.	Lack of energy/tiredness	1	2	3	4	5
3.	Feeling unwell a lot of the time	1	2	3	4	5
4.	Work around the home	1	2	3	4	5
5.	Not being able to do the things you used to do	1	2	3	4	5
6.	Anxiety	1	2	3	4	5
7.	Feeling down or depressed	1	2	3	4	5
8.	Feelings of sadness	1	2	3	4	5
9.	Fears about the cancer spreading	1	2	3	4	5
10.	Worry that the results of treatment are beyond your control	1	2	3	4	5
11.	Uncertainty about the future	1	2	3	4	5
12.	Learning to feel in control of your situation	1	2	3	4	5
13.	Keeping a positive outlook	1	2	3	4	5
14.	Feelings about death and dying	1	2	3	4	5
15.	Changes in sexual feelings	1	2	3	4	5
16.	Changes in your sexual relationships	1	2	3	4	5
17.	Concerns about the worries of those close to you	1	2	3	4	5
18.	More choice about which cancer specialists you see	1	2	3	4	5
19.	More choice about which hospital you attend	1	2	3	4	5
20.	Reassurance by medical staff that the way you feel is normal	1	2	3	4	5
21.	Hospital staff attending promptly to your physical needs	1	2	3	4	5
22.	Hospital staff acknowledging, and showing sensitivity to, your feelings and emotional needs	1	2	3	4	5



In the last month, what was your level of need for help with:		No need		Some need		
		Not applicable	Satisfied	Low need	Moderate need	High need
23.	Being given written information about the important aspects of your care	1	2	3	4	5
24.	Being given information (written, diagrams, drawings) about aspects of managing your illness and side-effects at home	1	2	3	4	5
25.	Being given explanations of those tests for which you would like explanations	1	2	3	4	5
26.	Being adequately informed about the benefits and side-effects of treatments before you choose to have them	1	2	3	4	5
27.	Being informed about your test results as soon as feasible	1	2	3	4	5
28.	Being informed about cancer which is under control or diminishing (that is, remission)	1	2	3	4	5
29.	Being informed about things you can do to help yourself to get well	1	2	3	4	5
30.	Having access to professional counselling (eg, psychologist, social worker, counsellor, nurse specialist) if you, family or friends need it	1	2	3	4	5
31.	Being given information about sexual relationships	1	2	3	4	5
32.	Being treated like a person not just another case	1	2	3	4	5
33.	Being treated in a hospital or clinic that is as physically pleasant as possible	1	2	3	4	5
34.	Having one member of hospital staff with whom you can talk to about all aspects of your condition, treatment and follow-up	1	2	3	4	5

Thank you for completing this survey

Appendix 7 Copy of GP Letter to inform of patient participation

Date

Dr ……
Address

Dear Dr.

Re: [add name and date of birth]

Study title: An exploration of the concerns of patients who have been diagnosed with a brain tumour and their carers when they are attending out patient clinics.

Your patient has recently agreed to participate in the above study. The study will be exploring concerns of patients who have been diagnosed with brain tumour and their carers when attending outpatient consultations. This study will involve participation in one focus group or an interview.
Full details of the study are outlined in the enclosed patient information sheet dated 17 April 2015.

If you have any concerns about your patient's participation in the trial or require any further information about the study please do not hesitate to contact me. My contact details are:
Janyne Afseth, Lecturer
Edinburgh Napier University
0131-455-5703
j.afseth@napier.ac.uk

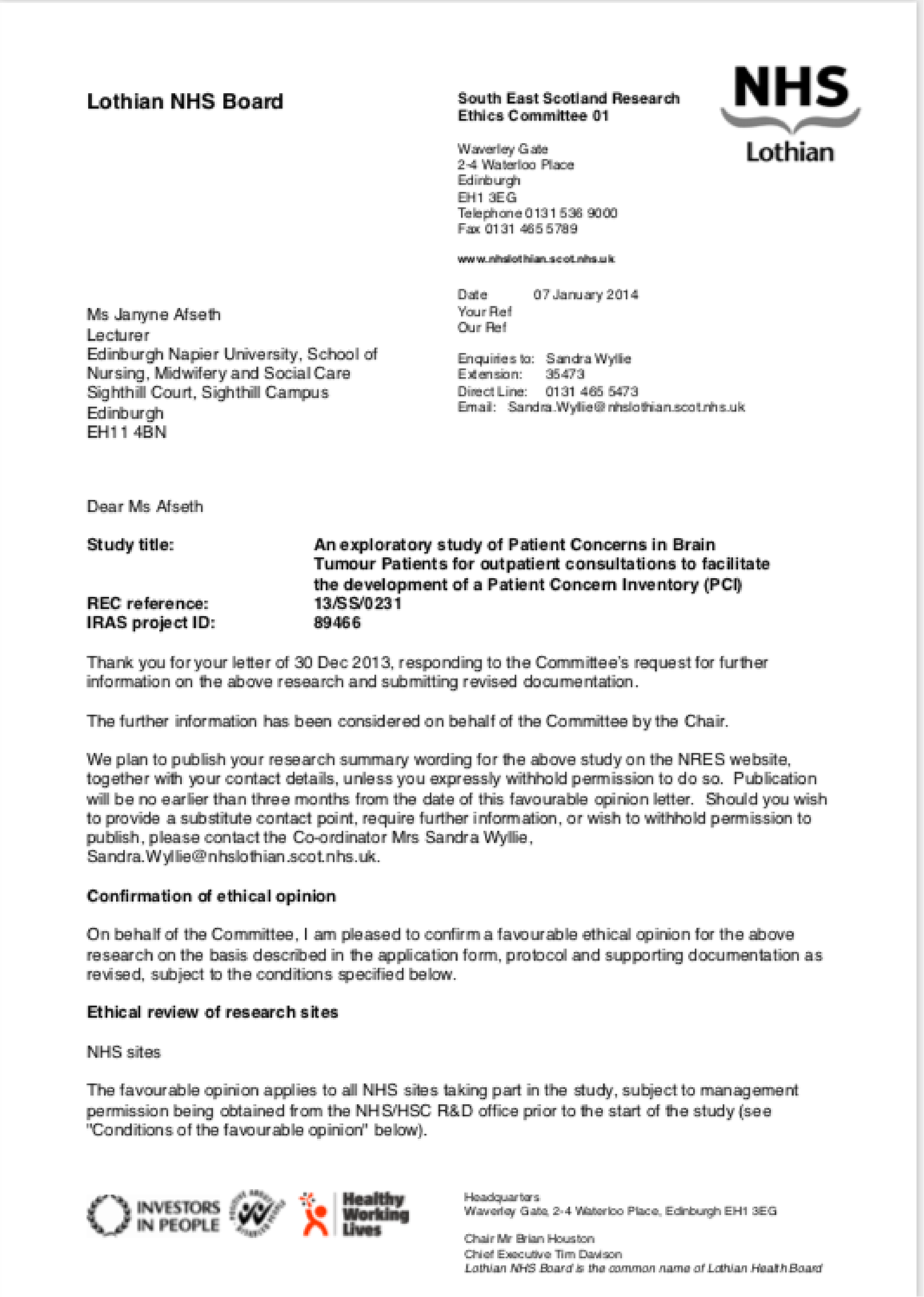
Yours sincerely

Janyne Afseth
Lecturer, Edinburgh Napier University

Enc: Patient information sheet

(on headed paper)

Appendix 8 – NHS Ethical approval letters



Lothian NHS Board

South East Scotland Research Ethics Committee 01

Waverley Gate
2-4 Waterloo Place
Edinburgh
EH1 3EG
Telephone 0131 536 9000
Fax 0131 465 5789

www.nhslothian.scot.nhs.uk

Date 07 January 2014
Your Ref
Our Ref

Enquiries to: Sandra Wyllie
Extension: 35473
Direct Line: 0131 465 5473
Email: Sandra.Wyllie@nhslothian.scot.nhs.uk

Ms Janyne Afseth
Lecturer
Edinburgh Napier University, School of Nursing, Midwifery and Social Care
Sighthill Court, Sighthill Campus
Edinburgh
EH11 4BN

Dear Ms Afseth

Study title: **An exploratory study of Patient Concerns in Brain Tumour Patients for outpatient consultations to facilitate the development of a Patient Concern Inventory (PCI)**
REC reference: **13/SS/0231**
IRAS project ID: **89466**

Thank you for your letter of 30 Dec 2013, responding to the Committee's request for further information on the above research and submitting revised documentation.

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

We plan to publish your research summary wording for the above study on the NRES website, together with your contact details, unless you expressly withhold permission to do so. Publication will be no earlier than three months from the date of this favourable opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to withhold permission to publish, please contact the Co-ordinator Mrs Sandra Wyllie, Sandra.Wyllie@nhslothian.scot.nhs.uk.

Confirmation of ethical opinion

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

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Headquarters
Waverley Gate, 2-4 Waterloo Place, Edinburgh EH1 3EG

Chair Mr Brian Houston
Chief Executive Tim Davison
Lothian NHS Board is the common name of Lothian Health Board

Conditions of the favourable opinion

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

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

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

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

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non clinical trials this is not currently mandatory.

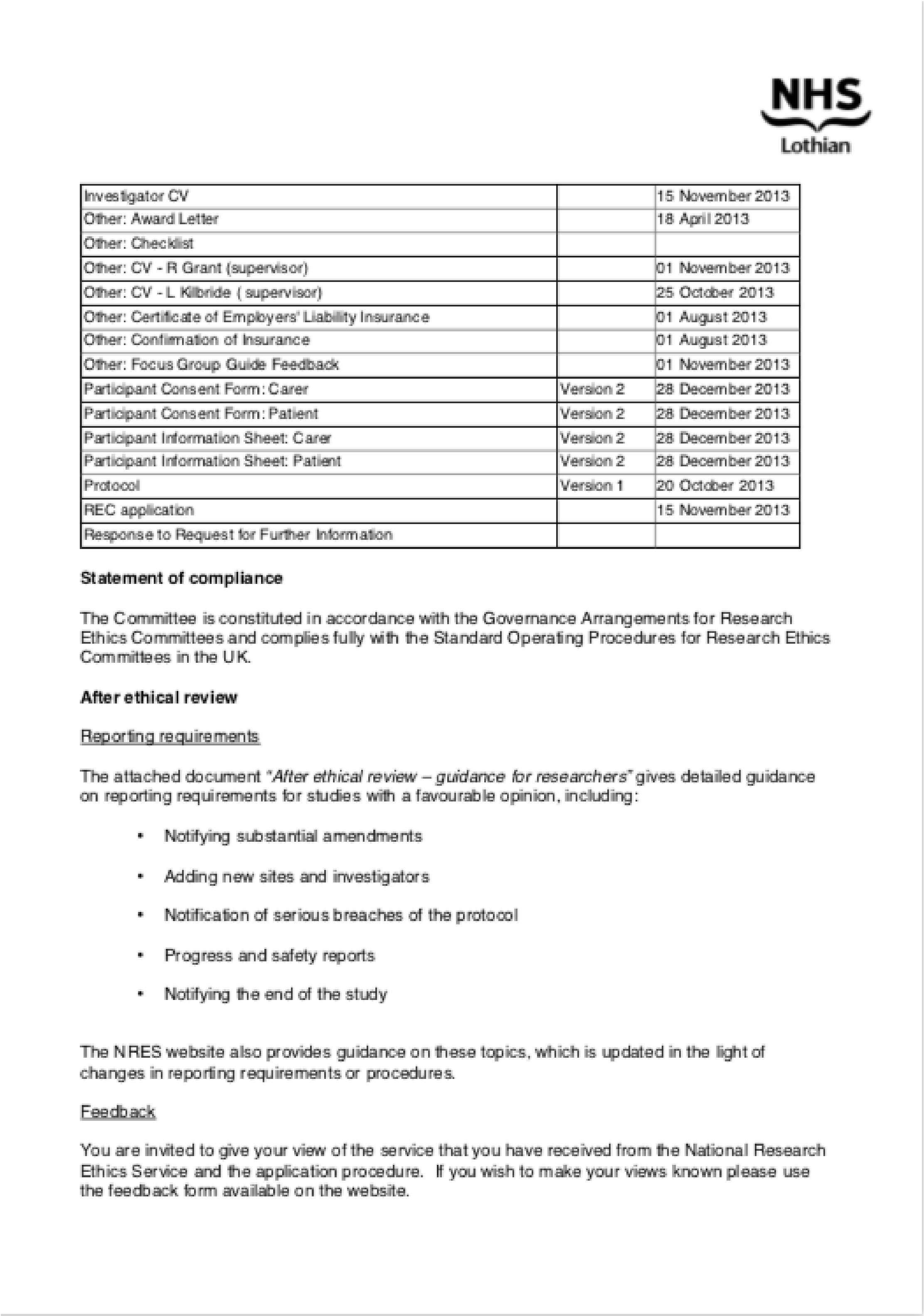
If a sponsor wishes to contest the need for registration they should contact Catherine Blewett (catherineblewett@nhs.net), the HRA does not, however, expect exceptions to be made. Guidance on where to register is provided within IRAS.

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

Approved documents

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

Document	*Version*	*Date*
GP/Consultant Information Sheets	Version 1.02	28 December 2013
Interview Schedules/Topic Guides	Version 1	01 November 2013



Investigator CV		15 November 2013
Other: Award Letter		18 April 2013
Other: Checklist		
Other: CV - R Grant (supervisor)		01 November 2013
Other: CV - L Kilbride (supervisor)		25 October 2013
Other: Certificate of Employers' Liability Insurance		01 August 2013
Other: Confirmation of Insurance		01 August 2013
Other: Focus Group Guide Feedback		01 November 2013
Participant Consent Form: Carer	Version 2	28 December 2013
Participant Consent Form: Patient	Version 2	28 December 2013
Participant Information Sheet: Carer	Version 2	28 December 2013
Participant Information Sheet: Patient	Version 2	28 December 2013
Protocol	Version 1	20 October 2013
REC application		15 November 2013
Response to Request for Further Information		

Statement of compliance

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

After ethical review

Reporting requirements

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

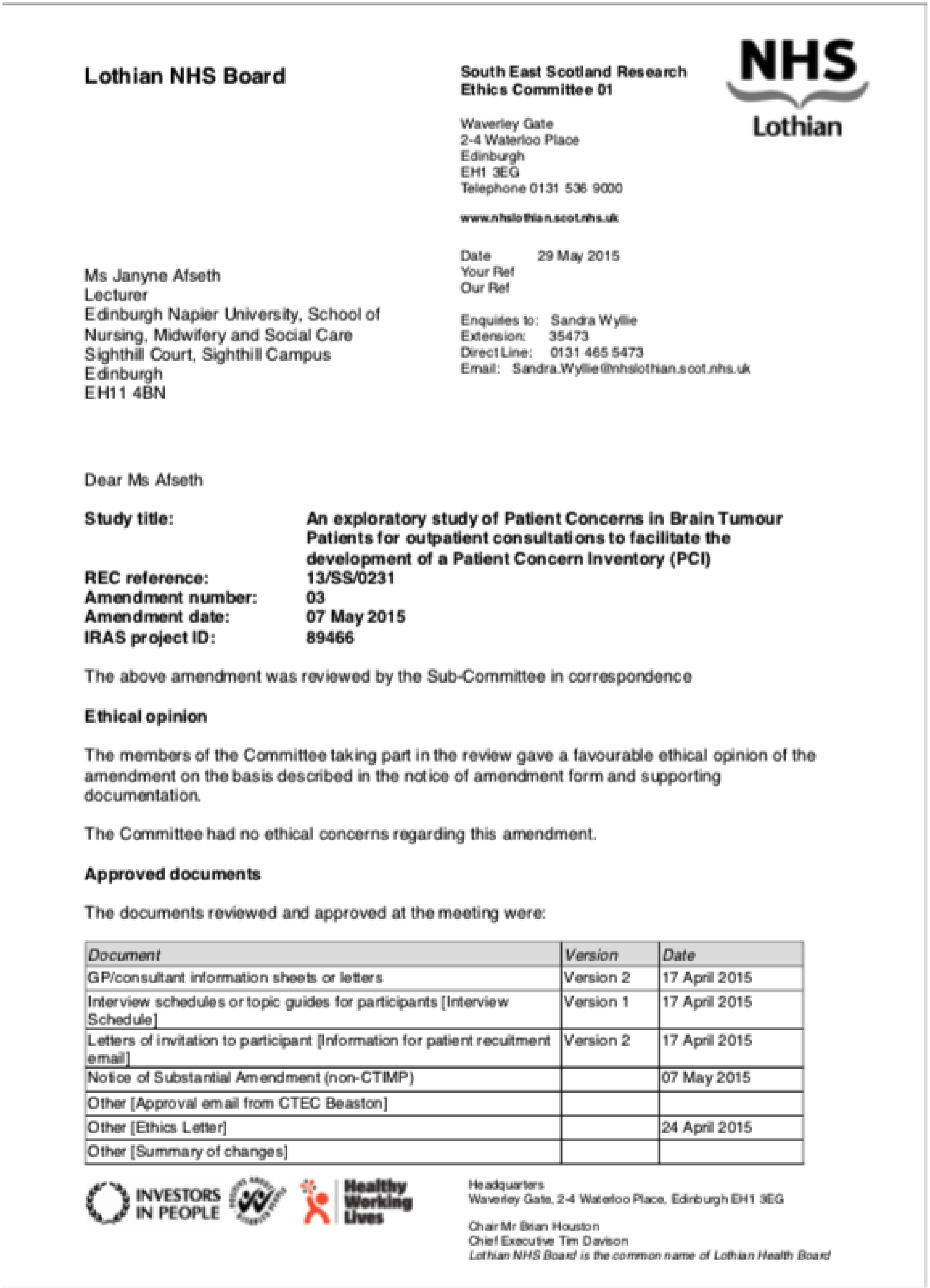
- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

Feedback

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

NHS approval – Amendment



Lothian NHS Board

South East Scotland Research
Ethics Committee 01

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Telephone 0131 536 9000

www.nhslothian.scot.nhs.uk

Date 29 May 2015
Your Ref
Our Ref

Enquiries to: Sandra Wyllie
Extension: 35473
Direct Line: 0131 465 5473
Email: Sandra.Wyllie@nhslothian.scot.nhs.uk

Ms Janyne Afseth
Lecturer
Edinburgh Napier University, School of
Nursing, Midwifery and Social Care
Sighthill Court, Sighthill Campus
Edinburgh
EH11 4BN

Dear Ms Afseth

Study title:	**An exploratory study of Patient Concerns in Brain Tumour Patients for outpatient consultations to facilitate the development of a Patient Concern Inventory (PCI)**
REC reference:	**13/SS/0231**
Amendment number:	**03**
Amendment date:	**07 May 2015**
IRAS project ID:	**89466**

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

Ethical opinion

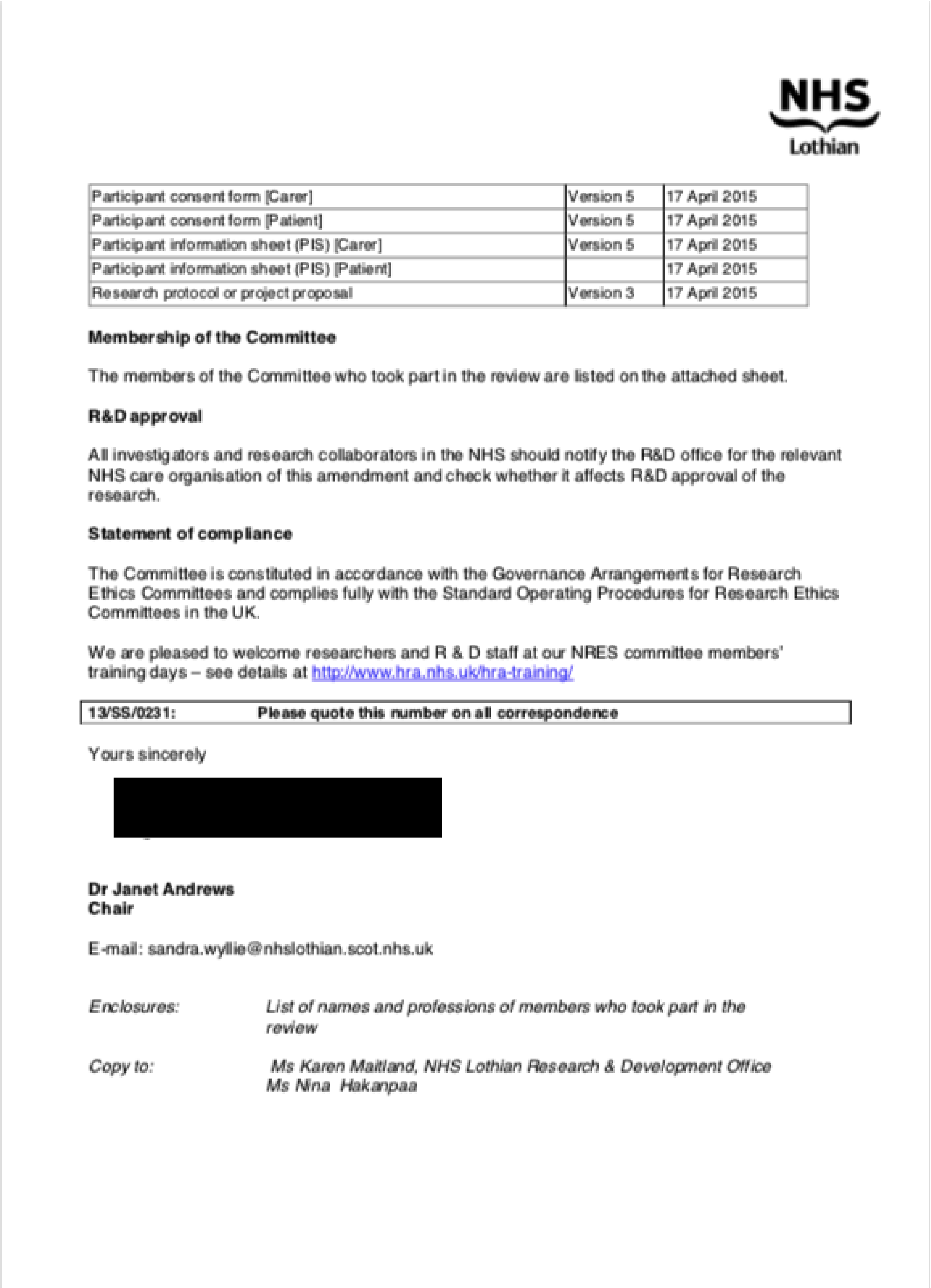
The members of the Committee taking part in the review gave a favourable ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

The Committee had no ethical concerns regarding this amendment.

Approved documents

The documents reviewed and approved at the meeting were:

Document	*Version*	*Date*
GP/consultant information sheets or letters	Version 2	17 April 2015
Interview schedules or topic guides for participants [Interview Schedule]	Version 1	17 April 2015
Letters of invitation to participant [Information for patient recuitment email]	Version 2	17 April 2015
Notice of Substantial Amendment (non-CTIMP)		07 May 2015
Other [Approval email from CTEC Beaston]		
Other [Ethics Letter]		24 April 2015
Other [Summary of changes]		



Participant consent form [Carer]	Version 5	17 April 2015
Participant consent form [Patient]	Version 5	17 April 2015
Participant information sheet (PIS) [Carer]	Version 5	17 April 2015
Participant information sheet (PIS) [Patient]		17 April 2015
Research protocol or project proposal	Version 3	17 April 2015

Membership of the Committee

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

R&D approval

All investigators and research collaborators in the NHS should notify the R&D office for the relevant NHS care organisation of this amendment and check whether it affects R&D approval of the research.

Statement of compliance

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

We are pleased to welcome researchers and R & D staff at our NRES committee members' training days – see details at http://www.hra.nhs.uk/hra-training/

13/SS/0231: Please quote this number on all correspondence

Yours sincerely

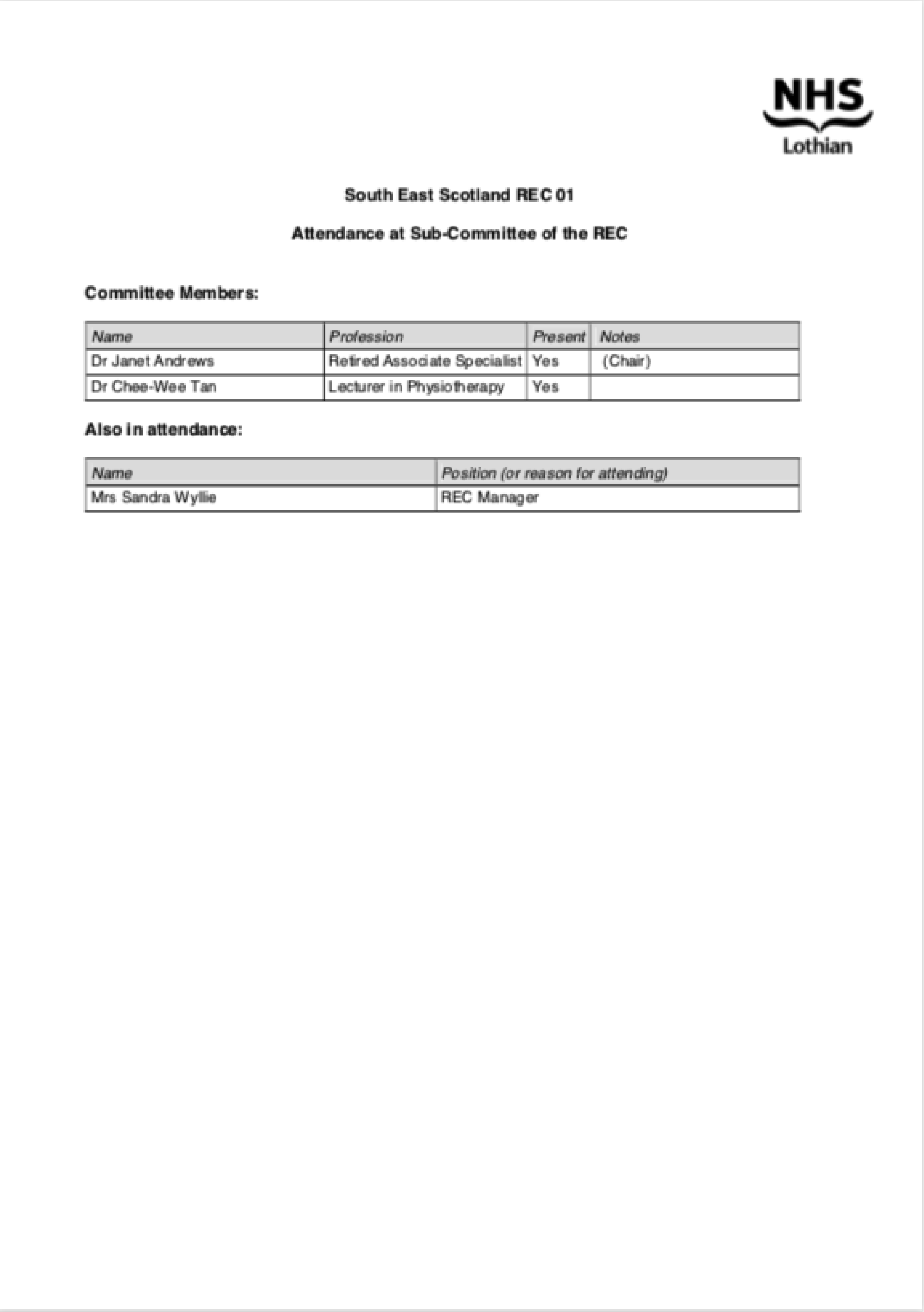
[redacted]

Dr Janet Andrews
Chair

E-mail: sandra.wyllie@nhslothian.scot.nhs.uk

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

Copy to: *Ms Karen Maitland, NHS Lothian Research & Development Office*
Ms Nina Hakanpaa



NHS
Lothian

South East Scotland REC 01

Attendance at Sub-Committee of the REC

Committee Members:

Name	*Profession*	*Present*	*Notes*
Dr Janet Andrews	Retired Associate Specialist	Yes	(Chair)
Dr Chee-Wee Tan	Lecturer in Physiotherapy	Yes	

Also in attendance:

Name	*Position (or reason for attending)*
Mrs Sandra Wyllie	REC Manager

Appendix 9 – Mind Map of Altered Self

Appendix 10 – Coding example

Interview 9 (patient)		
Transcript	Coding	Possible themes/Subthemes
R: … with the other fear, an anxiety, would be my dizzy spells (3), when I'm out and about (1). Personality changes, I feel I'm okay, but [my wife] thinks that I'm not the same person (2, 4), after my operation. But, but for me, personally, I feel okay. I've still, I've still got my, sort of, disabilities with my speech (3), but I must admit, I feel okay that way. And I, I'm always, I never have sadness. My outlook, now, is that I've had that operation, I could have been worse. And every day, ken, I look at things differently, now (5). And that way, I feel happy.	1. Neurocognitive change - causes anxiety or fear 2. Impact of neurocognitive changes on family 3. Specific neuro-cognitive symptoms 4. Different person 5. Adjustment – looking for positives	a) Neurocognitive changes and impact (1, 2, 3) b) Loss of self (2, 3) c) Coping and adjustment (5)

Appendix 11 - National policies and guidelines – informing initial theory generation

	United Kingdom – England	United Kingdom – Scotland	United States	Australia	Canada
Main purpose of Holistic Needs Assessment	Addressing holistic needs after completion of treatment to provide information for the individual and primary care and maximise wellbeing (through education, support and self-care)	Undertaking HNA throughout cancer 'journey' to facilitate support, advice/information and referral maximise wellbeing (through education, support and self-care) – part of mechanism to transform care after treatment	To determine what problems (or unmet needs) are contributing to distress to alleviate these to decrease distress	To assess holistic care needs (includes physical, psychological, social cultural, information and spiritual needs) of people affected by cancer. Based on this assessment, refer appropriately to promote optimal health and quality of life.	To determine what problems (or unmet needs) are contributing to distress to alleviate these to decrease distress To achieve a vision of person-centred care and ensure that each patient's concerns are being addressed by the right professional in a timely manner,

Focused time points for intervention	Completion of primary treatment	Suggest more than one timepoint and considered throughout each stage	Advised to be completed at each appointment (or set pathways) at all stages of cancer journey	Focused on cancer recovery but advised to be done throughout cancer continuum from diagnosis onwards	Advised to be completed at each appointment (or set pathways) at all stages of cancer journey
Description of associated interventions (part of HNA package)	HNA completed by health care professional. Macmillan problems checklist (specialist centre) Treatment summary (specialist centre). Completed by healthcare professional to be sent to primary care Cancer care review (primary care) to be completed by primary	Recommends elements of the recovery package as needed HNA completed by HCP. Macmillan problems checklist (specialist centre) Treatment summary (specialist centre). Completed by healthcare professional to be	Screening for distress and psychosocial needs (including, physical, social, practical and emotional, family, and spiritual/religious concerns,) Making and implementing a treatment plan to address these needs – the NCCN has evidence-based treatment	Tools and components to delivery positive outcomes: HNA tools Health literacy assessment Survivorship care plan (ideally electronic) Treatment summary	Screening for Distress includes five components: 1. completing a standardized screening tool (such as distress thermometer and problems checklist) 2. opening a dialogue and initiating a therapeutic relationship, 3. assessing risk factors and intensively

	care after treatment completed Education and support (all providers) – this may include referral, wellness events	sent to primary care Cancer care review (primary care) to be completed by primary care after treatment completed Education and support (all providers) – this may include referral, wellness events	algorithms based on assessment Referring to services as needed for psychosocial care Re-evaluating, with plan adjustment as appropriate.	Stratified care pathways Directory of services and accredited health professionals with Referral pathways; Motivational interviewing techniques and skills; Telehealth tools; Information and support services.	assessing concern(s), 4. ascertaining patient perceptions and negotiating a plan of care, and 5. providing interventions either directly or by referral when required.

Policy supporting HNA use	Move away from “one size fits all” solutions towards personalised care Patient experience on a par with clinical effectiveness and safety. Defined as a transformation in support for people living with and beyond cancer. By 2020 every person with cancer should have access to elements of a ‘Recovery Package’ – a comprehensive plan that outlines treatment as well as post-treatment support and care.	Scottish Government’s 2016 Cancer Strategy Beating Cancer: Ambition and Action “for health, social care and third sector services to deliver sustainable and innovative approaches to cancer care which meet the changing requirements of people with cancer to support them to live healthy lives at home” Catalyst to develop treatment summaries and after treatment care plans, while	HNA is a part of distress screening to allow the sources of problems which might be contributing to distress Distress causes decreased adherence, increased healthcare resource utilisations, increased anger, anxiety and depression, decreased adherence to self-care interventions such as stopping smoking or exercise and may	Evidence shows cancer survivors who have completed treatment experience a range of unmet needs. More efficient and effective ways of managing follow-up care of survivors are needed. The Australian health care system is fragmented and lacks clear coordination. (Ham & Timmins, 2015) An SCP is one way to try to ensure a consistent, coordinated management	Screening for Distress helps achieve a vision of person-centred care by allowing patients to indicate their concerns and worries and using those to drive assessment and intervention. Screening beyond typical physical symptoms to include common emotional and practical concerns. To that the health care team is in a position to address needs in a timely manner and

		also improving communication with primary care Individualising follow-up (reducing or increased based in individual need)	decrease life expectancy	plan and flow of information, to help ensure good survivorship outcomes. Improve patient satisfaction Decreased health worry/increased peace of mind in cancer survivors	respond to changing needs, this should result in the burden of suffering for patients, survivors and family members will be reduced. Better able to cope and thus experience heightened quality of life.
Possible contexts	C1 Individualised and personalised cancer care C2 Increased focus on patient decision making	C1 Individualised and personalised cancer care C2 increased focus on patient decision making	C1 Well developed care algorithms based on identified needs C2 – fragmented cancer services with funders (insurance)	C1 Individualised and personalised cancer care C2 Increased focus on patient decision making	C1 – treatment of distress is an agreed national priority for cancer care C2 – healthcare is a devolved service

	C3 Shifting services from acute sector into primary care or social care C4 Focus on wellness rather than disease in cancer care (particularly post treatment) C5 – The recovery package is an agreed funded service (and should be considered and funded through commissioning route)	C3 Shifting services from acute sector into primary care or social care C4 – Focus on wellness rather than disease in cancer care	focused on fee per service rather than holistic care C3 – Focus on increased patient decision making in care	C3 A wellness focus on cancer care survivorship (which starts at diagnosis) C4 Cross Australian guidance for survivorship care against a background of fragmentation	C3 – strong evidence based guideline for the assessment process and management of psychological distress, but limited guidance on strategies for other problems
Possible mechanisms Behavioural Responses and linkage with interventions	M1 – patient identification of holistic needs allows consultations to be to be focused on their most significant problems and this	M1 – patient identification of holistic needs with engaged communication allows consultations to be	M1 – through a problems checklist or HNA aligned with a distress assessment communication between the HCP	M1 – patient identification of holistic needs allows consultations to be to be focused on their most significant problems and this	M1 – Process is to implement to give patients options and choice in support measures

(resources)	gives empowerment and knowledge to provider to better plan care M2 The identification of problems can guide the HCP to provide information increasing knowledge of patients and self-management capabilities M3 – The information provided by the patient during an HNA can provide the basis for care plans that improving knowledge between various providers and the patient allowing more shared and aligned care	to be focused on patients most significant problems and this gives legitimisation and empowerment and knowledge to provider to better plan care M2 The identification of problems can guide the HCP to provide information increasing knowledge of patients and self-management capabilities M3 – The information	and patient can result in a perception of improved empathy M2 – the identification of needs which are contributing to distress and engaging in a dialogue can facilitated engagement in joint decision making M3 – A systematic assessment (HNA and distress screening) can mobilise resources to alleviate identified issues. Through this	gives empowerment and knowledge to provider to better plan care M2 – The assessment of health literacy and self-efficacy allows HCP to tailor individuals level of self-management which increases confidence and adherence to any strategies M3 The identification of problems can guide the HCP to provide information to the patient increasing knowledge of patients and self-management capabilities	which can give empowerment M2 - Increasing knowledge of patients can improve their confidence with dealing with their problems M3 - Individualising care to make it fit needs better and increasing compliance to suggested strategies M4 – Through integrated and regular distress screening and

	M4 – Using the HNA as a basis for discussion to develop a care plan based on patients' needs and capabilities can lead to Patient activation	provided by the patient during an HNA can provide the basis for care plans that improving knowledge between various providers and the patient allowing more shared and aligned care M4 – Using the HNA as a basis for discussion to develop a care plan based on patient's needs and capabilities can lead to Patient activation M5 – The HNA can be done in various	identification, patients can be empowered to set goals to improve their wellbeing M4 – The interactive dialogue about problems identified on the HNA and agreement about the appropriate resources helps improve patient's engagement in any planned interventions	M4 – The information provided by the patient during an HNA can provide the basis for care plans that support the affirmation of responsibility for care providers and increase the HCP commitment to survivorship care M5 – Using the HNA as a basis for discussion to develop a care plan based on patients' needs and capabilities can lead to Patient activation Facilitate communication	problem identification it can normalise and integrate the assessment and support of psychological issues changing the perception of the HCP on their role in patient support

		setting and support delivered by a variety of professionals to increase the relevance to the person affected by cancer which helps engagement		M6 – Using the HNA process can increase patient and provider understanding and communication around of an array of survivorship issues which can improve the patient’s trust of the HCP M7 – Discussion of problems identified though an HNA can provide a ‘teachable moment’ to allow HCP to promote of lifestyle and behavioural changes which are more likely to have acceptance if linked to identified problems .	

Outcomes proposed	O1 – stratify care based on the HNA to avoid unnecessary resources being directed to patient, improve self-management and or focus resources for those that need them more O2 – Increase patient participation in decision making and planning care O3 – increase self-management activity and self-efficacy. O4 – improve communication between all providers of care and patient to	O1 - to personalise care based on the HNA to avoid unnecessary resources being directed to patient, improve self-management. O2 – Increase patient participation in decision making and planning care O3 – increase self-management activity and self-efficacy. O4 – improve communication between all providers of care	O1 - through finding out the sources of distress (through problem checklist) and using the treatment algorithms distress is reduced O2 – increase patient participation in deciding support improves uptake of offered interventions O3 - Decrease psychological morbidity related to treatment and disease	O1 – To focus resources and through the knowledge of survivorship care allow services and HCP develop responsibility for these care needs O2 – Increase patient participation in decision making and planning care O3 – increase self-management activity and self-efficacy. O4 – improve understanding of HCP around survivorship issues and develop services appropriately including follow-up	O1 - through finding out the sources of distress (through problem checklist) and using the treatment algorithms distress is reduced O2 – increase patient participation in deciding support improves uptake of offered interventions O3 - Decrease psychological morbidity related to treatment and disease and increase QOL

	improve knowledge and confidence in management and where possible have care nearer to home O5 – decrease the level of unmet needs to allow patients to maximise their wellness (which may also include decreasing risks of recurrence) O6 – improve the patient experience and perception of support after treatment has completed. O7 – Decrease psychological	and patient to improve knowledge and confidence in management and where possible have care nearer to home. Where possible increase care and support in the community O5 – decrease the level of unmet needs to allow patients to maximise their wellness (which may also include decreasing risks of recurrence) O6 – improve the patient experience and perception of		decreasing duplication or omission of care O5 – decrease the level of unmet needs to allow patients to maximise their wellness (which may also include decreasing risks of recurrence) O6 – improve the patient knowledge to take actions as needed to minimise recurrence risks, manage late effects of disease and treatment and undertake surveillance of disease	O4 – clear national guidance will help ensure there is consistent compassion and support from all caregivers with a coordinated services. O5 – Improved accessibility of information (including patient’s medical information) for patients and other HCP

	morbidity related to treatment and disease	support after treatment has completed. O7 – Decrease psychological morbidity related to treatment and disease O8 To optimise communication and work between secondary and tertiary hospital cancer services and primary and community health services, social care and third sector to ensure acute care is not only supportive but meets the needs of			

		the individual.			
Strengths and limitations (evidence)	Has undertaken economic evaluation but no clear findings due to wide variability and poor data (Optimity Advisors, 2016) No other strong evidence	Economic evaluation of some projects but no overall evaluation possible to due variability in implementation projects (Johnston & Campbell, 2018a) Realist evaluation of HNA (Johnston & Campbell, 2018c) Individual evaluations of 25 projects available with additional information on	Good evidence linking distress to outcomes Strongly evidence base	Two reviews – Survivorship care plans – evidence viewed positively but no clear measurable evidence of benefit (Kinnane et al., 2016) and second on needs assessment tools and what to recommend – the psychometric testing was variable and no clear recommendation.(Jiao et al., 2015)	In supporting evidence distress and multiple unmet needs are linked (evidence based) but detection and actions do not necessarily support this.

		what worked well and what did not			
Scope	This is an integral part of the English Cancer policy and defines what service provision should be within the NHS	TCAT is named and the recovery package are part of Scotland's Cancer plan (Scottish Government, 2016a)	The NCCN is a US wide organisation and this is banded as national guideline. They have done In August 2012, the Commission on Cancer (CoC) of the American College of Surgeons (ACS) released new accreditation standards for hospital cancer programs. Their patient-centered focus now includes screening all patients with cancer for	Does not appear to be comprehensive country wide policy (possibly due to mix of federal, state and private funding and supply (Ham & Timmins, 2015) – but rather a recommendation of the Australian Cancer Survivorship Centre	

			psychosocial distress. These standards are required for accreditation and were enacted in 2015. Survey shows that under half of 70 centres have implemented before mandatory in 2015 (Lazenby et al., 2015)		
Organisations involved	Macmillan recovery package (England) England – Macmillan Cancer Support (3rd sector) and English commissioners for healthcare (England wide funding decision	Macmillan recovery package (England) England – Macmillan Cancer Support (3rd sector) and English commissioners for	Institute of Medicine (IOM) report, Cancer Care for the Whole Patient Endorsed by The American Psychosocial	Australian Cancer Survivorship Centre	Canadian Partnership Against Cancer Canadian Partnership Against Cancer

	makers), all health boards in England	healthcare (England wide funding decision makers), all health boards in England	Oncology Society (APOS), the Association of Oncology Social Work (AOSW), and the Oncology Nursing Society (ONS) published		(Cancer Journey Advisory Group) and Canadian Association of Psychosocial Oncology
Relevant policy documents	Living with and Beyond Cancer Programme Initiation Document Working Together Programme, (Webster, 2015), Living with and Beyond Cancer: Taking Action to Improve Outcomes (Macmillan Cancer Support, 2013), Achieving World-Class Cancer Outcomes (The Independent	Living with and beyond cancer Programme Initiation Document Working Together Programme, Webster, 2015), Living with and Beyond Cancer: Taking Action to Improve Outcomes (Macmillan Cancer Support, 2013), and Macmillan recovery Package	Institute of Medicine (IOM) report, Cancer Care for the Whole Patient	Clinical Oncology Society of Australia Model of Survivorship Care Working Group. Model of Survivorship Care: Critical Components of Cancer Survivorship Care in Australia Position Statement (Clinical Oncology Society of Australia, 2016) Australian Cancer Survivorship Centre;	Screening for Distress, the 6th Vital Sign: A Guide to Implementing Best Practices in Person-Centred Care. A Pan Canadian Practice Guideline: Screening, Assessment and Care of Psychosocial Distress (Depression, Anxiety) in Adults

	Cancer Taskforce, 2015) Macmillan recovery Package (Macmillan Cancer Support, 2018) Stratified pathways of care for people living with or beyond cancer. (NHS Improvement (Cancer), 2016) Implementing the Cancer Taskforce Recommendations: Commissioning person centred care for people affected by cancer. (NHS England, 2016)	(Macmillan Cancer Support, 2018). Scottish Government Cancer Plan Beating cancer: ambition and action (Scottish Government, 2016a)		Needs Assessment Tools for Post Treatment Cancer Survivors: Literature Review (Jiao et al., 2015) Australian Cancer Survivorship Centre; Needs Assessment for Cancer Survivors : Toolkit. (Wiley et al., 2015) Australian Cancer Survivorship Centre; Survivorship Care Plans: Literature Review. (Kinnane et al., 2016) Australian Cancer Survivorship Centre; Survivorship Care Plans: Toolkit (Wiley et al., 2015) Cancer Australia (2017). Principles of	with Cancer (Howell, Keshavarz et al., 2015)

				Cancer Survivorship.	

HCP – healthcare professional PAC – person affected by cancer – this will usually mean patients but can also extend to caregiver and family SCP – Survivorship Care Plan

Appendix 12 - Review literature informing initial theory generation

No	Author	Studies included	Intervention and controls and outcomes	Search strategy	Authors findings /conclusions	Relevance to theory	Justification and notes relevant to inclusion
R1	(Carey et al., 2012) Examination of the effectiveness of HNA and associated interventions to meet unmet care needs	Randomised controlled trials (7) Quasi randomised controlled trials (2)	Normal care vs specific interventions responding to an unmet need	Years 2000-2010 Medline, CINAHL, PsycINFO and Web of Science	6 studies had no effect 3 some intervention effect. One in a subgroup with high psychological needs and the other two across overall prevalence of unmet needs	Self-care interventions may be more effective in those with high needs or those with high problem-solving ability. Low levels of needs may preclude an intervention effect Broad needs assessment across a variety of needs may have limited use as a screening device or as an outcome to assess efficacy HNA for cancer 'survivor' as opposed to active patients' needs to be used (to capture appropriate needs based on time in journey)	Tested interventions to reduce unmet needs – but this was also limiting as would have excluded that had other outcomes such as improved QOL Only included comparative studies of interventions (to assess for treatment effects) 'unmet needs' was the only search term for

No	Author	Studies included	Intervention and controls and outcomes	Search strategy	Authors findings /conclusions	Relevance to theory	Justification and notes relevant to inclusion
						Dose effect may be relevant for interventions (such as intensity or duration) Low levels of adherence to interventions is an important component (healthcare professional and patient) Some needs may not be met such as fear of recurrence Macvean study reported much higher baseline needs in intervention (not clinically significant) but very high adherence to the developed self-care strategy (60% of patients reported using 75% of	this concept which tools commonly call problems or concerns as well

No	Author	Studies included	Intervention and controls and outcomes	Search strategy	Authors findings /conclusions	Relevance to theory	Justification and notes relevant to inclusion
						their shared developed interventions) This could indicated shared planning can improve compliance/outcomes	
R2	(Carlson et al., 2012) To examine studies which summarise the need and process for assessing distress and unmet needs as well as the benefits of assessing these unmet needs	14 trials in total Randomised controlled trials (7) Non randomised (7) Of these 5 examined needs 2 RCTs and 3 other as outcomes	Impact of screening vs no screening Screening and interventions vs screening Multiple outcomes such as distress (HADs), QOL and needs	Web of Knowledge and PubMed No start date to September 2011	Detection of needs does not impact reduction (included when fed back to clinician) Those with more symptoms access more services but does not impact distress or psychological morbidities There was no impact on reducing needs Four studies had an effect on distress and/or QOL but these did not have needs screening	This review was primarily focused on distress but others Screening might increase referrals Authors highlighted intensive training was a factor in successful trials – however this was on distress reduction and did not measure needs	High chance of type II errors due to small sample sizes

No	Author	Studies included	Intervention and controls and outcomes	Search strategy	Authors findings /conclusions	Relevance to theory	Justification and notes relevant to inclusion
R3	(Schouten, Avua, et al., 2019) To examine the effectiveness and safety of screening of psychosocial well-being and care needs of people with cancer. To explore the intervention characteristics that contribute to the effectiveness of these screening interventions	26 studies (18 RCTs and 8 NRCTs) with sample sizes of 41 to 1012 participants, involving a total of 7654 adults with cancer.	Impact of screening for psychosocial well-being and care needs of people vs normal care (no screening) Note screening was ; Distress screening +/- problems assessment (5) HRQOL (8) Needs assessment (8) psychosocial symptoms or	The Cochrane Central Register of Controlled Trials MEDLINE Ovid Embase Ovid PsycINFO CINAHL EBSCO (Cumulative Index to Nursing and Allied) Clinical trials registries US National Institutes of Health Ongoing Trials Register ClinicalTrials.gov (clinicaltrials.gov); UK National Research Register (webarchive.nationalarchives.gov.uk); ISRCTN registry (www.isrctn.com/); Dutch trial register (NRT)	Outcomes (HRQOL, Distress and Care needs) HRQOL (20 studies) Eight found beneficial effects of screening for several subdomains of HRQOL, and 10 found no effects of screening. One study found adverse effects, and the last study did not report quantitative results. Distress (16 studies) two found beneficial effects of screening, and 14 found no effects of screening. Care needs (7 studies) Three found beneficial effects of screening for several subdomains of care needs, and two found no effects of screening. One study	Review did not find a positive impact of screening Theorised that there is a level of distress or problems – and the effectiveness will not be seen unless there is a high baseline Reason for negative effects theorised at focus on problems or dependency possibly a consideration Single or multiple screening did not seem to make a difference	Authors highlighted this was the largest and most comprehensive review of this topic Evidence very heterogenous

No	Author	Studies included	Intervention and controls and outcomes	Search strategy	Authors findings /conclusions	Relevance to theory	Justification and notes relevant to inclusion
			overall well-being (6) (NOTE: one study used two methods de Leeuw)	(www.trialregister.nl/trialreg/ index) RePORT Expenditures and Results (RePORTER) query tool All from start of database to Jan 2018 Hand search of Psycho-Oncology and Supportive care in Cancer (2010 to 2018)	found adverse effects, and the last study did not report quantitative results. No recurring relationships were found between intervention characteristics and the effectiveness of screening interventions. Authors concluded there is not sufficient evidence to support the effectiveness of screening of psychosocial well-being and care needs in people	Supported (i.e. by nurse) or self completion did not make a difference	
R4	(Bidstrup et al., 2011) To examine the effectiveness of screening on	Seven RCTs. with sample sizes of 80 to 1134 participants, involving a total of 2597	All had screening intervention with tools (always distress but often additional tools)	Embase/Medline and Web of Knowledge abstract databases from inception to September 2010 and hand searched any similar previous reviews	Meta- analysis not possible due to heterogenicity of trials. Three showed positive effect, three showed no effect and one inconclusive (only impact on those depressed at baseline).	Lack of knowledge of desire for help (one study indicated only 1/3 took up offered help) but Accepting a referral was the best predictor of	Distress is a key rationale for undertaking HNA in some regions HNAs used in evaluation

No	Author	Studies included	Intervention and controls and outcomes	Search strategy	Authors findings /conclusions	Relevance to theory	Justification and notes relevant to inclusion
	psychological wellbeing	adults with cancer.	Three included specific needs assessment tools		Outcomes included distress, anxiety, depression and HRQOL	improvement in one of the studies Acceptability of treatments an important consideration Raise question if threshold of distress needed to demonstrate effectiveness	
R5	(Meijer et al., 2013) To evaluate the effect of screening cancer patients for psychological distress by examining the effects of	There were 14 eligible RCTs for treatment of distress, and 1 RCT on the effects of screening on patient distress.	Articles were included if they (1) compared treatment for patients with psychological distress to placebo or usual care in a randomized controlled trial (RCT); or (2)	CINAHL, Cochrane, EMBASE, ISI, MEDLINE, PsycINFO, and SCOPUS databases were searched through April 6, 2011 with manual searches of 45 relevant journals, reference list review, citation tracking of included articles, and	There were 14 eligible RCTs for treatment of distress, and 1 RCT on the effects of screening on patient distress. Pharmacological, psychotherapy and collaborative care interventions generally reduced distress with small to moderate effects. One study investigated effects of screening for distress on psychological	Only RCTs that limited inclusion to patients with high levels of distress, rather than all patients with cancer, were included because this is what would occur in a screening program. Indeed, patients with low levels of distress experience only negligible benefits from	Did not evaluation HNA – but did evaluate interventions after screening. May provide some guidance in what works in with high levels of distress

No	Author	Studies included	Intervention and controls and outcomes	Search strategy	Authors findings /conclusions	Relevance to theory	Justification and notes relevant to inclusion
	screening for distress on distress outcomes and secondly assessing the effectiveness of interventions to reduce distress among patients identified as distressed	total sample size per study ranged from 55 to 472.	assessed the effect of screening on psychological distress in a RCT.	trial registry reviews through June 30, 2012.	outcomes, and it found no improvement.	psychosocial interventions in cancer settings. Pharmacological, psychotherapy and collaborative care interventions generally reduced distress with small to moderate effects. Screening as an intervention has no effect Questionnaires can be used to support discussion about psychological issues Important to separate distress from physical issues	

Appendix 13 – Presentation of initial programme theories

How an HNA package can support of self-management in maximising wellness		
Bandura Agency categorization		Linked behavioural factor from policy and research
	Theory	
Collective	1. **Shared decision making** - When practitioners seek to involve patients in their care (C) they will use HNA (M1- resource) to support the identification of problems as the patient's active participation (M2 - response) improves both the patient satisfaction (O) with the service they receive, and the provider feels reassured (M3 - response) that they are meeting the needs of the patient (O)	Shared decision making (Australia) Giving choice and empowerment (UK and Canada) Individualising care (Canada) Some evidence joint planning after HNA improves outcomes (and adherence (Carey et al., 2012) Uptake of interventions can be poor, so it is import to assess acceptably (Bidstrup et al., 2011)
Proxy	2. **Legitimising Needs** – In a consultation with a HCP (C) and patients are provided with a HNA (M) it may legitimise their problems and issues (M1 -response) and provide 'permission' to ask	Legitimacy (Study 1)

	questions as this legitimises their support needs (O) and also provides confidence (M2) what is on the form is OK to ask about	Can support discussion or brining up of psychological issues (Meijer et al., 2013)
Proxy	3. **Cognitive problems –** Patients who experience significant loss of cognitive function (C) related to their brain the tumour may lack confidence in their ability to self-manage or make decisions related to their care (C), however a disease specific HNA (focused on neuro cognitive symptoms) undertaken by a practitioner (M1 - resource) who can give specialist advice on activities which are achievable could improve motivation (O)	Confidence (or lack of) in management of neurocognitive impacts was common (study 1)
Collective	4. **Change of relationship and dependency –** In patients who experience personality or behavioural changes (C) carers may feel inadequate or distressed in managing care and isolated due to a change in relationship (C) however disease specific holistic assessment of both the carer and patient (M-response) with specialist support and advice (M- resource) can increase coping (O) and 'self' management (O)	Change in relationship (Study 1) Isolation (Study 1)
Personal	5. **Burden of unmet need -** If patients present with many needs (C) – particularly high levels of psychosocial needs causing a significant burden and discomfort (C) through an HNA which targets their problems and results in a care plan to address these (M-resource) can may allow patients to feel in control (M-response) and activated for change (M-response) and through this, support the adherence to advised self-care strategies (O) .	Motivation increases due to high discomfort (Carey et al., 2012) There may a threshold of (high) need before any intervention has an impact (Bidstrup et al., 2011; Schouten, Avua, et al., 2019) Patient activation (UK)

Collective	6. **Peer support** – For patients who participate in groups with other brain tumour patients as part of their recommended care plan within an HNA programme (C) that use methods of peer support to develop self-care knowledge and skills (M- resource) may show efficacy in improving outcomes through that building trust relationships (M - response) based on shared experience that allow a forum for exploration and validation of options (M-resource) and improve adherence to self-care strategies (O)	Peer support (USA) Patient activation (UK)
Collective	7. **Self-sufficiency -** When a brain tumour patient and their family view themselves as 'self-sufficient' (C) and a disease specific HNA is undertaken (M-resource) and they are involved in the shared development of this (M-resource), this can develop feelings of trust (M-response) which supports the development of a problem-solving approach (O) decreasing reliance on health care services (O)	Choice and Empowerment (UK and Canada) Build trust (USA) Trust in expertise (Study 1) Problem-solving (Study 1) Supporting individuals to self-manage according to preferences (Australia)
Proxy	8. **Improved communication between providers** – When primary treatment is completed for brain tumour patients with complex needs (C), and a disease specific HNA, care plan and treatment summary shared with the primary care team (M-resource), can improve patient engagement with this team (O) as patient may feel confidence (M-response) that the non-specialist providers, or primary care have knowledge give advice and suggest interventions which are appropriate to their situation and therefore improve the utilisation of primary care services (O)	Improving knowledge between providers (UK) Ensure continuity of care (USA) Confidence

Personal	**9. Individualised care –** In consultations with brain tumour patients (C) when HNA is used to help develop individualised care plans and information (M - resource) which supports tailored self-management that is achievable for individuals, it improves their feelings of confidence (M - response), thereby improving adherence behavioural changes which support wellbeing (O)	Giving choice and empowerment (UK) Patient activation (UK) Individualising care (Canada) Supporting individuals to self-manage according to preferences (Australia)
Collective	10. **Trust in practitioners** – Due to the rarity of the disease and variable presentation patterns, brain tumour patients often have had sub-optimal management on diagnostic pathways from primary care (C) and some patient then feel mistrust in the knowledge of practitioners outside their specialist team. When HNA is completed the patient and a specialist practitioner (M-resource) and part of the care planning includes advising self-care interventions (M- resource) patients may have feelings of confidence and trust (M - response) that the advice or action is correct and this supports their development of self-management strategies (O)	Study 1 – needed expert practitioner who they trusted to advise Build trust (USA) Increasing knowledge of patients (UK) Patient activation (UK) Supporting individuals to self-manage according to preferences (Australia) Improve coping strategies (USA) Some evidence joint planning after HNA improves outcomes (and adherence (Carey et al., 2012)

Proxy	11. **Not responding to HNA findings or lack of holistic approach** If needs are identified (C) the practitioner does not respond to the needs identified in an HNA (M- resource) it causes the patient to feel their needs are not important (M response) and they become passive actors in their healthcare and wellbeing may decrease (O) and distress may increase (O)	Lack of response decreases wellbeing (Carey et al., 2012) Decrease distress (USA and Canada) Use of HNA alone does not reduce distress (Carlson et al., 2012)

C = context, M – mechanism, O = outcome

Appendix 14 – Exemplar of meeting notes in theory discussions

Meeting notes: initial programme theory developing towards candidate theories.

Initial theory review - 12th of July 2019

Attendees: JA, LN, and LJ.

- Consider looking at linear progression of HNA package and how this there may be precursors to get from one point to the other (for example, what contexts need to be in place). Also think about that an outcome in the early stage may then become the mechanism to take the individuals to the next stage (for example, identification of need as an early outcome then becomes the mechanisms for personalised care)
- Is the bold labelling helping or constricting at this point (for example is this restricting thinking)?
- There needs to be some prioritisation. It may be useful to think about main outcomes first – and then grouping according this.
    - What do I consider to be the most important outcomes - ultimately self-management but what is the one before this – is it the manifestation of self-efficacy (which can improve the 'coping' of these individuals in self-management). Question raised about the role of distress
- Theories 1 and 4 possibly need to think whether the main aspect is shared decision making.
    - What is the mechanism of importance here, need to distil this? The involvement of carers when disability is high is of paramount importance – and within this theory 7 links in as well – so there is a consideration of shared decision making, involvement of the family and perception of self-efficacy. On the other side is trust …
- With theory 2 and 11 there are clear links - Is it legitimacy? Or permission to discuss (and input and direct the discussion to what they think is important. Is it trust – think about mapping elements of this to determine what are the important issues Distress – is this a concept I want to bring in – This may relate to both to some extent as not listening devalues – but I think it is the longer-term issues of both no reaction in consultation vs not following through or not giving 'good' support or advice that is trusted? The outcome is a psychological outcome – and distress is the probably the right word – but it is not cancer related distress – as it is more of the unpleasant experience

- Theory 3 and 10 possibly link – there is the rarity of the disease – the neuro cognitive side effects and need for specialist care – with very good knowledge (of what is appropriate) trust is gained and increases confidence of the individuals – core ‘things’ are trust and expertise
- Theory 5 – need to have some discussion about the process of elimination of this. Due to the high level of symptoms and unmet need in this group – the issue of threshold might not be as significant (need to discuss further and bring in Rooney study and other HNA study in brain)
- Theory 6 – peer support – probably eliminate as well – challenges with coordination of this group – but maybe has some relevance for discussion with decision making (shared), or validation
- Theory 7 ties in with Theory 1 and maybe the main outcome is self-management and mechanism of importance is problem-solving
- Theory 8 – probably remove or look at how this ties in with theory 10 in the longer term. Revisit relevance under Theory 10 in discussions
- Theory 9 can probably be considered under shared decision making

Appendix 15 – Example of data extraction and analysis for literature

<table>
<tr><td>Reference</td><td colspan="2">Hickmann, A. K., Hechtner, M., Nadji-Ohl, M., Janko, M., Reuter, A. K., Kohlmann, K., … Renovanz, M. (2017). Evaluating patients for psychosocial distress and supportive care needs based on health-related QOL in primary brain tumors: a prospective multicenter analysis of patients with gliomas in an outpatient setting. <i>Journal of Neuro-Oncology</i>, <i>131</i>(1), 135–151. https://doi.org/10.1007/s11060-016-2280-0</td></tr>
<tr><td>Date Read</td><td colspan="2">23 August 2019 and 10 February 2020</td></tr>
<tr><td>Location on Mendeley (folder)</td><td colspan="2">Realist review – Neurocognitive</td></tr>
<tr><td colspan="3">Brief description of study or document:

This is quantitative study looking at correlations. It looks at the relationship between the distress thermometer, EORTC QOL questionnaire/BN 20 (HRQOL), the supportive care needs survey SF 34 (HNA). Interestingly they did not use the brain additional questions for the supportive care needs (designed by Janda et al., 2006). They found high distress and high supportive caring needs correlate with the worst quality-of-life this involved the study of 167 patients it was undertaken in Germany.</td></tr>
<tr><td>Relevant comments related to methodological quality of literature relevant to realist review (quality appraisal)</td><td colspan="2">This study is well reported arms statistical tests seem valid. Group variability is well reported.</td></tr>
<tr><td>Notes on usefulness and contribution to theory</td><td colspan="2">This was the study focused on the same clinical population looking at needs for brain tumour patients. It is one of the few studies looking at unmet needs in brain tumour patients

This provides some relationship between context and outcomes so was retained (although mechanisms were mentioned in discussion)</td></tr>
<tr><td colspan="2">List of potential CMO combinations which appear to be in play</td><td>Relevance rating
High</td></tr>
</table>

	Which theories are these relevant to?
This study supports that it is important to ask the right questions. This study used the generic supportive care needs survey (without the Brain tumour SCNS designed by Janda et al 2006 – they did not mention this so they may not have been aware or it may be it was not validated for this population (German). They highlighted that this generic cancer needs assessment tool did not identify these but that patients should be carefully screened for their subjective perception of cognitive problems. The presence of even low levels of cognitive impairment was most highly correlated with distress. The authors also highlighted that these cognitive changes do not necessarily correlate with measurable cognitive deficits but rather are related to the patient's perception and impact of these on their life in areas such as their emotional or social functioning.	1
Possible CMOs *Neurocognitive deficits link with distress (C)* *A brain specific HNA is important to provide a subjective assessment (M) due to the underlying perceived cognitive impairments as recognition (M) and support/treatment (M) of these which ensures the patient voice is heard. It may be the most important factor in decreasing distress (O) and improving HRQOL (O).*	
They say that in many clinical trials, quality-of-life is a surrogate marker for a good outcome, but they feel there is no threshold for brain tumour patients to discriminate between a symptom requiring intervention and one that is manageable	3
Possible CMO *Patients may have difficulty in problem-solving around their symptoms (M) due to neurocognitive issues (C) – or maybe could this be lack of knowledge on what can be done – this therefore could be a mechanism that need to be introduced*	
There is one interesting contribution to theory they mention the level or type of symptoms, particularly cognitive impairments, may influence functioning this may necessitate the need for supportive care as opposed to self-care.	3
Possible CMO *This could highlight the need for stratified care (M) in those that have higher neurocognitive deficits (C)*	

They also highlight that this is different from other cancers where often the co-existing psychological comorbidities may indicate a need for supportive care however in brain tumours is because of the location in the CNS the results obtained on these questionnaires may be linked to the impact of the tumour rather than a reaction to coping with a life-threatening illness. Possible CMO *This highlights possibly the need to consider this in a Brain Specific HNA – what is it assessing – possible context of how assessment is done (M)?* *Possibly might be something about care planning (M)*	1 or 3 (maybe)
Any additional contribution of what works for whom in what circumstances (do theories need refined) The impact of cognitive impairment on problem-solving (theory 3) Is there something about the origins of depression related to tumour location … not sure if relevant to consider in assessment (theory 1)	

Appendix 16 Contribution to theories table

Author (s) and date	Type of study	Theory 1	Theory 2	Theory 3	Theory 4
Ahmed et al., 2014	Narrative review	x			x
Armstrong et al., 2012	Quantitative study	x			
Bainbridge et al., 2011	Survey				x
Bergo et al., 2016	Systematic review			x	
Biddle et al., 2016	Qualitative study	x			x
Blum et al., 2014	Mixed methods				x
Boele et al., 2017	Observational quantitative study		x	x	
Boele et al., 2013	RCT		x		
Børøsund et al., 2014	Qualitative study	x			x
Cavers et al., 2013	Qualitative study	x	x		
Clarke, et al., 2019	Two phase qualitative study	x		x	x
Dilworth et al., 2014	Systematic review				x
Ford et al., 2012	Systematic review	x			
Girgis et al., 2009	RCT	x	x		x
Green et al., 2017	Mixed methods	x			x
Greenhalgh et al., 2013	Qualitative study				x
Halkett et al., 2015	Observational quantitative study		x	x	
Halkett et al., 2018	Observational quantitative study		x		
Handberg et al., 2018	Qualitative study				x
Heyn et al., 2013	Mixed methods	x			
Hickmann et al., 2017	Observational quantitative study	x		x	

Author (s) and date	Type of study	Theory 1	Theory 2	Theory 3	Theory 4
Janda et al., 2006a	Qualitative study	x			
Johnston & Campbell, 2018c	Realist evaluation	x			x
Korzeniowski et al., 2016	Qualitative study				x
Langbecker et al., 2017)	Qualitative study		x	x	
Langbecker & Yates, 2016	Observational quantitative study	x	x		x
Légaré et al., 2018	Systematic review	x			
Lucchiari et al., 2010	Cohort study			x	
Lucchiari et al., 2015	Mixed methods			x	
Madsen & Poulsen, 2011	Systematic review		x		
Newberry et al., 2012	Observational quantitative study		x		
Paterson et al., 2015	Cohort study			x	
Pereira et al., 2016	Survey				x
Piil et al., 2015a	Qualitative study			x	
Piil et al., 2015b	Qualitative study	x	x	x	x
Renovanz, Maurer, et al., 2018	Cohort study	x			x
Renovanz et al., 2017	Observational quantitative study			x	
Rotenstein et al., 2017	Opinion	x			x
Rogers et al., 2011	Survey				x
Rogers & Lowe, 2014	Evaluation	x			
Seekatz et al., 2017	Observational quantitative study	x	x		

Author (s) and date	Type of study	Theory 1	Theory 2	Theory 3	Theory 4
Sloane et al., 2016	Observational quantitative study	x		x	
Trad et al., 2015	Observational quantitative study		x		
Thewes et al., 2016	Qualitative study	x			x
Wells, Semple, et al., 2015	Survey				x
Wells, Cunningham, et al., 2015	Survey				x