An exploration of apatheia and the definition of apathy: Understanding people's experience of apathy in Huntington's disease.

by

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Abstract

Apathy is broadly defined as a loss of motivation and seems to be a relatively common clinical problem in neurodegenerative disorders such as Huntington's disease. The definition and conceptualisation of apathy, however, is unstandardised, which leads to confusion about what precisely apathy is and how to identify it. This thesis sought to clarify the concept of apathy.

First, an etymological exploration of the concept of apatheia, including comparing it to its modern derivative, apathy, helped to give context to what apathy may be. Building on this, a systematic review looked at how apathy is defined and measured in clinical literature, finding a lack of standardisation but some common ground in terms of how recent authors have thought about apathy. Semi-structured interviews with people with apathy in Huntington's disease, alongside measures of apathy, explored what it is like to experience apathy and found that people struggle with their identity following an experience of apathy. This led to the uncovering of two types of apathy; bewildered and empty apathy. These terms were discussed in relation to the work conducted in the previous chapters and compared with some of the conceptualisations of apathy in the literature. Directions for future research were discussed, with emphasis on identifying different apathy phenomena and using the positive elements of apatheia in helping to realign people's identity. This would enable future work to concentrate on identifying appropriate treatment and management techniques to alleviate the burden of apathy in chronic illness.

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Author’s declaration

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I confirm that any patient information obtained to produce this piece of work has been appropriately anonymised.

Preface

My interest in the study of apathy stems from observing complexities surrounding this subject matter both as a phenomenon in societal life and as a concept in medical literature. On first inspection, apathy is a relatively simple word, seldom used in everyday conversation, but relatively easily understood. Describing either oneself or someone else as apathetic suggests that a person does not care about something or is indifferent with respect to a certain issue. People may talk about apathy in relation to a specific task or activity; one may be apathetic about cricket, for example, meaning that a person simply does not care about watching, playing, or understanding that sport. In this context, apathy does not imply a clinical or medical problem but merely a lack of caring that is specific to one phenomenon. If, however, a person presents to a medical professional and is disinterested in much of their life, appears to lack motivation, and has little emotional response, it is possible that they are suffering from a clinical problem we may call apathy. We can suggest that the way in which the word apathy is generally used is similar to how the word depression is used; it can be both a way of exaggerating one's mood for comic or conversational effect or, more unusually in the case of apathy, a sensitive disclosure of a serious clinical problem. With the growing recognition of mental health problems, the former, slightly trivialising way of talking about depression is arguably becoming stigmatised, but some may describe themselves as depressed when it is more likely that they are simply sad or frustrated because of a specific problem at that time. Similarly, people describing themselves as apathetic are likely exaggerating or simply do not care about a specific activity, rather than being apathetic about a range of activities they previously enjoyed and therefore are potentially suffering from a clinical problem.

Thus, we can see a clear distinction between a social understanding of the term 'apathy', and the medical, clinical importance of the concept of apathy. This distinction will be revisited throughout this thesis.

When apathy is used in a medical context, it is generally referring to either a symptom of another condition or, less commonly, a separate syndrome. This is an important characteristic that remains difficult to resolve, particularly when looking at apathy in a population with many comorbid symptoms. The term itself is broadly used to refer to a lack of motivation in a patient, a definition pioneered by Robert Marin in the late 80s and early 90s (see e.g. Marin, 1991; discussed further in this introductory chapter and chapter two). Apathy is relatively common in neurodegenerative and neuropsychiatric conditions and is an important clinical issue that causes distress for the individual and their family; watching a family member or friend slowly lose interest in the things they used to be passionate about can be extremely distressing. Linked to this, the association of emotional blunting with apathy often leads to an assumption that apathy is not as distressing for patients as it is for family, but this is an issue that will be challenged in this thesis, particularly with the results from the semi-structured interview data. Developing an understanding of a person's experience of apathy will allow this thesis to develop a better conceptual understanding of what it is to be apathetic, as well as to identify areas for further research which will lead to ways to manage and treat apathy.

It was with these issues in mind that I decided to examine the experience of apathy as a problem in Huntington's disease (HD). The symptoms and clinical problems associated with HD are relatively well understood (although some of the pathophysiological mechanisms remain unidentified) but, although apathy has been noted to be a potentially common problem, no studies have reported people's experience of apathy in HD. As well as in HD, apathy has been noted in people with Parkinson's disease (PD), various dementias, and other neurodegenerative disorders. Apathy is a relatively under-researched area across all neurodegenerative conditions, but is currently best represented in studies involving people with PD. Only two studies have looked at apathy using qualitative data, both involving people with PD, and only one of them, a study by Simpson et al. (2015), focusses on people's experience of apathy.

Aim of this thesis:

The aim of this thesis is two-fold. The first aim is to understand the concept of apathy, both historically and currently, with an exploration of the etymology of apathy and a systematic review of current literature. The second aim is to explore people's experience of apathy in HD using semi-structured interviews and three measures of apathy. By conducting a conceptual study and using this to inform exploration of people's experience of apathy in HD, this thesis will add a previously unheard voice to the current literature and begin to unravel the concept of apathy and people's experience of apathy in HD, as well as be in a position to suggest avenues for future research into apathy.

Thesis structure:

The thesis consists of the following eight chapters.

Chapter 1 explores the concept of apathy at key historical points and in medical literature. It examines the etymology of apathy, its relationship to passions and emotions, changes in the meaning of the concept, and how these changes influence our understanding of apathy and 'apatheia' (its etymological derivation). It compares various definitions of apathy (both lay and medical). It explores how apathy is presented in contemporary clinical literature, particularly in PD, and explores its confusion with and distinction from depression.

Chapter 2 presents a systematic review of definitions and diagnostic criteria of apathy and their conceptual problems in HD and other neurodegenerative conditions. It also examines measurement scales of apathy. It overviews strategies, inclusion, exclusion, and selection criteria for research in this area and discusses extraction, assessment, and analysis of data. It includes a discussion about how apathy is currently viewed and what problems there may be with the concept of apathy. Of interest is the question of how the measures of apathy formulate the concept and how they therefore theoretically operationalise apathy.

Chapter 3 focuses on choosing appropriate methods to study this complex field. In order to understand the choice of methods, I discuss psychology as both a natural and social science, the problem of objectivity and subjectivity and practical implications of these concepts, and how these concepts are useful in psychological research. This chapter also explains why a predominantly qualitative technique is currently most appropriate to explore the concept of apathy. The last part of this chapter presents a summary of qualitative research methods and justifies the reasons for choosing interpretative phenomenological analysis (IPA) as the most appropriate tool to study the experience and definition of apathy.

Chapter 4 presents the protocol for this exploration and outlines the methods used for the collection of empirical data. It outlines the rationale for further exploration of apathy and how that exploration was undertaken. Eight people with apathy and HD were interviewed as well as three of the participants' family members (two participants' spouses and a parent of one participant). These interviews asked participants to talk about their experience of apathy, including how they defined apathy. The participants were also asked to discuss their general feelings about apathy. These interviews were one of the key strengths of this thesis as they presented people's experience of apathy. No study has reported experiential data on apathy; instead, studies of apathy tend towards neurological assessment or theoretical description. Three measures of apathy, that were identified in the systematic review, were also used with participants in order to test the extent to which the measures are able to operationalise apathy. This extensive exploration leads to discussion of a novel approach to thinking about apathy in subsequent chapters.

Chapter 5 reports the results of the data collection, concentrating on setting the scene for the subsequent analysis of the interviews in chapter six. Each participant and each interview is described in detail. The results from the validated measures of apathy are also reported in this chapter.

Chapter 6 analyses the results in more detail, focussing on three themes that illustrate participants' experience of apathy. The first theme concentrates on the interpretation that apathy introduced an altered or changed sense of self in the participants. The second theme identifies the emotional changes that many participants experienced, and in turn relates this to their change in identity, part of their change in self. The final theme then explores how apathy changed participants' behaviour, in particular introducing the idea of confusion or bewilderment as a behavioural difficulty. This chapter then ties these themes together, presenting a novel understanding of apathy that suggests different types of apathy exist.

Chapter 7 then discusses the topics covered throughout the thesis, incorporating findings. It discusses further the two novel types of apathy identified in chapter six. It discusses the similarities between apathy and depression, which present diagnostic and treatment difficulties, particularly when using diagnostic measures, and inquires whether apathy is an appropriate emotional response to developing HD. It continues the discussion from chapter one as to whether apathy is a unidimensional or multidimensional construct. The chapter also reflects on the use of IPA to explore apathy, on the limitations of this study, and suggests direction for future research.

Chapter 8 then draws the thesis together, offering a summary of the key findings. Appropriate, useful avenues for further research are suggested to both continue the exploration of apathy and also to develop effective management and treatment of apathy.

For ease of reading, numbering of chapters will start here from number one.

1. Chapter One – Introduction

To develop an understanding of how the term apathy is understood, this chapter will explore the definition of apathy, including:

- The history of apathy and apatheia;
- Theories of emotion related to apathy;
- How apathy is defined in lay contexts;
- How apathy is viewed in clinical literature;
- Comparisons between apathy and depression:
- A brief look at Huntington's disease; and
- A conclusion leading on to chapter two.

1.1 How has the concept of apathy developed over time?

The concept of apathy has changed significantly over a protracted length of time. Its original derivation, apatheia, referred to a state of mind sought-after by Stoic philosophers, while the modern term is largely negative in its connotations. This chapter will track that journey, a process that will help unravel people's experience of apathy and shed new light on the construct of apathy in the final chapter of this thesis. The difference between our understanding of apatheia and of apathy will give context to the difficulty in unravelling the construct of apathy.

1.1.1 The origins of apathy:

In Greek and Roman society, to describe oneself as a philosopher aligned to a particular school could be considered the equivalent to describing oneself within a religious framework. Philosophy was not merely an academic study of life but dictated a way of living one's life. Indeed, the diametrically opposed conclusions philosophers came to about how best to live a good philosophical life have been likened to the differences between the major religions (Cooper, 2012). The Greek philosophers, most notably Chrysippus, sought to bring philosophy to the masses through public oration and engagement, believing this to be of key importance to ensuring that philosophical study was able to accurately reflect the society within which it existed. In doing this, Chrysippus developed a desire to observe and document people's ordinary, everyday language usage, belief systems, and gestures (Nussbaum, 1987); a form of qualitative data collection that sociologists, psychologists, and many other social sciences still heavily rely upon. Indeed, emphasising the connection between philosophy and the social world, and as described by Diogenes Laertius (Attalus, 2015), the philosopher Zeno of Citium (c.334-261 BC) founded the Stoic school of philosophy (of which Chrysippus is widely accepted as the second founder) after conducting philosophical discourse around a painted colonnade (the *'stoa poikilê'*) in Athens, thus reaching as wide an audience as possible.

In modern life, however, standing underneath the same colonnade in Athens describing one's views on how best to live life through striving for a form of virtue, Zeno of Citium would likely be ignored and met, ironically given the word's derivation, with a form of apathy and disinterest. Philosophy has become somewhat abstracted from the world we live in and reserved for academic scholars to discuss, rather than being a tool with which to inform the general population. The lack of exposure philosophy now enjoys seems to be due to the professionalisation of the subject:

"Philosophy has become a highly organised discipline, done by specialists primarily for other specialists." Soames, 2005, p463.

Philosophical study is now so vast and all-encompassing that it is no longer accessible to a casual observer or listener but reserved for those with specialist knowledge. Further, the nature of the knowledge created is no longer distributed to a wide audience but exists, as Soames points out, within a small population. It is reasonable to suggest that the success of philosophy in antiquity widened the field to such an extent that orating from a *'stoa'* in hope of reaching 'the people' is no longer plausible (it could also be argued that the explosion in population breadth and density has a negative impact on the ability of one person to disseminate knowledge in this relatively antiquated manner). Indeed, as Soames (2005) continues, the proliferation of fields and practitioners in philosophy means that no single person can have an exhaustive knowledge of the subject. It must be pointed out that it is implausible to argue that Zeno or Chrysippus had an exhaustive knowledge of all philosophical study since this implicitly suggests that Greek philosophy was the beginning of philosophical thought, ignoring the impact of prior religious teaching, in particular Buddhist thinking, on Greek theories. Soames' point, however, that the field of philosophy is a highly specialist discipline that requires many years' study before one can begin to understand its complexity and that *stoa*-teaching methods are defunct, remains. This may seem defamatory to Greek philosophy, suggesting that it was easier to be a philosopher of antiquity, preaching from a porch, but is meant as the opposite, rather that the relatively pioneering work of Zeno and Chrysippus has rendered modern philosophy further advanced.

Hume (1896) points out that to understand the world, to be able to come up with a falsifiable explanation of what we term philosophical truth (that is a theory about the nature of being, or of being human), we must suffer. If the greatest philosophical minds could not come up with a satisfactory model of thought that could fully explain what it is to be human, how can we expect to do anything of the sort? It is irrational and conceited to suppose that we, as individuals and as a species, are able to fully comprehend life while existing within it. This is exemplified by the oxymoron inherent in knowledge acquisition; that what we see as correct today will be a mistake tomorrow and if this were not the case, we would stop making any progress in our advancement. Living with and suffering through some of our mistakes are important pathways to understanding complex phenomenon.

This notion, that knowledge acquisition is to a certain extent always at least partially wrong and prone to correction, is something analogous to modern, fast-paced, information-driven life. We often seek knowledge but are unwilling or unable to spend appreciable time on the journey towards an answer, instead engaging in solution-focussed exercises (another reason, perhaps, why philosophical study has become specialised). Philosophical reasoning teaches that the journey towards a truth is the most important part of knowledge acquisition. In the Hitchhiker's Guide to the Galaxy, Douglas Adams states that the answer to life, the universe, and everything is 42 but that we do not know the question. When a machine was subsequently built to discover the question, it was destroyed, impatiently, to make way for a hyper-space bypass; this fictional story encapsulates the difference in thought process between modern life and philosophical reasoning. Modern life (or in Adams' case, the purpose of the supercomputer Earth) is quick to find an answer but potentially does not ask the right question, whereas philosophical reasoning does not necessarily provide answers but posits a cognitive route that shapes our everyday thoughts. Again, this juxtaposition has ostracised philosophy from modern thinking and, in doing so, reserved it for the academic elite.

This mismatch in cognitive style is perhaps why we are quick to translate complex terms often with scant regard for historical and cultural context. In exploring the Greek word apatheia and the modern derivative apathy, there are some obvious disconnects that are explained by this modern trait for rejection of philosophical reason. The Stoic philosophers believed that identifying, controlling, and potentially extirpating one's *'pathe'* (often referred to as 'the passions') was key to achieving *'ataraxia'* (equanimity). If we translate this literally, we decide that the Stoics wished to dispel *emotion* (*pathe*) in order to achieve *tranquillity* or *contentment* (*eudaimonia*). This translation, while relatively easy to understand and succinct in its explanation of Stoic theorem, is overly simplistic and loses all the complexity that was carefully woven into the ideas extolled in the various works of Zeno, Cicero, Chrysippus, and Aristotle, among myriad others. A fuller explanation of *pathe* and *emotions* reveals the need for an anti-teleological approach that prioritises understanding and explaining people's experience of apathy, rather than focussing purely on the result of the experience of apathy.

1.1.2 The passions and emotion:

The Greek word *apatheia* ("a", without; "pathos", suffering, passion, experience; Oxford English Dictionary, OED, 2010) referred to a state of active freedom from *pathe* (*'the passions'*), and was considered a sought after, virtuous state of being. To elaborate on the notion of apatheia and its place within Stoic philosophical thinking, it will first be necessary to briefly explain the passions.

Pathos (plural *pathe*) is the Greek word for suffering and the antitheses of *apatheia* ("without suffering"). *Pathe*, the passions, are loosely and simply translated as emotions, although this definition should be treated cautiously to avoid the teleological approach mentioned above. Merriam-Webster (2014) states a range of definitions, including defining passion as *"emotion; the emotions as distinguished from reason"* thereby drawing a direct parallel from passions to emotion, yet this definition loses the complexity of the original phrase and directly comparing the passions and emotions is extremely difficult given their historical and cultural underpinnings (see Dryden, 2016). The passions were, briefly but more accurately, a form of emotional suffering, not just the extremely broad modern catch-all category of 'emotion'.

Dixon (2003) points out that describing human experience and cognitive response to stimuli purely in terms of one phrase, 'emotion', is a relatively new concept, perhaps around 200 years old. Prior to 'emotion' being the dominant discourse in describing human experience, philosophers and writers would use a range of words to describe what we now term merely emotions; passions, sentiments, and affections of the soul were commonly used in classical literature until somewhere between 1800-1850 when the phrase emotion became a synonym for the previously diverse language (this is discussed further below). Baldwin (1905) points out the broad nature of the term emotion in his '*Dictionary of philosophy and psychology*', describing its use as:

"…the use of the word emotion in English psychology is comparatively modern. It is found in Hume, but even he speaks generally rather of passions or affections. When the word emotion did become current its application was very wide, covering all possible varieties of feeling, except those that are purely sensational in their origin." (p316)

While the Merriam-Webster dictionary, among many others, does stick to the somewhat traditional view that the passions and emotion are synonyms, directly related and referring to the same concept, the linguistic differences identified above suggest this is overly simplistic. Indeed, the broadness of the contemporary category of emotions compared with the relative succinct nature of the passions (being a phrase used to determine emotional suffering rather than describing every emotion) would seem to distinguish the two. The use of the terms 'passions', 'sentiments', and 'affections of the soul' seem to introduce a much more nuanced state of affairs, in which one's feelings are considered part of one's whole rather than separate. In addition, the plural is almost always used in conjunction with these statements, emphasising their inherent complexity. Emotion, on the other hand, is often used in the singular; one refers to one's emotional well-being or describes oneself as emotional without being more specific. The passions should not be translated as simply emotions but instead the two should be viewed as distinct categories, perhaps emotion encompassing the passions. The passions are emotional suffering that can lead to irrational thought and are fundamentally a negative form of being, whereas emotions are a more general category that encompasses positive and negative agencies. It is interesting to note that passions were connected with secular language, while emotions were entirely non-secular; no translation of the Bible, for instance, included talk of emotions but consistently referred to the passion of the Christ or the affections of the soul. Although the passions are often described in literature as the Greek word for the emotions, this interpretation can be viewed as overly simplistic and losing the passion of the passions. In the same way that translating *pathe* as *emotion*, translating *apatheia* as *apathy* is equally simplistic and should be treated with caution.

Apatheia represents the Stoic ideal of being able to resist and eventually extirpate the passions (although it should be noted that the idea of extirpation is potentially contentious – ridding oneself of the passions would deny the existence of the passions), thereby freeing oneself from all unnecessary, distracting emotional suffering and devoting one's life to rational, errorless thought (Sorabji, 2002). Succumbing to the passions, the Stoics state, results in one existing in a state of tumult, unable to think clearly or objectively and prone to making irrational decisions, something the Stoics saw as detrimental and damaging to one's life. The passions are not, as may be logically inferred therefore, illogical feelings that do not make sense but are the extension of rational thought that may lead, subsequently, to irrational, undesirable, and unnecessary behaviour (Nussbaum, 1987). This difference is key to the Stoics' rationale, as it implies that resisting the passions in search of the state of apatheia can be achieved through the development of rational thought and logic. This is a particularly salient point as it dispels part of the oft-discussed philosophical dichotomy between reason-passion, where the Stoics can be easily misunderstood.

Various philosophers, from antiquity and modernity, have seen reason and passion as a hierarchical duality; the two are diametrically opposed and therefore behaving rationally and purely with reason remains a far more desirable state than succumbing to one's emotion. Indeed, when talking about emotion (and the passions), the word 'succumb' is often used, implying that one has failed to achieve something. Gailliot and Tice (2007), for instance, describe people as *"succumbing to their impulses in order to feel better"* when describing controlling one's emotional responses. This view, despite common misconception, is unsupported by Stoic thinking. The reason-passion duality is used as tacit agreement for Stoic apatheia; the argument stating that the Stoics would agree that reason is wholly better than passion (or emotion, if we adopt the socially accepted loose translation). This, however, displays a lack of understanding in Stoic philosophy as it presupposes that reason is logical thought and the passions illogical thought, hence being diametrically opposed. The Stoics saw the passions as extensions of, rather than a complete lack of, rational, logical thought. Therefore, the passions exist in the same cognitive space as reason and logic; to act with reason and virtue, one must extirpate the impact of the passions on one's cognition but remain aware of their presence. A comparison can perhaps be made with implicit bias. Everyone has implicit biases either for or against certain concepts or cultural practices; one may have grown up in a vegetarian family and may therefore implicitly agree with the practice without question (or rebel against that and vociferously eat meat). That bias may go unchallenged for a significant portion of a person's life. Being unaware of this bias does not mean that one does not have that implicit bias. The only way to overcome that implicit bias would be to recognise that bias and actively seek alternatives. It is the recognition and ability to control one's passions that leads to a state where reason can be sought. Hume (1896) argues against the reason-passion dichotomy and describes what may be termed the blending of reason-passion, stating that *"reason alone can never be a motive to any action of will"* (T413) and that *"reason is wholly inactive"* (T458).

According to Stoic theory, as medicine treats physical ailments, philosophy, when used and applied appropriately, is able to treat the diseased soul that is unable to think without the negative impact of the passions. When one finds oneself existing in a tumultuous state, affected by the passions and unable to think clearly and achieve apatheia, philosophical teachings can guide one back to a virtuous, balanced state that the Stoics referred to as apatheia. The passions are not to be wholly eradicated as acknowledgement of their existence precludes the possibility of destroying them – in the same way that revolution is tacit acknowledgement (albeit negative) that a system exists (Argyrou, 2013), the existence of the passions means they cannot be destroyed. It is impossible to fully extirpate the passions but instead necessary to extirpate one's negative reactions to the tumultuous impact of the passions.

1.2 Theories of emotion:

Moving on from the passions, many scientists and philosophers were interested in identifying the systems involved in people's response to external stimuli. In around 1800-1850, people stopped talking about the 'passions' or the 'affections of the soul' and started using the term 'emotions' when discussing love, hate, anger, or sorrow. As Dixon (2003) points out convincingly at length, this linguistic shift from the rich discourse of the passions to the narrow, often ill-defined discourse around emotions introduced an unintended complexity into the language and interpretation of cognition and behaviour. The three main theories that predict how emotions are experienced and felt, for instance, vary enormously, each with a different explanation of how emotions are triggered in the brain. The James-Lange theory (Angell, 1916) was developed independently by William James and Carl Lange, both early scholars of what may be deemed modern psychology. The theory stated that any physiological arousal from the autonomous elements of the nervous system are interpreted by the brain and induce an emotion. This theory suggests that emotion is a secondary feeling that is almost a pre-programmed response to physiological changes in the body. This also meant that if a body stopped experiencing physiological changes for some reason, it would not experience any emotion as the body would have nothing to interpret and induce an instinctive emotion. Often described as a common-sense model of emotional experience, the James-Lange theory is criticised for being a reductionist view of a complex process. Perhaps it suffers somewhat from its timing. The dawn of modern psychology, around the turn of the 20th century, heralded a new era for science; not only was science capable of incredible feats in the natural sciences, the social sciences were employing the scientific method in an attempt to explain human behaviour and reduce it to its constituent elements to make order from chaos. The turn of the 20th century heralded new techniques in everything from Marie Curie's pioneering work in radioactivity to the adoption of moving pictures in cinematography, leading to an era of change and adaption in much of life. The 1927 film Metropolis depicts a scientist designing and building a 'Maschinenmensch', or human machine, in order to resurrect his lost love. This hugely influential film, both in societal and technological terms, perhaps demonstrates the feeling of possibility that the turn of the 20th century heralded. The James-Lange theory, then, is perhaps a very good example of this desire to copy and reproduce human behaviour by breaking it down into small, manageable, and easily comprehendible components; if the James-Lange theory was correct, then humans were not far away from a non-fictitious 'Maschinenmensch'. The James-Lange theory also implied that humans were pre-programmed to react in certain ways to certain physiological arousals, which in turn were dictated by external stimuli. This very linear explanation of emotion means that emotional experience must be purely an external source, open to little or no interpretation.

The Cannon-Bard theory (Cannon, 1927), first published in 1927, the same year as Metropolis was released, was proposed by Walter Cannon and Philip Bard at Harvard University and was developed through a reasonably comprehensive corpus of empirical data using cats with and without intact autonomic nervous systems. Their theory differed from the now-outmoded (although still used) James-Lange and stated that physiological and emotional responses to stimuli are separate, independent but potentially parallel processes. External stimuli are thought to have elicited both physiological and emotional reactions in the nervous system potentially simultaneously. Cannon and Bard recognised that the cerebrum was key in interpreting external stimuli and that the processes involved were more complicated than the James-Lange theory allowed. The Cannon-Bard theory, opposed to the James-Lange, does allow for some internal interpretation of external stimuli as physiological and emotional reactions occur independently, so there is the potential for a mix of physiological and emotional reactions in different situations.

Finally, the Schachter-Singer (1962) two-factor theory of emotion is the latest and most complex early theory of emotional experience. This model stated that emotion is based on two factors; physiological arousal and cognitive identification. Entirely opposed to the James-Lange theory, this theory suggests that emotion precedes physiological arousal, which then influences a search for environmental factors as the source of the emotion that preceded the physiological response. The Schachter-Singer theory allows for the most agency in the experience of emotion as it allows for both interpretation and misinterpretation of external stimuli due to erroneous identification of physiological arousal, potentially in the same experience (the first emotion or the last may be misinterpretation). In other words, agents can make a mistake in their interpretation of events, implying a cognitively aware process occurs.

It is particularly interesting to note that these three theories of emotion do not discuss the impact of the emotions on the person experiencing them but are trying to break emotion down into a series of physiological, cognitive, and interpretative events. These theories are not interested in the utility of emotion, per se, but rather in the neurological processes underpinning the experience. This again emphasises the societal shift from Greek philosophising about the nature of apatheia and pathe, to the modern, scientifically minded drive towards explaining what it is to be human, not by subjectively examining one's lifeworld but by attempting to explain away neurological influence on conscience.

1.3 Apatheia:

Moving back to apatheia briefly, we can see these changes in thought between the passions and emotion and how that influences our understanding of apatheia and apathy. Apatheia, as can be seen above by its relationship with the passions, is a positive state and implies that one is able to objectively and rationally understand the world without being drawn towards irrational or illogical behaviour by extreme emotions such as distress, fear, lust, and delight (Nussbaum, 1987) and is similar to the Buddhist concept of *'upekkha'* (*'equanimity'*). It was a phrase used in many branches of Hellenistic philosophy, is a central tenet of Stoic philosophy, and was later used in many Christian writings (although with a slightly altered meaning, generally focussed on people's loss of belief in a higher being). Apatheia was key in a Stoic philosopher's journey to seeking virtue. Stoic philosophy stated that seeking virtue in life surpasses all other needs and one can only achieve the state of tranquillity or contentment (eudaimonia) by seeking virtue; in other words, without apatheia and, eventually, virtue in one's life, there is no point to life. For the Stoics, a virtuous life involved obtaining knowledge, objective wisdom, and objective truths, in ontological terms, about the world (Algra, Barnes, Mansfeld, and Schofield, 2008). Further, the Stoics valued wisdom (and therefore virtue) over everything else. The Stoic philosopher Cicero stated that:

"It is not the case that wisdom plus health is worth more than wisdom by itself separately." Nussbaum, 1987.

To obtain virtue, the Stoics developed a complex system of knowledge acquisition that posited several assumptions about the world and the nature of 'truth'. Among these was the assumption that there exists a truth in the world (a *'phantasia katalêptikê'*) but that truth can only be identified and apprehended with certainty by a disciplined, passionless individual, whereas a truth which is thought to have any flaws or imperfections in its conviction is defined as a probable truth (or *'phantasia akatalêptos'*) (Stockl, 1887). In essence, in order to apprehend the truth through absolute certainty and live a virtuous life, it is first necessary to practice apatheia, thereby emphasising the fundamental importance of apatheia.

In early Christian writing, apatheia was seen in much the same way as the Stoics and other philosophers viewed it, but also as a divine attribute displayed in Christ's humanity (Parry, Melling, Brady, et al., 2001). Apatheia was seen as a state of being in which negative emotions are not extirpated, as Stoicism practiced, but where emotions are recognised and suppressed through prayer, perhaps similar to Buddhist meditation; if one were to achieve apatheia in this sense, one would be able to recognise and resist temptation to sin and thereby truly repent (Parry, Melling, Brady et al., 2001). It is interesting that this definition implies that the process of apatheia removes agency from the individual and implies that apatheia is a divine state perhaps bestowed on a person by a divinity, whereas the Stoics were adamant that the responsibility of apatheia lay purely at the individual's feet. For the Stoics, apatheia was an active choice that required extensive effort to achieve, while early Christian writing begins to remove that active choice. This is perhaps where apatheia begins to lose some of its Stoic meaning and begins to transform into its modern and somewhat passive derivative, apathy.

1.4 Modern definitions and uses of apathy:

In contrast to apatheia, the modern derivative, apathy, refers to a *"lack of feeling or emotion, [and] a lack of interest or concern"* (Merriam-Webster, 2014). It is particularly interesting to note that apathy represents a lack of emotion which a person should want back (thereby making emotion a desirable state), whereas apatheia is the purposeful omission of emotional suffering. Indeed, the 'Tresor de la Langue Française informatisé' (the Treasury of the French Language computerised) defines apathy (*'apathie'*) as a *"state in which one becomes voluntarily estranged to the passions"* (Taylor, 2007) and the OED describes apathy in a similar vein as *"Freedom from, or insensibility to, suffering"* (OED, 2014). It is interesting that these three intensions of apathy differ slightly in their implication of the state of the person. The Merriam-Webster intension describes apathy in terms of loss, a lack of being, implying that apathy is an entirely passive process outside of one's control. The TLF uses a contradictory passive sentence structure to describe apathy while stressing the voluntary, active nature of the estrangement and uses the phrase passions instead of emotion. This seemingly odd combination is perhaps indicative of the confusion concerning apathy's definition; is apathy a similar active concept to apatheia? Or has the modern definition become entirely passive, emphasising loss over freedom? The TLF, understandably, seems unsure which is more appropriate. The OED's definition, again, seems unsure whether apathy is an active or passive state, the words 'freedom' and 'insensibility' being very different ways of experiencing suffering. Both are passive in their intent but also imply that apathy is not a lack of emotion or motivation, but a state where one is entirely ignorant of emotion. If we understand suffering in Greek terms, we can think of it further as suffering emotion, hence freedom from emotion could also imply that one is adept at understanding and controlling one's emotional response; very much an active state. This latter definition is much closer to apatheia than Merriam-Webster's definition of apathy which sticks doggedly to the more commonplace definition of apathy as a lack of agency, control, or interest.

The differences between these three intensions of apathy highlights the confusion that the word suffers and is in part due to the confusion over the use of the concept of emotion. If we take the basic, modern understanding of apathy (in particular the Merriam-Webster intension), the main features are negative loss, passivity, and a lack of feeling (avoiding the use of the word 'emotion' for clarity). This is entirely juxtaposed to apatheia, which was a positive, active, sought-after state, yet the two terms, apathy and apatheia, are confused in dictionaries, attributes of one appearing in the definition of the other. There is, evidently therefore, overlap between the two but the term apathy, in inheriting negative connotations, has become defined by loss and by what it is not, rather than what it encapsulates. Apathy exists in a strange state. It is a negative lack of emotion that one should avoid as it is implicated in becoming socially ostracised, yet the popular reason-emotion hierarchical duality implies that reason should always be sought over emotion. Does that imply that one should always be in touch with one's emotions, but never act on them, always preferring reasoned action? When put that way, apathy becomes very similar to apatheia, a previously desirable state, but is doubtless seen as a negative state of being in modern culture.

In the 20th century, apathy not only garnered negative personal connotations surrounding a loss of emotion and loss of connection from society, but also became dangerous politically and societally. The Lutheran pastor and theologian Martin Niemoller described a state of apathy in German society in his statement "First they came", describing a scenario in which an individual stands idly by while others are exterminated (Gerlach, 2000). The statement is somewhat reminiscent of Eliot's prophetic lines in The Hollow Men (Eliot, 2002), "This is how the world ends | Not with a bang but with a whimper." It is interesting that both these quotations describe the negative consequences of apathy and align it with the destruction of democracy through a civil laissez-faire, apolitical attitude. This concern for apathetic behaviour is something which certainly resonates; the New York Times columnist Charles M. Blow (2014) stated that *"Voter apathy is a civic abdication"*. The message is clear – apathy is a dangerous state of being; a far cry from the Stoics' desire to seek apatheia and align it with eudaimonia.

Apathy has also become a nuanced, context-dependent phrase that can have several interpretations in the same situation. As Lertzman (2009) points out in her thesis on the myth of apathy in climate change politics, seeming apathetic and being apathetic to climate change are two completely different states. Seeming apathetic and therefore unmotivated to engage in positive action about climate change can merely be a mask for not having enough knowledge, being fearful of change, or feeling disenfranchised by the enormity of the task. Lertzman describes this gulf between seeming and being apathetic using psychoanalysis and dubs it the "myth of apathy". When looking back at Niemoller's lambast of German society or Blow's anger at voter apathy, perhaps Lertzman's myth of apathy helps to mitigate harsh judgement of a society which seems apathetic. Indeed, Blow (2014) points out that those in power (particularly those to the right of the political spectrum) often obfuscate the truth and purposefully disenfranchise those who may otherwise oppose them. Apathy, then, is not only a dangerous personal state, but a dangerous political tool that the state can wield by proxy.

1.5 Exploration of apathy in clinical literature:

The definitions and conceptualisations of apathy will be further unpacked in the systematic review that follows in chapter two, but a broader discussion of clinical apathy is relevant here. It is important to make the distinction between dictionary definitions of apathy that a wider audience generally associate with apathy and how apathy is defined and viewed in clinical settings. There are, of course, similarities in how the concept of apathy is understood across lay and medical settings. Apathy is generally seen as a negative influence on someone's life, whether defined from a clinical or dictionary setting. Culturally speaking, apathy is a negative that, as shown above, is often viewed as an apolitical danger. Clinically, apathy could be viewed as a negative symptom in that it is often defined as a loss or lack of something, generally motivation. These definitions are slightly different; the cultural view infers a negative feeling, whereas the clinical definition explicitly talks about a negative symptom, a technical definition with a specific meaning.

Perhaps the most important clinical definition of apathy, certainly in terms of its influence on the field, is Marin's early work of the 80s and 90s. Marin's work has had an obvious and large effect on all future work, with most subsequent papers that have examined apathy either directly quoting or borrowing heavily from his definition of apathy. For this reason, a slightly extended quotation that lays out this definition of apathy is appropriate. Marin (1990) defined apathy as:

"…apathy describes only those patients whose lack of motivation is not attributable to a diminished lack of consciousness, an intellectual deficit, or emotional distress. Apathy is, therefore, a state of primary motivational impairment." Marin, 1990.

In a subsequent paper, he elaborated:

"The term apathy describes the lack of motivation seen in a variety of neuropsychiatric disorders. It is employed by clinicians to describe such familiar attributes as loss of interests, loss of emotions, flattening of affect, or loss of energy." Marin, 1991.

These two quotations are important in both defining apathy and setting the direction for future research. As mentioned above, Marin's work did seem to set the tone for future work, with most authors then using this broad outline of apathy. The central idea that Marin developed, that of apathy being a loss of motivation, continues to be a dominant theme in apathy research. While this is arguably an appropriate definition of apathy, it could also be characterised as overly broad and Levy and Dubois (2006), in particular, were critical of defining apathy as a lack of motivation as it is a psychological interpretation of a behavioural state. Marin himself notes that a lack of motivation should not be attributable to another reasonable explanation and must itself be a primary behavioural characteristic but does not explain how that would manifest. Similarly, research that relies on Marin's definition does not explain how a lack of motivation can be viewed as the primary symptom of an apathy syndrome. This is one of the main reasons Levy and Dubois (2006) specifically moved away from talking about apathy as a loss of motivation, a direction that the field seems to be following with Robert et al.'s (2018) most recent diagnostic criteria for apathy avoiding the term motivation and instead using the idea of goal-directed activity as a proxy for apathy. These problems with the definitions and conceptualisations of apathy will be addressed further in subsequent chapters.

In the fourth revised edition of Diagnostic and Statistical Manual of Mental Disorders (DSM-IV-TR; American Psychiatric Association, APA, 2000), apathy is mentioned in four disorders and only 15 times in total. There were calls before the release of the fifth edition of the DSM to include apathy as a separate construct (Tagariello, Girardi, and Amore, 2008). The American Psychiatric Association did not include a separate apathy construct, but the DSM-V (American Psychiatric Association, 2013) does now include 34 references to apathy, the main increase being in the neurocognitive disorders section.

An interesting set of studies reported in one article by Leander, Shah, and Sanders (2014) looks at people's responses to the apathy of others in health populations. In their study, they use indifference as a synonym for apathy; while this is an extremely broad definition of apathy, the study findings do help to explain some elements of apathy, specifically related to the manipulation of goal-directed behaviour (GDB), an important facet of apathy. In these studies, Leander, Shah, and Sanders broke down GDB into goal commitment (how committed a participant was to performing a goal or behaviour) and goal accessibility (how easy it was to achieve a certain goal or behaviour) and manipulated both to see the effect on levels of indifference. Indifference tended to be highest when goal accessibility was not manipulated, and goal commitment was positively manipulated to ensure participants were committed to their goal. In their conclusion, Leander, Shah, and Sanders state that their five studies show that indifference to goals is not a novel change of preference in an individual, but an external impact on pre-existing (potentially latent) preference. In other words, a person is more likely to show indifference to an activity if there is a barrier to them doing that activity that they cannot control. This lack of control may then exaggerate their desire to not do that activity, resulting in indifference.

1.5.1 Apathy and depression:

Depression and apathy are often conflated but consensus seems to be growing that they are phenomenologically different. Some have stated that apathy is not distinct from depression; Bogart (2011), for example, argued that apathy is not a meaningful syndrome in PD until it was demonstrated that apathy is highly correlated with negative patient outcomes, while Tate et al. (2003) state that depression is a much more pressing issue than apathy in people with human immunodeficiency virus (HIV). Since then, a comprehensive systematic review and meta-analysis by den Brok et al. (2015) has showed that apathy is pervasive in PD and that successful treatment of apathy would improve patients' quality of life. Den Brok et al. (2015) demonstrated that half of the patients included in their meta-analysis were suffering with apathy and no concomitant depression, and notes that different treatment strategies may be required depending on the neurological mechanisms affected by apathy, effectively stating that apathy does have its own underlying mechanism.

That there is overlap between depression and apathy is also accurate, however. Starkstein (2005) noted that both depression and apathy, in their modern guises, are associated with "reduced volition" and states that the difference between apathy and depression lies in a person's emotional mood. If a person shows reduced volition and emotional changes congruent with their mood, they are likely depressed. If reduced volition occurs without those emotional changes, a person is likely suffering from apathy. Similarly, Tagariello, Girardi, and Amore (2009) differentiate apathy and depression (when both are viewed as symptoms of a wider systemic problem) by the presence, indicating depression, or absence, indicating apathy, of negative mood. These solutions, however, are overly simplistic; depression can be diagnosed without consistent negative mood and apathy can induce sporadic negative mood (Starkstein and Leentjens, 2008). These distinctions also imply that the two cannot co-occur, although Starkstein later demonstrates that apathy and depression do often co-occur in patients when assessed with multiple rating scales. Levy et al. (1998), in their paper titled "Apathy is not depression", use a cross-sectional comparison of neuropsychiatric symptoms in a range of disorders and show that diagnosis of apathy did not correlate with depression. As Levy et al. (1998) point out, some of the previously reported overlap between apathy and depression could be down to items in depression scales using apathy as a signifier of depression rather than there being a clinical overlap. It is certainly possible for a person to suffer from depression and apathy concurrently (den Brok et al.'s (2015) systematic review stipulates this), but this does not mean that depression and apathy are always related or must occur together. Lyketsos et al. (2000) demonstrated this in their report of a large study on ageing, and the prevalence of apathy without depression is common in several types of neurocognitive disorder (Levy, Cummings, Fairbanks et al., 1998; Starkstein et al., 2001).

1.5.2 Uni-dimensional and multi-dimensional depression and apathy:

Another issue that is still being debated about depression, and has implications for the study of apathy, is the relative merits of a uni-dimensional or multi-dimensional model of depression. The DSM-V (APA, 2013), published in 2013, uses a summing of symptoms to determine if a major episode of depression is present or not, thereby assuming that depression is a unidimensional construct. While the DSM-V does contain diagnostic criteria for a large variety of types of depression (e.g. post-natal depression, bipolar disorder, and major depression), these are all diagnosed via a summing of symptoms, creating a unidimensional construct. There have been many calls for this to change and for psychiatric conditions including depression to be given a useful multi-dimensional approach. Elhai et al. (2012), for instance, found that a two-factor model of major depressive disorder (MDD), separating somatic from non-somatic symptoms, provided more explanatory power (via confirmatory factor analysis) than a one-factor model. While there are some problems with Elhai et al.'s (2012) study in terms of the generalisability of their data and issues with self-reporting of mild depression rather than clinically diagnosed MDD (problems that Elhai et al. openly discuss), the support for a two-factor model of depression seems to represent a general shift of approach (e.g. Cheung and Power, 2012; Darharaj et al., 2016).

One problem with the distinction between a uni-dimensional and multi-dimensional construct, particularly in predominantly psychological constructs, is presented by Fried et al. (2016). In this paper, Fried et al. note that to track depression, it must be assumed that any differences in sum-scores over time on measures such as Beck's Depression Inventory (BDI;

Beck, Ward, Mendelson, Mock, and Erbaugh, 1961) reflect changes in a person's depression. In assuming this, depression must be assumed to be a uni-dimensional construct that a measure such as the BDI tracks uni-dimensionally. That is, the BDI must load strongly on to one primary factor. If a measure loads on to multiple factors, then a resulting single sum-score would also represent change in all of those multiple factors and violate the assumed uni-dimensionality of the measure.

When it comes to apathy, authors frequently describe it as a concept that can be identified as uni-dimensional but also tend to refer to it as consisting of a triadic structure. Marin's (1991) key conceptualisation of apathy implies that a lack of motivation is a direct proxy for apathy, but also emphasises a triadic structure of apathy, based on behaviour, cognition, and emotion. Many other papers then paraphrase Marin's definition of apathy, that apathy is characterised by a lack of motivation, and consider that a sufficient definition without going further into possible concomitants. Similarly, Levy and Dubois (2006) refer to apathy as a pathology of goal-directed behaviour, but then immediately refer to three underlying mechanisms related to emotion, cognition, and behaviour initiation. This desire to simplify and reduce apathy to its constituent elements is laudable in its efforts to explain a complex phenomenon, but ultimately seems to lead to confusion as to whether apathy is a uni-dimensional or multi-dimensional construct. Given Fried et al.'s (2016) identification of uni-dimensional depression scales as problematic, the relative proliferation of sum-score apathy scales is potentially worrying when much of the literature already identifies apathy as a conceptually ill-defined construct that is described in confusing terms as both uni- and multi-dimensional.

Chapter two summarises these multiple definitions of apathy and provides further background.

1.5.3 Apathy in Parkinson's disease and qualitative research:

Much of the research into apathy looks at people with PD, largely because of the high prevalence of apathy in neurodegenerative conditions, with prevalence in PD of an estimated 17-70%, and because much of the underlying neurological dysfunction of PD is reasonably well understood (Pagonabarraga et al., 2015). Bogart (2011), in a review of the status of apathy in PD, notes that apathy is generally considered causally related to the neurodegenerative effects of PD rather than a psychological or psychiatric reaction. Levy and Dubois's (2006) characterisation of apathy as a pathology of goal-directed behaviour in neurodegenerative conditions adopts this understanding. In comparison, McKinlay et al.

(2008), in their study of neuropsychiatric problems in PD, suggest that apathy is correlated with both cognitive impairment and a person's perception of cognitive impairment. That is, their experience of apathy seemed to increase both when any cognitive impairment worsened, but also when they thought that their cognitive impairment was worsening. This leads to the possibility that apathy is a psychological reaction to some aspects of neurodegeneration, something that may occur in both PD and HD.

There are currently only two studies that use a qualitative form of data collection to study apathy. Both studies interview patients with PD and as such deserve exploration here. Mele et al. (2019) used the Theoretical Domains Framework (Atkins et al., 2017) and Behaviour Change Wheel (Michie, van Stralen, and West, 2011) to inform a theoretical thematic analysis of the barriers and facilitators to diagnosing and managing apathy in PD. Mele et al. (2019) interviewed healthcare professionals, people with PD, and primary caregivers to gather evidence from key stakeholders about how apathy is recognised and managed in PD. Their focus was on identifying how healthcare providers and systems can better react to potential apathy in people with PD, rather than looking at the construct of apathy itself. Mele et al. (2019) do not specifically focus on the problems with the conceptualisation of apathy, but report that the unstandardised definition of apathy is a barrier to successful treatment. In terms of treatment options, they identify non-pharmacologic treatment options as more likely to be effective, a similar conclusion to Krishnamoorthy and Craufurd's (2011) review of potential treatments for apathy in HD.

The second qualitative study of apathy in PD is Simpson et al.'s (2014) experientially-driven, phenomenological exploration. This is a similar approach to that used in the data collection in chapters five and six, and because of this direct methodological comparison, it will be particularly interesting to compare Simpson et al.'s (2014) results to those reported here. Simpson et al. (2014) interviewed seven people with PD who had or were currently experiencing apathy over the past year. Experienced clinicians who worked in a memory clinic screened potential participants for apathy. Participants with other psychiatric conditions, such as depression, were not excluded because of the large potential for overlap. In their analysis of the data, Simpson et al. (2014) present three themes related to participants' motivational impairment, participants' apathy as a protective factor in avoiding exposure to their worsening symptoms, and external factors contriving to feed into their apathy. The participants talked about the relationship between apathy and their symptoms of PD, many noting that worsening physical symptoms made them less inclined to do things. Participants noted that it was technically possible to do things, but they did not want to do something to a lesser degree or quality than they used to do. Additionally, their reduced positive mood due to symptom burden made doing potentially difficult things less appealing. There was often conflict within a participant, with doing nothing seeming appealing but a sense of pride then making them want to override that apathy. Socially, participants noted that external barriers that impacted their ability to move around or work were worsened by their feelings of apathy. If a task became harder for them, not because of worsening symptoms but because of an external change, participants were likely to avoid that task. Simpson et al. (2014) note that participants' experience of apathy as interrelated to their neurodegeneration and impairment caused by PD was contrary to earlier conclusions in the field that apathy was purely a neurological symptom and not a psychological syndrome. In looking at the future of apathy, Simpson et al. (2014) conclude that further research into non-pharmacological treatment of apathy is essential. Some studies have begun to look at behavioural interventions to help apathy, with varying success (e.g. Roth et al., 2007 looked at treating apathy in dementia), but Simpson et al.'s (2014) findings support the development of behavioural and psychosocial interventions to help people overcome their apathy. It is particularly interesting to note that participants in Simpson et al. (2014) study were particularly active in considering and talking about apathy. Most definitions or conceptualisations of apathy focus on describing apathy as a state of loss and often imply that people with apathy have lost so much motivation and emotional response that they are passive recipients of apathy. Simpson et al. (2014) demonstrate that, contrary to the idea that family members suffer from a patient's apathy more than the patient themselves, living with apathy can be a damaging, distressing experience.

Given the reasonable amount of research undertaken around apathy in PD, particularly Simpson et al.'s (2014) experiential study, and the dearth of research into apathy in HD, this thesis will focus on the exploration of apathy in HD, but will also suggest implications for the wider study of apathy in neurodegenerative conditions. While it would have been plausible to conduct a similar study to Simpson et al.'s (2014) and interview people with PD, this would leave people with HD with no voice in the field at a crucial time in the study of apathy. By exploring apathy with people in HD, comparisons can be drawn between this study and Simpson et al.'s (2014) to suggest ways in which the experience of apathy may converge and differ between neurodegenerative conditions.

1.6 Huntington's disease:

Huntington's disease is a devastating disease that blights families and causes early death in most sufferers, usually following an extended period of severely limited quality of life (Novak and Tabrizi, 2010). It is an autosomal-dominant, progressive, neurodegenerative, genetic disorder that causes symptoms including chorea, muscle spasm, a lack of muscular control, cognitive decline, and behavioural difficulties (Walker, 2007). George Huntington described the disease in 1872 (Lanska, 2000), noting in particular people's pronounced chorea. HD is a hereditary disease with a one in two chance of a parent passing the defective gene to their offspring. Despite good knowledge of some of the pathophysiological mechanisms behind the cause, there is still much that is not well understood, and no cure currently exists. Treatment tends to revolve around improving symptoms where possible and managing risk of complication or increased degeneration (Novak and Tabrizi, 2010). Onset varies from adolescence to old age, with juvenile HD being diagnosed in someone symptomatic before age 20. Disease burden worsens over time, with time from diagnosis to death being around 20 years (Folstein, 1989), and loss of insight into disease burden increases over time (Sitek, Thompson, Craufurd, and Snowden, 2014). The risk of suicide in people with HD is markedly increased over the risk in the general population, with some studies showing that 25% of people with HD attempt suicide over the course of their illness (Farrer, 1986).

Psychiatric symptoms of HD include depression, apathy, anxiety, mood disorders, and suicidal ideation (Walker, 2007). Estimates of prevalence of apathy in HD vary broadly (van Duijn, Kingma, and van der Mast, 2007) but prevalence and severity of apathy is reported to increase with disease progression in HD and is generally associated with severe cognitive deficits (Baudic et al., 2006). Krishnamoorthy and Craufurd (2011) suggest that apathy is an inevitable part of HD that worsens the longer a patient is symptomatic and is a direct consequence of advanced disease. Additionally, they state that the only approaches likely to appropriately treat apathy in HD must involve multidisciplinary teams, extensive modifications of a person's environment, provision of regular psychosocial support, and psychoeducation interventions designed to support both patients and caregivers.

1.7 Neuropsychology and underlying mechanisms of apathy:

Much of the current research into apathy looks at the neurological mechanisms responsible for apathy, most focussing on lesions or impairments of the pre-frontal cortex (e.g. Moretti and Sognori, 2016; Caravaggio et al., 2018) and how this may affect brain function in individuals with cognitive impairment due to neurodegenerative conditions. This is a key area in the progression of our understanding in apathy but is a different subject to the experiential examination of apathy. As this thesis focusses on outlining the conceptualisation of apathy and then people's experience of apathy, these neurological observations of apathy are not discussed at length. The theoretical and experiential implications of what apathy may consist are assessed. In the future, experiential data in people with apathy can be combined with underlying neurological mechanisms to form a holistic theory about the concept of apathy, but this is beyond the reach of the current understanding of apathy and of this thesis.

1.8 Conclusion:

The complicated history of apathy, from a Stoic ideal to an unwanted loss of motivation, is emblematic of several cultural shifts in both cognitive and emotional styles. Apatheia, the passions, and affections of the soul have been replaced by apathy and emotions. This shift does not necessarily carry negative repercussions, but an understanding of these derivations is useful in trying to unravel how to define, conceptualise, and measure apathy. The complex medical status of apathy further emphasises the need for conceptual clarity when discussing and potentially diagnosing apathy. Chapter two clarifies the construct of apathy by conducting a systematic review of the academic and clinical literature and using this to understand how apathy is currently thought about. Later in this thesis, these definitions will be combined with analysis of participants' experience of apathy to suggest ways in which experience can further develop the construct of apathy.

2. Chapter Two – A narrative systematic review of the definition and measurement of apathy in Huntington's disease

2.1 Introduction:

As noted in chapter one, apathy is a complex term that has been used in different ways since moving on from its derivation, apatheia. There are multiple ways of defining apathy and these must be understood when trying to conceptualise apathy. This review seeks to identify the myriad definitions and measurements of apathy and use this knowledge to inform further study of the construct of apathy.

In doing this, it is necessary to provide a brief background about apathy and HD. HD, as mentioned in chapter one, is a progressive, neurodegenerative, genetic disorder that causes symptoms including chorea, muscle spasm, a lack of muscular control, cognitive decline, and behavioural difficulties (Walker, 2007). Apathy is often defined using Marin's (1991) criteria which refers to apathy as primarily a "lack of motivation". More commonly, apathy is defined as a relative lack of interest or emotion. Developing a measure or diagnostic criteria for apathy is the subject of an increasing number of papers as apathy is recognised as a destructive condition. Apathy is a common feature in many progressive disorders such as PD, Alzheimer's disease (AD), HIV, and HD, where incidence seems to be particularly high and is estimated at between 33-76% (van Duijn, Kingma, & van der Mast, 2007). In comparison, Starkstein et al. (2009) found the prevalence of apathy in people with PD ranged from 17-70% across five studies.

There are multiple definitions of apathy, several theories about its causation, and four main sets of diagnostic criteria. Exploring the diversity of definitions and conceptualisations of apathy, as well as considering the conceptual challenges this diversity presents, are the main focusses of this systematic review. Previous reviews by Clarke et al. (2011) and Radakovic, Harley, Abrahams, and Starr (2015) have pointed out the lack of consensus in the area and this review will build on these works by outlining how the relevant papers conceptualise and measure apathy differently.

Perhaps the most diverse aspect of the literature is the relatively large number of measures of apathy that exist. There are eight apathy-specific measures, and at least five other measures include an apathy sub-scale. Part of the reason behind this relatively large number of measures is the diversity across the field and the different approaches authors take to discussing and conceptualising apathy. The diversity and difference in reported prevalence rates of apathy shown above in both HD and PD are most likely causally related to the conceptual confusion across the literature. These extremely wide prevalence rates suggest that, as well as a difference in rates across disorders, methodological variability when measuring apathy leads to a poor understanding of the phenomenon of apathy. It is likely that each paper interprets apathy slightly differently, meaning that when we discuss apathy in the context of the literature, we are referring to a subjective, non-standardised concept that is challenging to compare across the literature. The repeated calls for standardisation in the field must be addressed both here and in future research.

Another problem with apathy is that it is difficult to ascertain how much of a problem it presents as there are relatively few studies that look at people's experience of apathy and those that look at quality of life tend to be looking at a wide range of issues, not just apathy. It is therefore difficult to understood the impact of apathy on a person's life. Additionally, there is little understanding about people's ability to express their emotions in apathy. This difficulty in expressing one's feelings is perhaps one of the reasons research tends to focus on developing measures of apathy rather than developing the concept itself. There are very few papers that explore the impact of apathy on patients' lives and the lives of those around them. Tate et al. (2003) explored the impact that apathy and depression had on the health-related quality of life (QoL) of people with HIV and found that apathy occurs in around 20-30% of cases and depression occurs in 60-80% of cases. Further, Tate et al. found that depression accounted for 40% of the variance in QoL and apathy only 9-14%, leading the authors to conclude that depression has a much greater impact on QoL than apathy. This is a good example of one of the problems with a non-standardised approach to investigating the impact of apathy. As it remains unclear what the impact of apathy is on a person's life, assessing a person's QoL using the SF-36 cannot reveal the impact of apathy on their life. It is also unlikely that an apathetic person will report negative emotions or reduced physical activity as apathy often seems to cause a person to lose insight into their behaviour and feelings (or vice versa). Apathy is also less likely to impact the eight scales of the SF-36, the QoL measure used in Tate et al.'s (2003) study, than depression. The negative mood associated with depression is more likely to include awareness of one's functioning, a major component of the measure. Additionally, and perhaps more pertinently, depression is a well-researched, homogenised mood disorder with very stringent diagnostic criteria. Apathy remains an unstandardised concept. Therefore, comparing the impact of depression and apathy is unhelpful as the two disorders are so conceptually different.

This conceptual confusion over what apathy is, how it manifests, and how it is best measured is perhaps masking the clinical problem of apathy. Apathy is a distressing symptom for both patients and their family, although little research focusses on the experiential impact of apathy. Additionally, when apathy is a primary focus, the problems induced by apathy seem to be masked by the easier to identify and easier to treat problems associated with depression (as in Tate et al., 2003). The research that has looked at the impact of apathy on patients' lives suggests that apathy negatively affects self-care and glycaemic control in diabetes (Padala et al., 2008), and Benito-León, Cubo, and Coronell (2012) showed that apathy is a major cause of clinical concern in people recently diagnosed with PD, and that people with PD and apathy had significantly lower QoL scores than those without apathy.

Previous reviews that have explored the concept of apathy have tended to explore the validity of measures of apathy without exploring how those measures defined apathy, how those definitions may differ, and why there are substantial differences between the measures. This systematic review is part of a larger thesis that concentrates on examining apathy by combining an exploration of the current definitions of apathy with in-depth semi-structured interviews with people with HD who have personal experience of apathy. The review will explore the various definitions of apathy in detail, looking at how authors examine and report the definition of apathy and how the definition has been developed. By combining these data, a comprehensive picture of how apathy is viewed will provide a useful step in understanding what parts of the concept of apathy we understand, and what parts need further exploration and clarification. In addition, this review will suggest possible directions for research to understand what apathy is and how it impacts patients' lives.

2.2 Aims of this review:

Based on the previous literature, the questions posed in this review were:

1. How is apathy defined and conceptually discussed in Huntington's disease and other neuropsychiatric conditions?
2. How is apathy measured in HD and other neuropsychiatric conditions?

It must be noted that although this is a systematic review in that the methodology used to perform the searches and retrieve data is systematic, following the PRISMA guidelines, the review itself follows a more narrative structure than most systematic reviews. The aims lend themselves to a more discursive approach to interpreting the data available.

2.3 Methods:

2.3.1 Search strategy:

This review sought to determine how apathy is defined and conceptually discussed in HD and other neuropsychiatric conditions. In addition, the review was interested in how apathy is measured and how the items included in the measures of apathy reflect the definitions of apathy. Due to the complexity and range of aims of this review, the review included different types of article. Previous reviews that examined the measures of apathy or discussed the concept of apathy were included, as well papers that developed a novel measure of apathy, presented potential diagnostic criteria, or extensively discussed the definition of apathy. This systematic review followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Moher, 2009). The review protocol was registered on the PROSPERO web site (www.crd.york.ac.uk/PROSPERO; registration no. CRD42016037046) and reports a PRISMA checklist in Appendix 10.5.

In March 2016, the electronic databases MEDLINE, Ovid EMBASE, Web of Science, and PsychInfo were searched. Google Scholar was used to supplement and clarify the search (e.g. in case of conflict between databases). Search strategies included both Medical Subject Headings (MESH) where possible and text word searches to increase sensitivity. A combination of search terms from three groups were used to broaden the search results. Main search terms included "apathy", "Huntington's disease", "neurodegenerative", "definition", and "measure". The search strategy was tailored for each database (e.g. PsychInfo does not support MESH terms). A full table displaying the search strategy is below. An updated second search was conducted in September 2019 to add missing papers to the original search due to the length of time between first search and thesis submission. In the intervening period, a Google Alert had been set up to send any new papers matching the search terms to an email address. To ensure no papers were missed, however, the second search cross-referenced these Google Alert papers with a separate search on Web of Science in September 2019. The results of the second search have been reported as a second PRISMA flow diagram of this thesis and additional papers added are reported in the results section of this systematic review. Both PRISMA diagrams detailing the search results diagrams are below.

2.3.1.1 Search strategy detail:

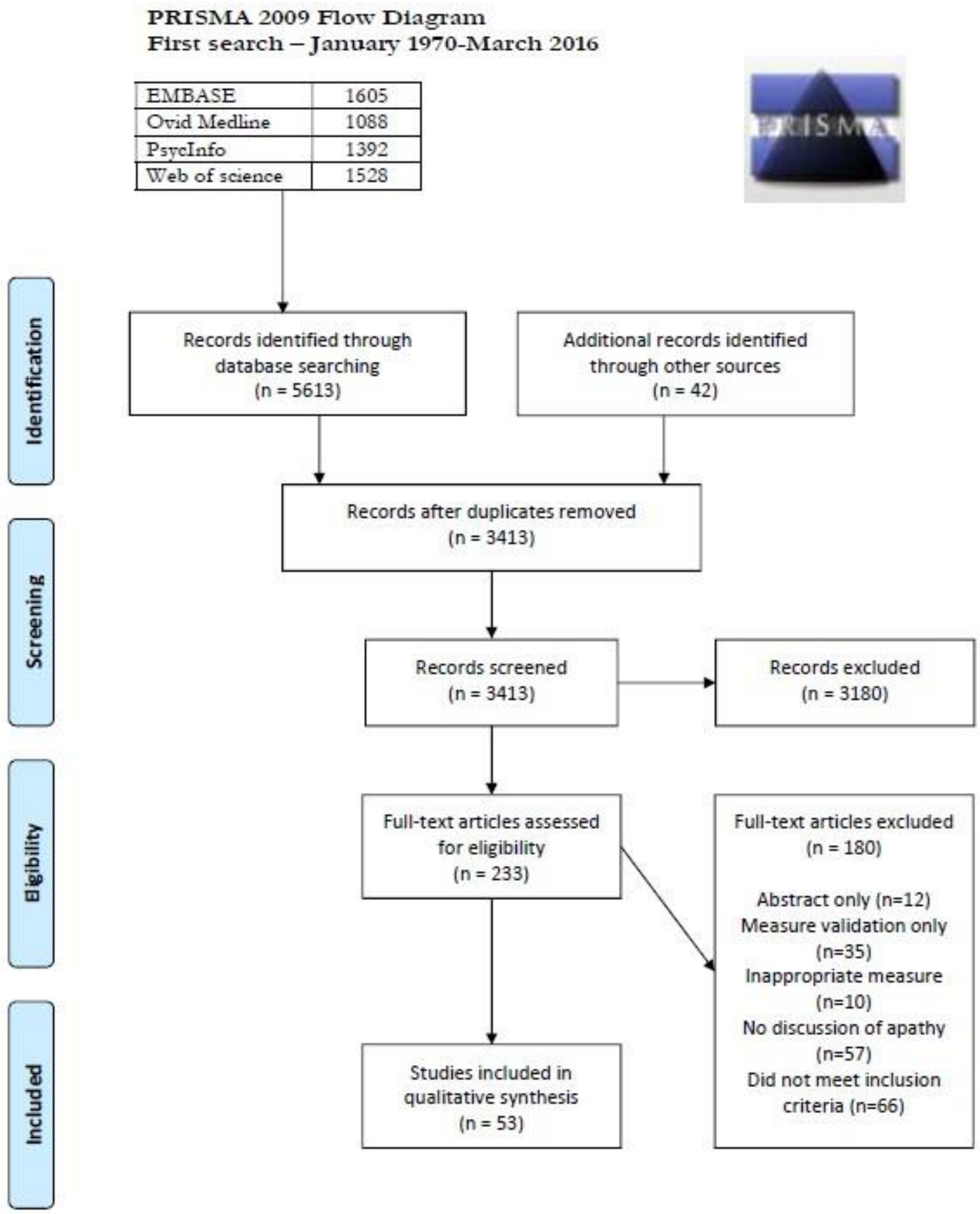
Date: January 1970-March 2016 (second search covered January 2016-September 2019) Language: All (translation help from colleagues and translation programmes) Study design: Searches were not limited by design Participants: Studies were not limited by participant type or age; no included studies were with participants <18 years
Searches included a combination of the following search terms: 1. Apath*.mp OR Apathy/ (MESH) AND 2. Huntington* disease OR Huntington disease/ (MESH) OR Huntington* chorea OR 3. Parkinson* disease (MESH) OR 4. Dementia (MESH) OR 5. stroke (MESH) OR 6. neurops* (MESH) OR 7. neurodeg* (MESH) OR 8. Depression (MESH) AND 9. definition OR 10. concept OR 11. scale OR 12. tool OR 13. psychometric OR 14. measur*

2.3.2 Inclusion, exclusion, and selection criteria:

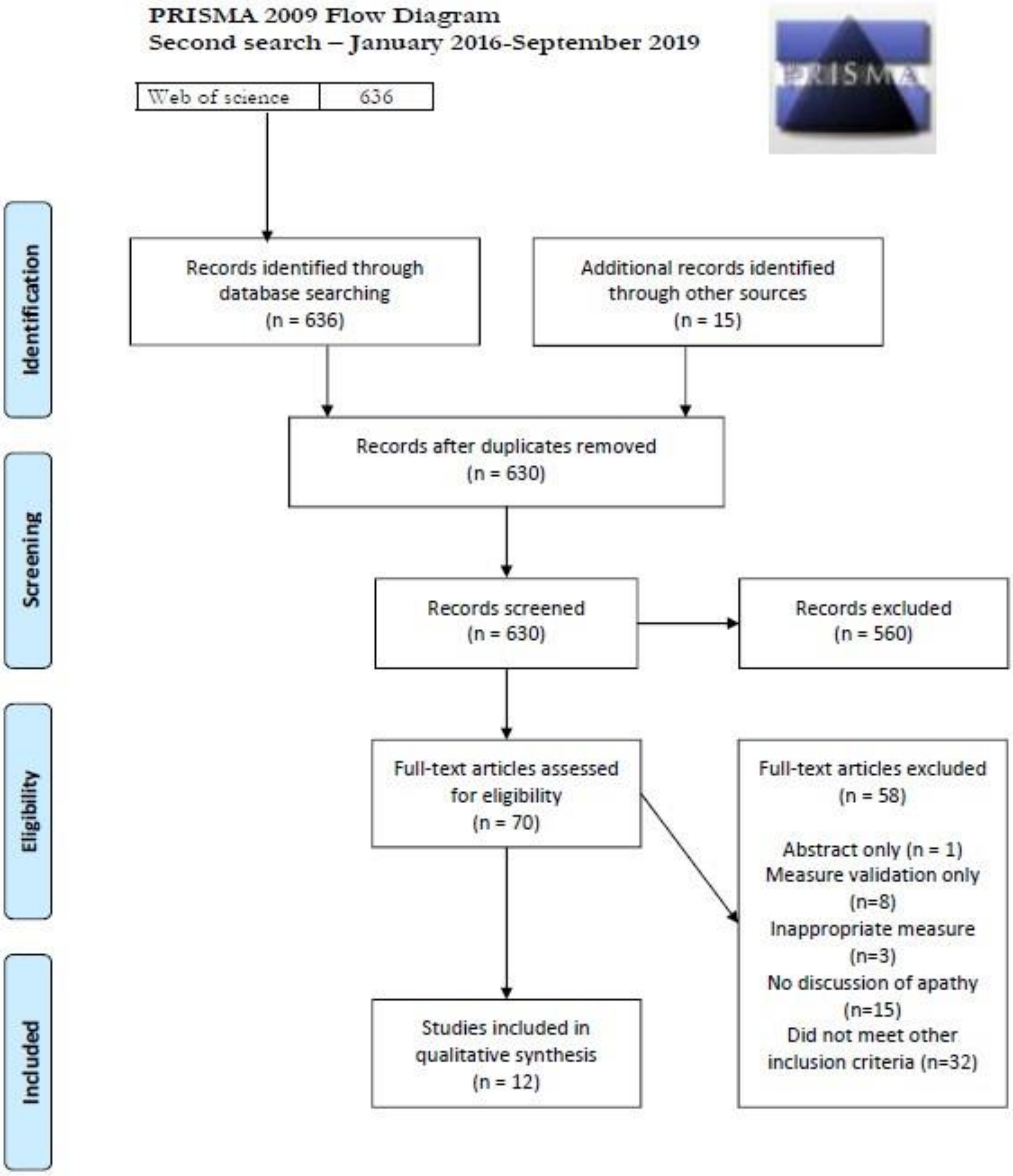
Papers that defined apathy, explored potential diagnostic criteria of apathy, or presented the development of a novel measure of apathy were included. All study types were checked against these inclusion criteria for inclusion. No studies were excluded on the grounds of methodology. Papers that verified the accuracy of an existing measure without presenting a novel measure of apathy were excluded. Brief or shortened versions of existing measures were not included (e.g. Radakovic et al., 2019, validation of brief-Dimensional Apathy Scale). Any paper that used a previous author's definition of apathy and did not present a novel measure was excluded.

Author one reviewed all titles and abstracts to assess their relevance for inclusion. Full-text papers were retrieved for all papers that remained. The first author then assessed these full texts for eligibility. Author two cross-checked 20% of the results during both stages. The results of these searches are shown in the two PRISMA flow diagrams, below, one for the first search covering the period January 1970-March 2016 and the second covering the period January 2016-September 2019. A slight overlap of the searches ensured that all papers from 2016 were included in the search.

2.3.2.1 PRISMA flow diagram for first literature search – conducted March 2016:



2.3.2.2 PRISMA flow diagram for second literature search – September 2019:



2.3.3 Data extraction, assessment, and analysis:

To answer the questions posed in this review, included papers needed to show how they conceptualised, defined, and measured apathy, and how their conceptualisation of apathy has been developed. These data are not routinely extracted or focussed on by existing systematic review quality assessment and data extraction tools. Additionally, the issue of bias (e.g. related to study design or analysis conduct) is often an important consideration in systematic reviews. Here, however, bias is not considered as important as identifying the concept of apathy that each paper develops. The Hawker disparate data tool (Hawker, Payne, Kerr, Hardey, & Powell, 2002) was considered as it is specifically designed to accommodate a variety of methodologies and paradigms in one review. Data extraction was initially trialled using an adapted version of the Hawker disparate data tool. After review, the Hawker tool was deemed not appropriate for this review, however, as it focussed on quality assessment and methodological rigour, two elements that are not useful when assessing a paper's conceptualisation of apathy.

As no appropriate validated data extraction tool was available, author one extracted data using a Word document that focussed on extracting the most useful data relevant to the review. These data included standard identifying information such as author, title, year, and publication. To answer the review questions, data extraction also included, where possible:

- Definition of apathy;
- Concept of apathy (see below);
- Type of paper;
    - Measure: paper that develops a novel measure of apathy;
    - Discussion: paper that discusses apathy conceptually, sometimes referencing a specific illness/injury;
    - Review: paper that reviews either the concept of apathy or the available measures of apathy in a systematic manner (although not necessarily a systematic review);

The concept of apathy that each paper demonstrates is of key importance. To establish how each paper conceptualised apathy was often difficult as most authors did not explicitly state what they thought apathy is and how it can be identified. Each paper was therefore closely examined to identify first how the paper defined apathy (usually there was at least a small section in the introduction with this explanation), and then to see if any other symptoms or elements of apathy were mentioned later in the paper that were not included in the initial definition (often in the discussion section). These conceptualisations are shown in table 1.

The exact wording from the paper was used where possible to reduce the amount of interpretation. Some change of wording was necessary in places, however, to ensure the conceptualisation presented made sense.

It is important to briefly note the difference between a definition and a concept of apathy. A definition of apathy is limited to how the word itself is being interpreted. Apathy may be defined, for instance, as a loss of motivation. A concept of apathy differs as this may include symptoms or behaviours associated with the definition of apathy presented. A concept is therefore a more cohesive, inclusive analysis of apathy whereas a definition of apathy is limited to how authors describe the word itself.

2.4 Results:

The first search identified 5613 studies, 53 of which met inclusion criteria. The second search identified a further 636 papers that were published between March 2016-September 2019, 12 of which met inclusion criteria. A total of 65 papers were identified across the two searches. Two detailed PRISMA flow diagrams for both searches are included in the appendices. Table 1 (in the appendices) outlines the papers. Although the inclusion of 65 papers is relatively large for a systematic review, the remit of the review was kept purposefully broad to include definitions and conceptual discussion from a diverse group of papers. This has enabled the review to discuss several issues related to the conceptual exploration of apathy, organised under the three questions posed by this review.

All papers were published in psychiatric, neuroscientific, or general academic journals. The main disorders associated with apathy were PD and various forms of dementia (AD, FTD, bvFTD, dementia). Other disorders mentioned less frequently were HD, region-specific stroke, schizophrenia, TBI, depression, PSP, and MCI. A few disorders, such as corticobasal degeneration, were mentioned by one or two papers. All papers were directly linked to clinical work, either through empirical data taken from patients or a theory was developed in the paper and linked to a clinical population.

2.4.1 How is apathy defined and conceptually discussed in Huntington's disease and other neuropsychiatric conditions?

This section will focus on:

- Describing four key definitions and conceptualisations of apathy; and
- Outlining three sets of diagnostic criteria for apathy.

2.4.2 Key definitions of apathy:

Most of the 65 papers either did not discuss apathy at all or presented only a very limited definition and discussion. It was common for papers to discuss apathy without offering a clear definition, particularly papers that developed a measure that included apathy as a sub-scale (e.g. #45, the development of the KBCI). Thirty-seven of the 65 papers did not define or conceptualise apathy beyond using dictionary definitions or quoting one other authors' work. Thirteen papers discussed apathy in a more detailed manner, quoting multiple definitions from multiple authors. These thirteen, however, did not further elaborate or discuss the complexities of apathy. Some papers (#26, #30, #43, #47) did seek to explore the nosology of apathy, but none could outline a clear, cohesive nosological position of apathy that was well evidenced. Eight papers (Marin, 1990 and 1991; Levy and Dubois, 2006; Arnould et al., 2013; Radakovic and Abrahams, 2014, Radakovic et al., 2015, Radakovic et al., 2017, and Radakovic and Abrahams, 2018) sought to define and conceptualise apathy and are discussed in detail below.

Table 1 (in the appendices) outlines all 65 papers included in the review, providing a brief description and, where possible, a discussion of the definition and conceptualisation of apathy that the paper presents. Table 2 (in the appendices) outlines the identified diagnostic criteria for apathy, and table 3 (in the appendices) outlines the 14 measures of apathy.

Marin's concept of apathy focusses on describing a clinically useful interpretation of apathy that relies heavily on identifying psychological components of apathy and differentiating between syndromic and symptomatic apathy. Levy and Dubois (2006) take a different approach to Marin, favouring a neurological interpretation of apathy and characterising it as a behavioural syndrome that can be split into three subtypes, all of which are correlated with dysfunction in specific parts of the brain. Arnould et al. (2013) sit somewhere between the two, offering an assessment of apathy that draws on a range of elements in a multidimensional model to conceptualise apathy. Radakovic and Abraham (2014), Radakovic et al. (2015), Radakovic et al. (2017), and Radakovic and Abrahams (2018) present a novel, multi-dimensional measure of apathy, the Dimensional Apathy Scale (DAS), that does not use a single sum-score (as many measures do) and later develop this into a Dimensional Apathy Framework (DAF) that takes Stuss et al.'s (2011) model of executive functioning and adapts it for apathy.

2.4.2.1 Marin (1990, 1991):

Marin focusses on three points in his definition and conceptualisation of apathy. Firstly, that apathy syndrome is a primary lack of motivation. Secondly, that apathy syndrome and apathy as a symptom are distinct clinical problems. Thirdly, that there are key features to apathetic patients that are important to note.

Marin (1990, 1991) focusses on defining apathy from a clinical perspective and providing a definition that is logical and usable for clinicians. He observes that the conventional, dictionary definition describes apathy as an "absence or lack of feeling, emotions, interest, or concern" (1990, p22). He notes that this definition is not useful in a clinical setting as it lacks specificity and clinical cases are more complex than this suggests. Patients with depression, for instance, often profess a lack of motivation and are subsequently described as apathetic, although the presence of emotional pain would seem to be incompatible with that label. Similarly, patients with frontal lobe injuries often lack interest but also display euphoria. In these cases, Marin notes that a lack of motivation is the key driver in describing these patients as apathetic, even though they show a lot of emotion.

Marin therefore posits that a lack of motivation is the primary component of apathy. Amotivation, however, is present in many clinical disorders. To separate apathy from other states where amotivation may be present (for example abulia, despair, or delirium), Marin suggests that an apathy syndrome should be considered a primary impairment of motivation in "patients whose lack of motivation is not attributable to a diminished lack of consciousness, an intellectual deficit, or emotional distress" (1990, p22). Apathy syndrome therefore describes a lack of motivation that is not accompanied by a behavioural, cognitive, or emotional problem. If a lack of motivation is present as well as any of these clinical problems, Marin describes apathy as a symptom or feature of another syndrome or disorder. Marin draws a helpful conceptual parallel here to patients with language difficulty following brain injury. Aphasia is only diagnosed if a patient's language is primarily affected by the brain injury itself, and not by diminished consciousness, attention or intellect. For Marin, the difference between apathy syndrome and apathy as a symptom inhabits a similar clinical definition.

To further explicate the difference between apathy syndrome and apathy as a symptom, Marin gives clinical examples. A patient with schizophrenia may present with entirely negative symptoms, including loss of interest, flat affect, and reduced engagement, and be therefore characterised as apathetic. A depressed patient may show similar negative symptoms, but also be in significant emotional distress, in part due to the negative symptoms.

In these cases, the patient with schizophrenia could likely be described as displaying apathy syndrome since the apathy seems to be a primary motivational impairment without obvious cause, while the patient with depression would be more likely to be experiencing apathy as a symptom of depression as their emotional distress is likely causing a loss of motivation. Marin surmises this, stating:

"The syndrome of apathy is defined as primary absence of motivation, that is, lack of motivation not attributable to disturbance of intellect, emotion, or level of consciousness. If loss of motivation is attributable to disturbance of intellect, emotion, or level of consciousness, apathy is considered a symptom." (p244, 1991)

In Marin's view, the nosological position of apathy as either a symptom or a syndrome is predicated on whether the clinician can attribute the motivational loss to a cognitive, emotional, or behavioural cause. If this is clear, apathy is likely occurring as a symptom, and if not, likely a syndrome. Marin's intention here is to clarify how and when apathy occurs and give clinicians flexible guidance pertaining to the identification and nosological positioning of apathy.

In this section, Marin also describes the likelihood of apathy being a "disease entity itself" as "doubtful" (p244, 1991). This is an important statement on his part and is something he reinforces with his careful use of terminology. Instead of describing apathy as primary or secondary, and perhaps implying that apathy must be a separate disease entity, Marin refers to apathy as a syndrome or symptom. Marin uses the term primary to describe motivational impairment in apathy syndrome. By this, he means that it is the motivational impairment that is primary, not the presence of apathy itself. When apathy is present as a symptom, the motivational impairment is secondary to a behavioural, cognitive, or emotional problem.

In outlining apathy's presentation and features, Marin explores three avenues. Marin takes a broad look at apathy's relationship with individual personality. In a similar way that a person can be depressed without suffering from the medical condition depression, Marin describes apathy as a personality feature present in all but to differing degrees. It is normal to be apathetic about some things, to demonstrate selective apathy, and merely reflects a person's disinterest in some activities. Marin sees this as unproblematic if the individual remains normally motivated in other areas of their life (much as a temporary sadness or depressive moment can be a normal part of one's personality). In individuals whose adult personality lends itself more towards disinterest and apathy, this may show itself either in limited aspects of their psychosocial functioning or in a more pervasive manner. Marin posits here that a level of apathy sufficient to pervasively interfere with psychosocial functioning may be indicative of a diagnosable personality disorder, although points out that it is more likely that a personality disorder would be recognised first, with apathy a secondary symptom.

Marin also considers the role of perceived environmental rewards and their inevitable decrease as part of the normal ageing process. Any major changes in a person's functioning, due to ageing or biological events, may lead to a perceived reduction in environmental rewards which in turn impairs motivation. Marin gives the example of enforced stays in hospital for patients with psychiatric problems as being a possible cause of apathy. Less drastically, change or loss of societal role induced by retirement or divorce can cause an apathetic reaction. The normal transition from one stage of life to another can induce confusion and reduce perceived environmental rewards, impairing motivation and causing apathy. Here, as well, Marin's description of the link between motivation and emotion is key in recognising apathy. Referring to his conventional definition of apathy, Marin points out that a lack of emotion characterises apathy. When considering a diagnosis of apathy, however, it is not just the level of emotion that is important. The relationship between a person's "intensity, persistence, or fixity" (1990, p245) of emotional responsivity is key in identifying a lack of motivation. Marin states that a person's emotional responsivity provides clinicians with information about how motivated they are to respond to environmental events. In other words, a person's flat emotional response to situations can be an indicator of a lack of motivation, which in turn demonstrates the presence of apathy.

Lastly, Marin considers the role and features of apathy in various clinical disorders, including psychotic and affective disorders, organic disorders such as dementia and frontal lobe injury, and drug-induced apathy. Marin presents a quotation from Raskin and Sathananthan (1979) that points out that apathy may come from multiple sources and change the presentation of the person in multiple ways. Particularly in the elderly and people with schizophrenia, people may demonstrate apathy secondary to depression but also related to their perceived reduction in socioenvironmental rewards. The two are not necessarily part of the same apathy symptom or syndrome, and indeed could be a symptom and a syndrome of apathy co-occurring.

2.4.2.2 Levy and Dubois (2006):

Levy and Dubois (2006) consider a "novel perspective" (p916) of apathy, describing it as a syndrome consisting of clinical signs that are indicative of neurological damage to the prefrontal-cortex (PFC)-basal ganglia circuits. Levy and Dubois present apathy as a neurological problem that, if better understood, would provide better understanding of the PFC-basal ganglia circuitry. Levy and Dubois use neurological language to explicitly move away from Marin's clinical, psychological description of apathy as a lack of motivation. They see this as an overly subjective way of inferring a person's mental state from their behavioural state:

"Apathy is not defined here as the clinical consequence of a 'lack of motivation' – a rather blurred and inhomogeneous psychological concept – but as an observable behavioural syndrome consisting in a quantitative reduction of voluntary (or goal-directed) behaviours." (p916)

It is important to note that Levy and Dubois look at apathy from a different clinical field, and therefore purposefully adopt a different approach to defining and conceptualising apathy to Marin's. They do not necessarily disagree with Marin's concept of apathy, but instead adopt an approach whose underlying assumptions about the mind and the brain clash with Marin's. Where Marin focusses on identifying clinical features and prioritising the psychological implications of a lack of motivation, Levy and Dubois focus on identifying apathy as a neurological problem. They see apathy as a behavioural state that is indicative of neurological dysfunction and eschew the psychological as fundamentally irrelevant. Their concept of apathy therefore seeks to identify apathy as an objective, observable, quantitatively measurable reduction in voluntary behaviour. They explicitly state that an apathetic syndrome should be "objectively measurable" (p916), something they view as not possible using Marin's flexible and subjective interpretation of an apathy symptom and syndrome. Levy and Dubois reinforce these ideas and expand their definition of apathy, including parts of an explanatory model of motivation and goal-directed behaviour (GDB) by Brown and Pluck (2000):

"…we propose to define apathy as the quantitative reduction of self-generated voluntary and purposeful behaviours. It is therefore observable and can be quantified… apathy is a pathology of voluntary goal-directed behaviour and the underlying mechanisms responsible for apathy may be seen as dysfunctions occurring at the level of elaboration, execution, and control of GDB." p916

Levy and Dubois present data that show a link between apathy and a dysfunction of the PFC-basal ganglia axis, which they describe as a key component in the "generation and control of self-generated purposeful behaviour" (p917). As the PFC-basal ganglia circuits are heavily involved in GDB (Brown and Pluck, 2000), and Levy and Dubois describe apathy as a pathology of GDB, they describe disruption at different stages of the GDB process and thus three distinct types of apathy in terms of different locations of lesions: emotional-affective, related to orbital-medial PFC lesions; cognitive, related to the lateral PFC; and auto-activation, related to basal ganglia lesions. They also specify how the three types of apathy can impact patients. Firstly, apathy related to emotional-affective processing can reduce

GDB due to a failure to identify emotional signals and planned behaviour. Patients with this deficit find it difficult to demonstrate positive or negative emotion and are likely to display a flattened affect. Secondly, disruption of cognitive processing can reduce GDB due to an inability to expand on thoughts and form a detailed plan of action. Patients with disrupted cognitive processing therefore find it difficult to develop ideas about what to do. Lastly, auto-activation deficit is the most significant sub-type of apathy, whereby any plan of action is near impossible to initiate or complete. Patients with this deficit find it very difficult to achieve anything or understand their own desires or motivation.

As Levy and Dubois acknowledge, the proposition of different 'types' of apathy is suggested by Stuss et al. (2002) who propose that damage to five frontal and subcortical circuits produces different types of apathy. While Stuss et al. (2002) identify a broad range of circuits across the frontal cortex as producing different types of apathy, Levy and Dubois offer a narrower focus, describing how damage to the PFC-basal ganglia circuits may cause apathy sub-types. According to Levy and Dubois, signals coming from basal ganglia sub-regions that normally contribute to effective decision-making are diminished due to damage in the basal ganglia. This diminished signal is then not sufficient to promote the decision-making part of GDB in the PFC and any behaviour is effectively stalled. If damage in the basal ganglia or PFC is sufficient, no signal and therefore no behaviour may be initiated or maintained. Hence, the degree or amount of damage to this circuitry may create different sub-types of apathy. They describe apathy as secondary to dysfunctions of the PFC and basal ganglia, which causes a diminishment or stopping of signals through these areas that are key to inducing or maintaining GDB.

2.4.2.3 Arnould et al. (2013):

Arnould et al. (2013) describe apathy specifically in traumatic brain injury (TBI) and outline a multidimensional, four-factor model of apathy. Their aim was to develop a model of apathy that had the potential to improve the clinical assessment and treatment of people who suffer apathy following a TBI. They suggest that revealing the underlying mechanisms of symptoms is key in accurately conceptualising and diagnosing apathy. Arnould et al. acknowledge that previous literature has sought to explore underlying mechanisms of apathy symptoms (particularly Levy and Dubois) but states that insufficient weight was given to this exploration, with previous authors instead focussing on identifying behavioural or psychological symptoms. They adopt an approach somewhere between Levy and Dubois' entirely neurobehavioural interpretation of apathy that relies heavily on underlying neurological mechanisms and Marin's reliance on the psychological features and presentation of apathy.

Arnould et al.'s (2013) model seeks to identify these underlying psychological mechanisms and processes that are present in each dimension of apathy. They describe apathy as comprised of four dimensions; cognitive, motivational, affective, and a dimension that is not described by other authors as a key dimension, personal identity. The cognitive dimension describes a series of problems related to cognition such as difficulty in resolving complex situations which may subsequently lead to a reduction in the desire to set goals, and a decrease in ability to assess goal-directed behaviour leading to a reduction in goal-setting, among other cognitive problems. The motivational dimension relates to an inability to anticipate pleasure or seek sensation, which also results in a lack of positive reinforcement. Arnould et al. suggest that a lack of ability to anticipate reward leads to a reduction in motivation and increased apathy. The affective dimension relates predominantly to the presence of negative mood, which increases patients' perception of pain, may increase perceived difficulty in task initiation and completion, and reduces the ability to experience pleasurable emotions. The personal identity dimension postulates that an individual's pre-existing beliefs and norms play a large role in how and when apathy is experienced, and how an individual may react to the impact of the preceding dimensions. Arnould et al. suggest that self-esteem may be closely linked to apathy; i.e. low self-esteem may reduce perceived risk-taking behaviour and thereby reduce the likelihood of experiencing positive reinforcement or emotions related to reward. Arnould et al. present this dimension as a unique feature of their model, although it does resemble Marin's exploration of individual personality and reduction in perceived socioenvironmental rewards.

2.4.2.4 Radakovic and Abraham (2014), Radakovic et al. (2015), Radakovic et al. (2017), and Radakovic and Abrahams (2018):

Across four papers, Radakovic and other authors present a multi-dimensional measure of apathy and subsequently develop the Dimensional Apathy Framework (DAF). Much of this work builds on the descriptions and conceptualisations of apathy above, particularly those of Marin (1991) and Levy and Dubois (2006). This is not an unusual approach, and indeed is broadly similar to Arnould et al.'s (2013) model, but Radakovic et al. come to a slightly different conclusion to others who have taken this approach. Radakovic notes that most of the existing measures of apathy recognise the potential multi-dimensional aspect of apathy but then present a global sum-score that means apathy must be considered a singular concept. This has also been pointed out in Clarke et al.'s (2011) review of the field, which Radakovic uses as appropriate justification for developing a measure that maintains the separation and distinction of three dimensions of apathy.

The Dimensional Apathy Scale (DAS) (see table 3 in the appendices for details) is based on Levy and Dubois' (2006) triadic apathy subtypes, emotional-affective, auto-activation, and cognitive apathy. After factor analysis of the data and multiple changes to the items included, a three-factor model was found to fit the measure best, with eight items per subscale. The three factors changed from Levy and Dubois' (2006) and were executive, cognitive/behavioural initiation, and emotional. The executive and cognitive/ behavioural initiation factors mapped respectively onto Levy and Dubois' (2006) cognitive apathy and auto-activation apathy, while the emotional subtype did not match the description of emotional-affective apathy and was therefore changed. Radakovic et al. defined the emotional subtype of apathy as an integration of emotional behaviours, while Levy and Dubois (2006) described emotional-affective apathy as a reduction in GDB related to emotion processing, a different process to Radakovic's emotional apathy.

Where Levy and Dubois (2006) consider apathy as secondary to dysfunctions of the PFC and basal ganglia, and therefore adopt a neurological position in assessing apathy, Radakovic et al. adopt a position somewhere between Marin's focus on psychological and behavioural interpretation of apathy and Levy and Dubois' focus on neural mechanisms. Again, this is similar to Arnould et al. (2013) but is perhaps better explained and developed across a series of papers. Radakovic and Abrahams (2018) develop a novel framework of apathy, the DAF, which is an extension of the theory behind the DAS that emphasises their multi-dimensional conceptualisation of apathy. The DAF consists of three subtypes of apathy: initiation apathy, characterised by impaired self-generation of thoughts and decreased spontaneity; executive apathy, characterised by impairment in executive functions of planning and inability to manage goals; and emotional apathy, characterised by affective flattening, indifference, and emotional blunting. Overarching all of these subtypes, they propose that self-awareness mediates between and among them. This inclusion of self-awareness (or the lack of) as key to how people experience apathy is a particularly useful and novel element. It allows the framework to show that a person can be more or less aware of how their apathy affects different parts of their life and begins to bring the possibility of discussing people's experience of apathy into the mainstream discussion of apathy.

2.4.3 Diagnostic criteria:

Another important part of the literature's definition and conceptual discussion of apathy is the development of diagnostic criteria. Alongside defining apathy, several authors identified the need for consistent, agreed-upon diagnostic criteria that could be used clinically to identify potentially apathetic patients. Four diagnostic criteria for apathy have been developed by Marin (1991), Starkstein (2001), Robert et al. (2009), and Robert et al. (2018). The latest criteria from Robert et al. (2018) are an updating of the 2009 criterion by the same authors that acknowledges that research has significantly developed the concept of apathy in the intervening years. The criteria follow a similar pattern to the definitions discussed above in that Marin proposed a set of criteria and this was then adapted by Starkstein et al. (2001) and then by Robert et al. in 2009 and 2018. Mulin et al. (2011) also present a set of diagnostic criteria but these are a slightly amended version of Robert et al.'s (2009) criteria and are therefore not considered separately. An abridged version of each of the criteria is set out in table 2 (in the appendices due to length).

It is interesting to note that each set of criteria builds on the previous with relatively little change. Moving from Marin's criteria to Robert et al.'s, perhaps one of the major changes is the adoption of a specific time-period for considering a person's potentially apathetic behaviour. Marin leaves this relatively undefined whereas Robert et al. lay out a reasonably short period of four weeks to observe change. This is perhaps reflective of the idea that people's preferences and interests will naturally change over particularly extended periods, whereas someone experiencing apathy is more likely to change their preferences or display disinterest relatively quickly. Given much of Robert et al.'s (2009) work considers apathy in Alzheimer's disease and other neuropsychiatric conditions, four weeks seems like a short space of time to recognise major changes in behaviour.

Another key difference is the inclusion of a reasonably broad criteria in Robert et al.'s (2009) definition that includes identification of "clinically significant impairment". This in itself is a broad idea but emphasises the idea that apathy causes impairment in a person's life and that this is a key component in diagnosing apathy. Marin's criteria do not necessarily imply that apathy itself causes impairment but instead focusses on a lack of motivation as the key driver in diagnosing apathy. This is an important shift as Robert et al. (2009) also include diminished motivation as a criterion but would only consider diagnosing apathy if that motivational diminishment induces clinical impairment. Robert et al.'s (2018) later work again builds on the earlier criteria and represents perhaps the biggest shift in thinking. Criterion A shifts the focus of apathy away from discussion of motivation and instead talks purely about a reduction in goal-directed activity. This language is carefully chosen to avoid defining apathy as a loss of motivation as Robert et al. (2018) note that this broad definition has been criticised, with critics pointing out that motivation is a psychological interpretation of observable behavioural and therefore overly subjective (Levy and Dubois, 2006). Instead of motivation, apathy has become a quantitative reduction in goal-directed activity, a phrase that seems to encompass both GDB and goal-directed cognition (GDC). Criterion B1 now elaborates on both GDB and GDC together instead of separating them across two criteria. More importance is also given to a loss of emotion; five types of emotional loss are now considered a key part of apathy instead of the previous two. Social interaction is also considered as more important, now being a separate criterion, whereas in the 2009 criteria, it was only implied as part of B1.

2.4.4 How is apathy measured in HD and other neuropsychiatric conditions?

There are 14 novel apathy measures that met this review's inclusion criteria, comprising nine full-scales and five sub-scales. The 14 measures are summarised in Table 3 (in the appendices). There are important differences in how the measures approach the measurement of apathy. This section will focus on identifying difference in content, structure, and scoring.

2.4.4.1 Content:

The measures include items that assess a variety of domains or dimensions of apathy. Broadly, assessing a person's motivation by asking questions related to their behaviour, cognition, and emotion (BCE) is interpreted as a direct proxy for assessing apathy. A global sum-score of apathy was a common outcome for the measures apart from the DAS, which maintained a triadic structure of the resulting score. The assessment of motivation, either directly or indirectly, runs throughout the measures. Specifically, the items in the measures assess people's; motivation, level of interest, activities of daily living (ADL), energy, spontaneity, affect, initiative, withdrawal, emotional engagement or responsivity, socialisation, hygiene, amount of speech, and ability to complete tasks. The items assess these dimensions by asking questions about their presence, absence, amount, frequency, and severity. Most measures ask people to think back to the past four weeks when considering their BCE. The Scale for the Assessment of Negative Symptoms (SANS) and Irritability-Apathy Scale (IAS) do not stipulate a timeframe, while the Apathy Motivation Index (AMI) stipulates a shorter two-week period.

As mentioned above, assessment of motivation is common in the measures. Some items relate to motivation directly by using the word motivation in the item, while others will use various proxies to assess motivation. These proxies include asking questions about a person's level of interest in various activities (e.g. hobbies, friends, learning new things), the amount of effort a person puts into an activity, or ability to plan their day. The last item of the Apathy Evaluation Scale-Clinician (AES-C), for instance, poses the relatively simple statement, "S/he has motivation" and is answered with a Likert-type 1-4 scale. The first AES-C item states, "S/he is interested in things". The latter refers mainly to a person's level of interest but could also be construed as assessing a person's motivation through their interest in 'things' – if one is interested in things, one would have to be motivated to initiate and maintain that interest. The Apathy Inventory (IA) consists of three items that assess a person's emotional blunting, loss of initiative, and loss of interest. It could be reasonably argued that all three domains require motivation to overcome these problems and certainly reasonable to describe motivation as key in maintaining initiative and interest.

Assessing general level of interest is also a common item. Again, there are a range of approaches to assessing patient interest. The AES-C, as with a lot of the items it assesses, is relatively simplistic in its assessment. The first item of the AES-C states, "S/he is interested in things", and is scored on a Likert-type 1-4. In addition, items four, five, and nine are related to a person's level of interest in new experiences, new things, and amount of time spent doing things that interest them. The Lille Apathy Rating Scale (LARS) contains nine domains, one of which is 'interest' and contains multiple questions related to interest. The Dementia Apathy Interview and Rating (DAIR) also contains four items that are directly related to interest, and another two that are indirectly related.

Some measures are weighted toward a certain domain, for instance by asking more questions about a person's behaviour than their emotional state. Others are not weighted and ask the same number of questions about each domain they focus on. As described above, however, this is often not clear cut as some items refer, at least linguistically, to multiple domains. Items that ask questions about a person's level of interest can also be interpreted as asking about a person's motivation. A question about a person's level of emotional engagement could also be assessing a person's behaviour, i.e. if they demonstrate emotion. These differences are further complicated by the scoring system each measure adopts – some measures give a global apathy score, while others give a score for each domain measured and eschew a global score. No measures discuss the overlapping of domains such as interest and motivation or emotion and behaviour. When discussing the development of measures, various statistical techniques often show how many factors the measure loads onto, but these are presented as distinct factors with no overlap.

The level of detail and complexity of the items varies greatly across the measures. Level of detail and item complexity were compared by looking at the number of items included, how many domains the items looked at, and complexity of the design of the item itself (i.e. how the item is presented and what it includes). As expected, the full scales tended to be more detailed than the sub-scales by including more items and assessing a wider range of domains. The LARS, for instance, starts with a question about a person's everyday productivity which is rated by an open question as well as two further Likert-type questions. This is a complex design for the assessment of one domain and the LARS continues this complex, multi-faceted, well-designed format throughout each of the nine domains it assesses and uses 33 items. The IAS sub-scale includes five items and starts with a similar question about a person's interests in everyday life. The answers, in contrast to the LARS, are a closed set of Likert-type options ranging from interested to not interested. This format stays the same for the other four items in the IAS, and each item arguably refers to a person's interest.

There seemed to be an inverse relationship between item complexity and number of items per domain. Total number of items per measure did not seem to have any relation to complexity of item. When measures were examined more closely to look at how many dimensions they measured and how complex the items were, it was noted that there did seem to be a relationship. The measures that had fewer items per dimension, such as the IA, laid more importance on each item and more importance on the experience and subjective judgement of the assessor. The measures that include multiple items per dimension could afford to ask simpler questions that required less subjective judgement, but each item did not delve as deeply into the dimension.

The IA and DAS demonstrate this relationship between complexity and number of items well. The IA has three items, the least of all, and is relatively simple in style, asking for a score out of four (no problem-major problem) for three domains (emotional blunting, loss of initiative, and loss of interest), one item per domain. The domains, however, are relatively complex in that they ask questions that involve subjective clinical judgement about multiple aspects of a person's BCE. The first domain, emotional blunting, simply asks for an assessment based on the questions, "Does the patient show affection?" and, "Does s/he show emotions?". These are complicated questions that a clinician must then reduce to a score of out four. This measure was designed to be administered by a clinician to a patient, and necessitates the clinician having prior experience with the patient.

The DAS, presumably because it was designed to be self-administered, is much simpler. It includes 24 items, all of which are stylistically similar simple statements that are answered on a Likert-type scale from 1-4, almost always-hardly ever. The items relate to three domains; executive function, emotion, and behaviour/cognitive initiation, eight items per domain. Each item requires little assessment or judgement, but instead focusses on a small event or activity. When combined, the eight items give a fuller picture of that domain than the individual items can. The AMI is a similar structure to the DAS (the DAS and LARS were used in the development of the AMI), posing relatively simple statements that people are asked to report are completely true or completely untrue about them. The AMI also uses a triadic structure, referring to behavioural activation, emotional sensitivity, and social motivation as the three factors influencing a person's apathy.

2.4.4.2 Structure:

A key area of difference concerns who administers the measures and to whom they are meant to be administered. Some measures have different variants designed for different situations, but most have only one version. The AES and IA have three variants; a clinician talking to the patient, a clinician talking to a person who knows the patient well (a carer, spouse, or close relative), and, lastly, self-report for the AES and a form of self-report, with a clinician present, for the IA. The Apathy Scale (AS), Key Behaviours Change Inventory (KBCI), Structured Clinical Interview for Apathy (SCIA), Neuropsychiatric Inventory (NPI), Frontal Systems Behaviour Scale (FrSBE), SANS, IAS, and LARS are administered by a clinician talking to the patient. The DAIR is different, administered by a clinician talking to a carer, while the Apathy in Dementia Nursing Home (APADEM-NH) is administered by a clinician talking to a professional caregiver. Given the APADEM-NH is designed to be used only in a nursing home, it is likely that the professional caregiver is a member of the nursing home staff, although this is not specifically stated in the measure. The APADEM-NH also does not state if the term professional caregiver precludes family members. The DAS is a self-report measure while the AMI is clinician-administered; both were developed in healthy populations, although the DAS has since been validated in clinical populations and the AMI states that it can be used in clinical populations (although future validation is needed to prove this).

As well as a variety of administration techniques, a variety and combination of question styles are used. Some of the measures, such as the AES-C and LARS, begin with a reasonably open-ended question designed to assess a patient's activities of daily living and ability to recall and talk about themselves. The AES-C states that the interview should begin with a description of the patient's interests, activities, and daily routine, while the opening question in the LARS is "What do you do during the day? Tell me about your day-to-day life.". From these opening statements, a clinician must then make judgements about the patient's interest, motivation, and ability. Again, however, there is a marked difference between the assessment of this first question. The AES-C uses this open-ended question style to direct the clinician to answer the 18 items in the measure on a Likert-like scale from "Not at all characteristic" to "A lot characteristic". The LARS uses this open-ended beginning as a direct way to assess a person's activities of daily living, scoring by the time taken to reply and the number of activities mentioned. In addition, the LARS is scored on a scale that goes from "no reply" (a score of +2) through to "immediate reply" (a score of -2) for time taken to reply, and from "none" (+2) to "detailed organisation of a typical day…" (-2).

Other measures do not include an open-ended start, instead going straight into asking direct, closed questions (although it should be noted that the AES-C also follows this pattern but prefaced with an open-ended question). The self-report DAS poses closed statements that can be answered with one of four responses; "Almost always", "Often", "Occasionally", and "Hardly Ever". The NPI poses relative questions directly to the clinician administering the test such as, "Does the patient seem less spontaneous less active than usual?". These questions are answered dichotomously either "Yes" or "No". The IAS also has closed questions with a five-point Likert-type scale response.

The style and tone of question also changes among the measures. Some measures pose positive statements, while others pose negative statements. In the case of questions assessing motivation, this is particularly apparent. The AES-C presents motivation as a positive question, asking if a person is motivated. The DAS presents motivation as a negative, stating "I need a bit of encouragement to get things started". All three of the IA's items, which arguably all indirectly assess motivation, are posed negatively, for instance asking about emotional blunting instead of the more neutral (but open-ended) emotional state.

2.4.4.3 Scoring apathy:

The measures can be split into three groups based on how they score apathy. Firstly, the sub-scales included in this review, the SANS, IAS, KBCI, NPI, and FrSBE, all provide a global score for apathy that is often combined to give an overall score. The SANS, for instance, provides a global apathy score that becomes part of the overall assessment of a patient's negative symptoms.

Secondly, the AES, AS, DAIR, and SCIA provide a global, singular score of apathy. The AES provides limited guidance about interpreting severity; a score of over 42/72 indicates the presence of mild apathy in elderly populations, while a lower score may indicate the presence of mild apathy in younger populations. There is no defined cut-off, rather, researchers and clinicians are encouraged to develop their own norms based on their experience. The AS, being predominantly an adapted version of the AES, adopts similarly flexible guidance. The DAIR states that a higher score indicates more severe apathy but does not suggest or mention any cut-off. The SCIA offers the most structured scoring programme; the questions in the SCIA are based on adapted diagnostic criteria from Starkstein (2001) and the interviewer rates the symptoms as either absent, subclinical, or present. If symptoms are present in criteria A (lack of motivation relative to the patient's previous levels of functioning) and B (related to lack of effort, interest, or concern, and flat affect), and absent in criteria C (extent of social and occupational dysfunction caused by the symptoms of apathy) and D (exclusion of organic causes of behavioural changes other than apathy), then apathy is diagnosed. The SCIA is therefore a test of Starkstein et al.'s (2001) diagnostic criteria.

Finally, the IA, LARS, DAS, and APADEM-NH adopt a multi-dimensional interpretation of apathy by assessing different dimensions related to apathy. The IA assesses three dimensions of apathy (see Table 3) and includes both individual scores for the dimensions as well as a global apathy score (obtained by combining the three sub-scores). The LARS assesses nine dimensions of apathy in 33 items (see Table 3) that are then divided into four dimensional sub-scores and then summed for a global apathy score. Similarly, the AMI is scored on three dimensions and then summed. The DAS is scored in three dimensions (executive, emotive, and behaviour/cognitive initiation) with no global apathy score. The APADEM-NH is scored into three dimensions (see Table 3) and it is unclear if there is also a global score for apathy. The AES is scored in three dimensions relating to Marin's triadic structure of apathy (behavioural, cognitive, and emotional dimensions), and then summed to provide a global score.

2.5 Discussion:

This review systematically explored the conceptual discussion and measurement of apathy in HD and other neurodegenerative and neuropsychiatric disorders. No other systematic reviews exist of this scale in the field. There are other reviews, but most concentrate on one aspect of apathy, such as measurement or symptom clusters. This review is unparalleled in its scope, looking at the concept of apathy from a broad stance and taking into account definitions, conceptualisations, measurement, and diagnostic criteria. In covering all of these aspects, this review has been able to compare the impact of these different issues in apathy to begin to unravel how apathy is viewed. Of particular importance is the recognition that definitions of apathy varied across the literature, something previously recognised, but that the variance in definition was usually because an author took a different perspective on identifying apathy. The difference in approach between Marin, who concentrated on identifying behaviour, emotion, and cognition related to apathy, and Levy and Dubois, who explored potential underlying neurological mechanisms in apathy, is a good example of this difference in approach. These authors did not agree with each other about what apathy constitutes, but that was because they were talking about different concepts of apathy based on separate fields of study and with divergent aims. Therefore, when thinking about the definition, conceptualisation, and measurement of apathy, it is important to note from what perspective one is talking and interrogating one's aim.

At the start of the review, it was relatively unknown how much literature focussed on conceptually defining apathy. Previous reviews suggested that the definition of apathy varied across the literature and that there was a lack of consensus about the definition. It remained unclear, however, exactly what work existed to conceptually define apathy. The literature reviewed here shows that apathy is a complex phenomenon that is conceptually discussed by relatively few authors, but many recognise that it is an unstandardised concept. The authors included in this review gave at least a brief description of apathy, but often relied on a previous author's definition or defined apathy as a combination of behavioural, cognitive, and emotional components without going into detail. There was broad consensus that apathy is syndromic (Lanctot et al., 2017) and more recent explorations of apathy focussed on the multi-dimensional structure of apathy rather than relying solely on describing apathy as a loss of motivation, such as Radakovic and Abraham's (2018) paper developing a Dimensional Apathy Framework.

There are conceptual similarities about how apathy is discussed across the papers. A large portion of papers include a loss or lack of motivation as being key to apathy. A reduction in or impairment of goal-directed behaviour (GDB) was also common, as was using GDB as either a synonym for apathy or a main component of apathy. Other common phrases include disinterest or a lack of interest, emotional blunting, lack of concern, and flat affect. There are certainly common themes running through these terms, and in general, it is reasonable to say that most definitions or conceptualisations of apathy were similar. This is both reassuring and problematic in the context of conceptualising apathy. The similarity in how papers defined apathy mean that, broadly, it is true that authors think of apathy in similar terms. It is clear, however, that this means they do not think about apathy in exactly the same way. There are tens of versions of apathy across the papers in this review. As a consequence of the lack of standardisation of apathy, authors are forced to set out what they think apathy is, based on their experience and reading of the literature, which can be a double-edged sword as this usually introduces yet another definition of apathy. It must also be pointed out that many of the papers not included in this review were rejected precisely because they did not provide a definition or explanation of apathy. Authors therefore have a difficult choice; either present a very broad definition of apathy, such as merely defining it as a loss of motivation, or present their own understanding of apathy, and thereby contribute to the lack of standardisation in the field.

These comments are not criticisms but a demonstration of the difficulty in defining a concept. Apathy in particular is difficult to define in a way that can be standardised, particularly as it is a concept usually defined by loss. Although its etymological beginnings emphasised control and associated apatheia positively, the shift in the meaning of apathy means it is now likely to be associated with a loss or decrease of behavioural, cognitive, and emotional concomitants, and difficult to standardise.

Perhaps because of this conceptual difficulty, most authors do not elaborately define the concept of apathy, instead focussing on measuring apathy and acknowledging that it remains an ill-defined concept. The few authors that do conceptually define apathy (predominantly Marin, Levy and Dubois, Arnould et al., and Radakovic et al.) do so from different perspectives and disciplines and with different aims. It is reasonable to suggest that apathy has not been sufficiently conceptually defined and remains a varied concept across the literature, although recent developments in the acknowledgement of apathy as syndromic and as multi-dimensional are positive steps. Additionally, although three detailed diagnostic criteria exist, none have been adopted by a diagnostic manual and therefore have not become accepted clinical standards for assessing apathy, further emphasising the lack of conceptual clarity when discussing apathy.

In terms of the measurement of apathy, previous reviews focussed on the quality of the measures from a psychometric and methodological perspective. This review set aside those issues, instead focussing on the measures' differences in terms of item inclusion, scoring method, and question style. This comparison showed a large amount of variability in how the measures approach the measurement of apathy. Although there was overlap between measures and they contained items that related to similar dimensions, such as level of motivation or completion of activities of daily living, there were sufficient differences in all the measures to suggest that apathy could reasonably be described as a different construct in each measure. No measure of apathy seems to be preferred or represented well in the literature, with most measures seeming to be used by their proposing authors and relatively few other studies. The adoption of Carrozzino's (2019) clinimetric approach to construct and measure development that prioritises clinical utility and validity over psychometric properties is appealing and may help to alleviate the proliferation of measures. This is also problematic, however, as Carrozzino (2019) concludes that the LARS is the most clinically useful, potentially ignoring the complex language issues that the LARS demonstrates (discussed further below).

2.5.1 Discussion – How is apathy defined and conceptually discussed in Huntington's disease and other neuropsychiatric conditions?

Four main groups of authors have sought to define and conceptualise apathy, all taking different approaches to the task, with different underlying assumptions about what apathy constitutes and how best to identify it. It is important to recognise the main differences between these authors to clarify how apathy is currently viewed.

Marin (1990, 1991) defined apathy as a primary loss of motivation and made a distinction between apathy as a syndrome and as a symptom. Marin treated the problem of how best to identify and conceptualise apathy from a clinical, psychiatric perspective. This meant prioritising the clinical features of apathy, concentrating on its psychological and behavioural impact. To Marin, a person's lack of motivation was key in understanding apathy and identifying this relative lack of motivation was a clinician's job. When defining apathy in his 1991 paper, Marin focusses on how clinicians use the term apathy and compares apathy with other psychiatric disorders by looking at behavioural symptoms and mapping them on to his definition of apathy. He concentrates on symptoms like a lack of interest, flat affect, and dysphoria. By placing apathy within the context of psychiatric disorders, and by discussing those disorders from a psychiatrist's perspective, Marin is conceptualising apathy within the framework of psychiatry.

Levy and Dubois (2006) actively distanced themselves from what they saw as Marin's overly subjective approach that relied on predominantly psychological or behavioural symptoms and a psychiatric viewpoint. Instead, Levy and Dubois concentrated on trying to identify the underlying neurological mechanism(s) of apathy, thereby moving away from the subjective interpretation of behaviour and towards objective identification and measurement of apathy through brain dysfunction or damage. This stance is somewhat problematic as it views psychological symptoms as subjective and therefore not useful while presenting a theory about the underlying mechanisms involved in apathy as objective and therefore a better measurement. It is perhaps a little dismissive to brush psychological symptoms aside as merely subjective while suggesting that identifying neurological mechanisms is entirely objective and without an interpretative element.

This potential problem of Levy and Dubois, their insistence that identifying neurological problems is unproblematic, is emblematic of the difficulty in comparing these two interpretations of apathy. There is no specific disagreement or major rift between Marin's and Levy and Dubois' interpretations of apathy, apart from Levy and Dubois' protestations about the potentially subjective use of motivation as a psychological interpretation of behaviour. Instead, the two have a different starting point. The two interpretations think about the problem in different ways, approach identification of apathy from different perspectives, and sit on different sides of the mind/brain dualism debate. Marin seeks to identify psychological and behavioural elements of apathy and distinguish apathy from other psychiatric disorders. He sees the mind and the brain as essentially the same and has no problem in dealing with potentially subjective interpretations of a person's symptomatology. Levy and Dubois agree that apathy is a clinical issue, they agree that apathy is a psychiatric disorder, but instead of focussing on psychological or behavioural concomitants of apathy, that they view as overly subjective, immediately focus on underlying neurological mechanisms responsible for causing apathy, that they regard as objective. They see the mind and the brain as completely distinct and see no value in identifying psychological symptoms of apathy that reside in the mind as these are too subjective to be of enough import. Identifying objective brain damage or dysfunction is, to them, a more useful and reliable method of identifying apathy. This difference shows a fundamental divergence in thinking between Marin and Levy and Dubois. Both their concepts of apathy are guided by their clinical experience and knowledge, both of which have led them down different paths and different methods of identifying and thinking about apathy. Although they are thinking about the same problem, they are speaking in different languages that, in this early stage of research into apathy, have little overlap. The overlap is an important area, but one that inevitably must come when more is understood about the concept of apathy. It is important to note that neither approach is more or less correct, but significantly divergent. Levy and Dubois are seeking to identify the relationship between neurological impairment and apathy, while Marin seeks to identify what apathy looks like in people's behaviour, emotion, and cognition. Both approaches are valid but represent different assumptions and methodologies that, as yet, are not compatible.

Arnould et al. (2013) take what may be deemed a middle ground here, combining Marin's psychological and behavioural stance with Levy and Dubois' exploration of underlying neurological mechanisms. In a similar fashion to Levy and Dubois, Arnould et al. (2013) also present a dissatisfaction with the current interpretations of apathy. In their opinion, the existing conceptualisations of apathy rely on a "mostly descriptive" (p215) approach to defining apathy. Arnould et al. state that a descriptive approach, as is often taken in the DSM, focusses on identifying a cluster of symptoms that co-vary to produce a diagnosable disorder and therefore also show causation. Arnould et al. suggest that revealing underlying mechanisms of symptoms is key in accurately conceptualising and diagnosing a disorder. They do acknowledge that previous literature has sought to explore underlying mechanisms of apathy symptoms (particularly Levy and Dubois) but states that insufficient weight was given to this exploration. Levy and Dubois' exploration of apathy was predominantly concerned with identifying neurological mechanisms, occasionally using behavioural traits to identify underlying neurology, but perhaps Arnould et al.'s point is that no previous paper has combined all elements, the psychological and behavioural with the underlying mechanism. It seems a little unfair to criticise Levy and Dubois for not combining these fields, since their aim was explicitly to explore the areas of neuropsychology related to apathy. Further, given the relative infancy of the exploration of apathy, it is perhaps a little early to look for total explanation of apathy, combining neurology, cognition, emotion, and behaviour, before any single element is understood.

In contrast to Arnould et al.'s (2013) interesting but perhaps flawed work, Radakovic et al. (2014, 2015, 2017 2018), across four papers, develop a multi-dimensional theory of apathy that is both compelling and well-explained. Many papers still resist this multi-dimensional description of apathy by, for instance, using a global sum-score in a measure of apathy or by describing apathy as characterised by simply a loss of motivation; this is perhaps one of the motivations behind Radakovic and Abrahams (2018) development of the Dimensional Apathy Framework, that serves as an excellent base for future research. Their framework is triadic, relying on initiation, executive, and emotional subtypes of apathy, but also includes a fourth overarching dimension of self-awareness that allows the model to potentially grow, as they state in their article. Self-awareness, as they state, is not a separate factor but can influence all three subtypes of apathy. This overarching factor then presents apathy as a multi-faceted and multi-dimensional syndrome. The important implication here is that apathy can be experienced in different ways by different people, something that previous models of apathy, particularly those that relied on simple explanations of apathy as merely a single-factor loss of motivation, did not show. For clarity, it should also be noted that the

Dimensional Apathy Framework was developed by Radakovic et al. after the development and testing of the Dimensional Apathy Scale (the DAS). The DAS is used in later chapters of this thesis as part of the data collection. Parts of Radakovic et al.'s later framework, the Dimensional Apathy Framework, are useful in exploring the DAS but it is important to remember the order in which these were developed. This is important as one may expect the scale to come out of the framework, but in this case the reverse is true. The scale precedes the framework and does not necessarily reflect all elements of the framework.

2.5.2 Discussion – The concept of apathy:

Apathy appears to be a complex, subjective, damaging symptom or syndrome that occurs in many conditions and this complexity is reflected in the multiple definitions of apathy. The four concepts of apathy presented above are different in their focus and in terms of where they lay emphasis in describing apathy. Marin approached the problem from a purely clinical perspective, attempting to provide a description of how patients may behave and what it is they may experience while suffering from apathy. Levy and Dubois approach apathy from a functional neurophysiological and neurological perspective, prioritising the localisation of apathy rather than the experiential elements. Arnould combines the neurological and psychological, offering a version of apathy that identifies key relevant behaviours that may signify apathy but roots them in possible neurological causes. When viewed together, there are clear differences in how these approaches present different versions of apathy. Marin is interested in the behaviours that signify the presence of apathy, predominantly focussing on identifying a relative lack of motivation. Levy and Dubois focus on identifying neurological correlates that may be identifiable in changed behaviour in a person but are more likely to be identified by brain imaging. In this way, Levy and Dubois are looking to identify precise neurological problems that present as apathy and that future research can therefore alleviate. Arnould et al. suggest a novel four-dimension structure of apathy that includes both identification of behaviours that are suggestive of apathy and potential neurological correlates. Radakovic et al., across multiple papers, present a nuanced, balanced concept of apathy that shares its roots across both neurological mechanisms, psychology, and behavioural traits. The development of the Dimensional Apathy Framework (Radakovic and Abrahams, 2018) is an interesting step in explicitly trying to unify the varied concepts of apathy, but is in early stages of development. Future work in this area, attempting to unify the various assumptions and mechanisms underlying the concept of apathy, is key in uniting the field and furthering knowledge. As it stands, the fragmented nature of the concept of apathy makes research in the area difficult to unite.

2.5.3 Discussion – How is apathy measured in HD and other neuropsychiatric conditions?

The 14 novel measures of apathy identified in this review took a range of approaches to measuring apathy. The measures differed in terms of their content, structure, form, and scoring systems. Each measure was validated in a specific population, mostly with a neurological condition such as PD, HD, or a form of dementia. The DAS (Radakovic and Abrahams 2014) and AMI (Ang, Lockwood, Apps, et al., 2017) were validated in a healthy population. The differences in approaches meant that the apathy phenomenon elicited by each measure was at least slightly different.

There are varying items included in the measures that have different aims and explore different elements of a person's behaviour. The items that each measure uses will necessarily change the phenomenon elicited by the measure. While measures all report statistical factor loadings (usually single factor, although some report three factor models) that show the measure only loading onto one or three factors, these factors are not necessarily the same across all measures. The LARS, for instance, assesses nine domains that include everyday productivity, interests, and initiative-taking, but still reports a global sum-score. Therefore apathy, according to the LARS, is a combination of those nine domains that all load into one score that directly reports a person's level or degree of apathy. The IAS, meanwhile, only includes four items that (as they state in their paper) only cover one domain as they all assess a person's interest. Since the IAS is only interested in a person's level of interest, albeit from different perspectives, the phenomenon of apathy that it elicits will only account for a person's interest and not the other eight domains that the LARS assesses.

A more complex measure offers greater discrimination between participants, usually assesses more domains, and is subtler in its assessment of apathy. This is not necessarily a good thing, however. In terms of assessing apathy, the LARS is more discriminatory and offers more subtlety than the IAS as it includes both open- and closed-question formats, assesses nine rather than one domain, and gives individual domain scores as well as an indication of apathy. The inherent complexity of the LARS, however, could also hinder its utility by increasing participant burden and possibly confusing participants as it jumps from one domain to the next. There is a shorter version of the LARS which cuts down to seven domains, but this is still far more complex than the IAS.

The way in which questions are posed is also important in determining the phenomenon of apathy elicited by the measures. When assessing motivation, the AES-C asks if motivation is present, whereas the DAS asks if motivation is absent, and the AMI states that "I am motivated" and asks a participant to agree or disagree. Similarly, the NPI asks if the patient lacks motivation or has lost interest. These approaches assess a patient's level of motivation and interest but start from a different perspective. This happens throughout the measures, with some seeking to identify a loss of motivation, while others look to identify presence of motivation. These are two different questions that are likely to elicit different responses. Identifying a presence of motivation implies that a person's normal behaviour lacks motivation, while identifying a lack of motivation implies a person is usually motivated. A further problem with assessing motivation is that it is an unusual symptom to talk about. In everyday conversation, it is relatively unlikely for people to discuss their level of motivation. The unusual nature of the question, be it posed positively or negatively, is likely to have an impact on a person's response.

Another area of difference that is important in understanding the phenomenon of apathy elicited is the issue of who administers the measures and to whom they are meant to be administered. A different version of a person's behaviours, feelings, and emotions will be elicited from different perspectives. Most measures are aimed to be administered with patients by their clinicians, but a few measures are administered to carers by clinicians or are self-report. A self-report measure of apathy may cause problems related to a person's level of insight into their behaviour and emotional state, while a carer-administered measure of apathy may miss some subtleties that a self-report or clinician-administered measure is more likely to pick up. Radakovic et al. (2017), for example, found that self-rating of apathy was 38% in people with PD and 33% when rated by their caregiver and Valentino et al.'s (2018) specific study of this issue found significant discrepancies between caregiver and self-reported levels of apathy. These findings should be considered when assessing apathy, and further that interpretation of apathy differs from different perspectives. These issues are occasionally discussed in the development of measures, but it is also common for measures to report statistical successes and reliability of a measure and assume that this is a proxy for accurately revealing the construct of apathy.

2.5.3.1 Language usage:

The language used in the measures often seemed overly clinical, a problem compounded by some of the measures' insistence that the interviewer should use the exact language in the measure. During the validations of all the measures, there were no reports of participants being confused by the tests or having difficulty understanding the questions posed without further clarification. Given that one of the findings in the literature is that apathy is most highly correlated with cognitive decline or impairment, particularly in participants suffering from various forms and stages of dementia, it seems highly plausible that researchers would have had to clarify some of the questions at least occasionally. Take the following question from Starkstein's SCIA, a structured interview with a patient administered by a clinician:

"Are you indifferent to your health condition, working situation, hobbies or personal care?" (Starkstein et al., 2005, p1071).

This question is an operationalisation of criterion B4 in the diagnostic criteria Starkstein adapted from Marin's original work. This question clearly addresses B4 and provides a link between the diagnostic criteria and the measurement of apathy. The language, however, contains multiple clauses and is an unusual, confusing combination of words. If a participant does have any degree of cognitive impairment, at the very least their answer will suffer from a degree of the primacy-recency effect, where people remember only the first and last item they heard. In this case, a participant may not remember to include their working situation or hobbies. While this question may increase the likelihood that the SCIA demonstrates good operationalisation of Starkstein's diagnostic criteria, it seems likely that using it with participants with cognitive impairment would drastically reduce its ecological validity. In HD, particularly, it would be reasonable to expect a large proportion of people would have a degree of cognitive impairment and may therefore struggle with some of the questions in the various measures.

Even without cognitive impairment, the language used in measures often seems designed to be useful in a research situation but not necessarily in a clinical environment. The LARS is perhaps better than other measures when it comes to question design but still includes questions that seem more likely to confuse a participant than encourage them to discuss their state:

"Do you take part spontaneously in daily living activities or do you need to be asked?" (Dujardin et al., 2013, p2018).

A combination of an unusual word (spontaneous) and unusual phrase (daily living activities) seems likely to confuse most people, regardless of their health status. The LARS is another structured measure that states that it should be administered verbatim:

"To obtain the best validity, it is not advisable to change the vocabulary or to add additional comments to the questions." (LARS appendix)

From a methodological perspective, not changing the wording of an item is a rigorous, theoretically reliable way to administer a measure. To be able to administer a measure verbatim, however, it must be appropriately worded for the people who will use it. Sockeel et al. (2006) state that the LARS was designed for use in PD and acknowledge that it is likely that apathy is highly prevalent in neurological and psychiatric disorders, yet the wording of the LARS is often potentially challenging. Although a verbatim delivery allows a measure to be administered rigorously, it drastically reduces its ecological validity. Future measure development must take this into account and ensure that the language used in each item is easily understandable, that prompts are permitted to be used and explicitly stated, or a measure does not have to be administered verbatim. Allowing this variability will introduce methodological problems associated with rigour, but it is likely that anyone using potentially confusing measures already introduce this variability by going 'off-script' during measure administration.

2.5.3.2 Under-reporting of item inclusion:

One of the frustrations of looking at how papers conceptualise apathy and then the items included in the measure is that there is often no or little clear link. There is a logical progression in some measures; Strauss and Sperry in the DAIR consider a lack of initiation and interest a key part of apathy, and there are several items that clearly relate directly to a lack of initiation and lack of interest. Even here, however, there is no direct explanation as to why a lack of initiation and a lack of interest are necessarily symptoms of apathy. It is unclear if what Strauss and Sperry consider to be apathy is a restrictive conceptualisation whose conditions must be exhaustively met before a person can be said to be apathetic or if, as Robert et al. (2002) seem to suggest in their scoring of the IA, the presence of one of these symptoms is enough to call someone apathetic. Much of the focus of the development of measures of apathy is in ensuring the statistical validity of the measures; this is undoubtedly a key component of any measure and must be included in any reporting of an apparently good measure. In the case of apathy, a concept that much of the literature acknowledges is under-developed conceptually, a clear link between items in a measure and the experience of apathy needs to be shown in order for the field to advance.

Perhaps the best example of this demonstration of a link between item-inclusion and people's experience of apathy was the reporting of the development of the DAS (Radakovic and Abrahams, 2014). In this development, Radakovic and Abrahams report how the triadic structure of apathy is mapped onto eight items per subtype, all derived from previous literature but amended to improve some of the language and fit a triadic structure of apathy. While this is a rare good example of reporting of measure development, the development of the DAS in a healthy population and reliance on previous literature does still make for some unusual wording in the measure that may confuse people with cognitive impairment, particularly bearing in mind that the DAS is designed as a self-report measure.

2.5.4 Discussion – Diagnostic criteria:

There are four sets of diagnostic criteria, outlined in Table 2. All four use a similar layout, refer to apathy as a relative state, and present apathy as fundamentally a state of loss. Marin, Starkstein's, and Robert et al.'s (2009) first criteria refer to a loss of motivation, while Robert et al. (2018) refer to diminished goal-directed activity in a purposeful move away from describing apathy a loss of motivation. Robert et al.'s (2018) criteria are certainly the most comprehensive and also offer the most difference between the four in terms of how it conceptualises apathy. Prior to this, none of the older criteria drastically improved or altered the other's, meaning that it was difficult to determine which to use other than by using the most recently developed.

Robert et al.'s (2018) criteria, however, do offer an improvement over the previous three. The move away from describing apathy as a loss of motivation due to its association with subjective interpretation of a psychological state (see Levy and Dubois, 2006) is debatable, but it does demonstrate that this set of criteria incorporates recent developments in the interpretation of apathy. The description of apathy as a quantitative reduction of goal-directed activity is, and will be to many, very similar to describing apathy as a loss of motivation but does move the focus of diagnosis to identifying set behaviours that may be demonstrative of apathy. Other than this change of language, these criteria use a fuller description of changes in emotional and social states that mean a diagnosis of apathy can be a relatively broad diagnosis. In other words, a person can present as only experiencing a reduction in their empathy and still be considered potentially apathetic. Previous diagnostic criteria were relatively strict (in an effort to narrow down the conceptualisation of apathy), but recent research has shown a potentially larger set of symptoms associated with apathy that these criteria are able to consider.

One problem that remains with Robert et al.'s (2018) diagnostic criteria, however, is that they do not consider diagnosis of apathy in different domains or suggest different subtypes of apathy, as much newer research is starting to do. Not all of the criteria have to be fulfilled for a diagnosis of apathy, allowing for some flexibility as mentioned above, but there is still no recognition that a person may experience a subtype of apathy. Much in the same way that people can experience various types of depression that can impact them in different ways, research by Levy and Dubois (2006) demonstrated the possibility of different types of apathy (albeit only in terms of neurological correlates) and more recent research by Radakovic and Abrahams (2018) has demonstrated a plausible framework of apathy that suggests each subtype is likely to impact different people in different ways and at different times. As it stands, Robert et al.'s (2018) diagnostic criteria do not allow for this variability of experience.

2.5.5 Discussion – Conceptual difficulties:

One of the main purposes of conducting a systematic review is to assimilate a wide range of evidence in one place, understand that data, and provide a robust, evidenced conclusion to the question posed in the review. When the data being assimilated are quantitative and use relatively similar statistical tools, such as when assimilating randomised controlled trial evidence, there are clear methodologies that provide reasonably objective results. One of the difficulties of this review has been the assimilation of disparate, often unlabelled data. This review sought to understand how the literature conceptualises apathy, but in doing so, one of the findings has been that there are very limited instances of this happening. To further complicate this, determining and interpreting what authors mean when they do define or conceptualise apathy is not a straightforward task. Although several papers sought to conceptualise apathy, it was difficult to precisely understand what the authors meant. It is important to stress that this is not a reflection of poor-quality papers, but a linguistic, conceptual problem.

There is no set way to conceptualise a potential disorder like apathy. There is recognition in the literature that is ill-defined, but there are no suggestions as to how this lack of definition should be solved. Robert et al. (2009) sought to present a unified set of diagnostic criteria for apathy, presumably as a starting point to getting apathy included in the next edition of either the DSM or ICD. Apathy remains undefined in either of those manuals, and, based on the lack of consensus in the field and relatively sparse conceptual work in the literature, probably with good reason.

2.5.6 Discussion – Lack of standardisation:

Another relatively common theme in the later literature is the acknowledgement that the lack of consensus surrounding the definition of apathy is problematic. Unfortunately, while this is recognised in the literature, very few papers attempt to address this problem, instead often bypassing the problem and focussing on estimating prevalence rates using a potentially flawed methodology. Any prevalence rates or measures of apathy are inherently flawed until a standardised definition of apathy is agreed (and ideally with a conceptualisation of apathy that is recognised and adopted by the DSM or ICD).

Many papers acknowledged that the lack of standardisation was a problem in the field and that the lack of consensus meant comparing apathy prevalence rates or understanding patient experience of apathy was flawed. This acknowledgement generally did not get discussed in relation to the study itself, however. This lack of discussion of the conceptual issues related to apathy make a lot of the papers' conclusions difficult to corroborate. If a paper acknowledges that the definition of apathy is problematic and uncertain, it is then difficult to agree that the concept under investigation is necessarily apathy and that the conclusions drawn, for instance that apathy reduces quality of life in patients with cerebral autosomal dominant arteriopathy with subcortical infarcts and leukoencephalopathy syndrome (CADASIL, Reyes et al., 2009), are valid. Reyes et al. (2009) certainly showed that quality of life was reduced in patients who they identified as apathetic, but as they used the NPI and apathy scale to identify apathetic patients, the most certain statement we can make is that patients who scored highly on the NPI apathy sub-scale and the apathy scale demonstrated lower quality of life scores. In other words, the conclusions that Reyes et al. (2009) present, along with many author similar findings in other papers, are only as good as the measures used. As most authors conclude, the measures of apathy currently available present multiple conceptual problems as they define and conceptualise apathy differently.

2.6 Limitations of this review:

Conducting this narrative systematic review involved collating a large range of data from a variety of sources. Papers included ranged from discussions of the concept of apathy, to statistical analyses of measures of apathy, to reports of diagnostic criteria. This variability allowed the review to discuss many elements of the concepts of apathy, but also presented a problem of cohesion. No standardised data extraction tool was appropriate for use, so a method of data extraction based predominantly on the researcher's decision-making was used. While every step was taken to ensure that this process was rigorous and data extraction was a particularly lengthy process, it remains possible that papers were missed or data was incomplete.

Another potential limitation of the methodology chosen was that it relied on the researcher interpreting authors' conceptualisations of apathy from the writing in their papers. It is plausible that misunderstandings may have occurred here, particularly since many of the papers included in the review were not explicitly exploring the concept of apathy but rather brushed this issue aside. While this was one of the reasons conducting this review was necessary for this thesis, it remains a potential limitation.

No funding was acquired for this review. Future reviews in the area would benefit from dedicated funding.

2.7 Summarising the concept of apathy and moving towards the empirical work:

One of the most evident findings from this review is that the concept of apathy is defined in a variety of ways across a large number of papers. Many papers sought to estimate the prevalence of apathy within certain populations but did so without setting out a clear definition of apathy. There is no consensus in the literature surrounding a definition or conceptualisation of apathy, a finding supported by the lack of definition in either the DSM-V (APA, 2013) or ICD-10/11 (World Health Organization, WHO, 1992, 2018). Relatively new diagnostic criteria for apathy have been developed (Robert et al., 2018), but these have also not been adopted by either main diagnostic manual. Recent work in the field, however, has advanced our understanding of apathy and begun to find common conceptual ground. The work of Radakovic and Abrahams (2018) to develop the Dimensional Apathy Framework after developing the Dimensional Apathy Scale (one of three scales used in the later data collection for this thesis), in particular, is promising in bringing together a broad range of research into one reasonably unified theory. Continued work in this area seems both reasonable and potentially fruitful. There seems to be general agreement that apathy is syndromic in nature and that it is a multi-dimensional construct best defined (currently) in a triadic structure. There are still problems, however, with showing that even this most recent of research is able to map and understand people's experience of apathy and begin to relieve the heavy burden of apathy on patients and their families.

Indeed, it is the burden of apathy which perhaps is lacking in this review and in the literature more generally. While apathy is a relatively under researched area when compared to other fields, over 60 papers were included in this review, yet very few discussed the impact of apathy on individuals and their families. The literature tends to concentrate on identifying apathy from a particular standpoint (neurological, psychiatric, or other), and postulating how the concept may present. These are, of course, important questions for our understanding, but they are of academic interest to people currently suffering from apathy. Perhaps the idea of experiential data is anathema to the predominance of neurological and statistical approaches to identifying what apathy is and what it looks like, but exploring how people feel and how their lives change when apathetic will only serve to improve our understanding of the concept of apathy.

3. Chapter Three – Exploration of methodology

The aim of this thesis is to explore the concept of apathy, with specific examination of the etymology of apathy, how the literature conceptualises it, how people with HD experience apathy. Chapter one looked at the etymology of apathy, as well as how apathy is defined and thought about in both lay and medical usage. Chapter two then delved deeper into the medical definitions and conceptualisations of apathy, finding that there is no standardised definition of apathy and a range of conceptualisations and measures of apathy. This chapter will focus on how to explore the concept, given what is already known about apathy, and which methodologies allow for exploration of the experience of apathy.

To explain the appropriateness of the chosen methodology, there are several interrelated concepts that require exploration:

- Firstly, the *emergence of psychology* as a distinct field will be discussed with reference to the relationship between the natural and social sciences. This will help to explain how the study of apathy in people with HD straddles the boundaries between the natural and the social sciences.
- A short exploration of *objectivity and subjectivity* will follow, centred around their definitions and practical implications, and will explore to what extent these concepts are possible or desirable in psychological research. This will be directly related to the study of apathy and will focus on the middle-ground that the study of apathy occupies. A brief description of the importance of pragmatism in research will then conclude with an explanation of why a predominantly qualitative technique is most appropriate to exploring the concept of apathy in this thesis.
- The *methods and apathy* section will then outline various qualitative research tools and ends in a discussion of the approach chosen.
- The last section explores the roots of the chosen methodology, including how Heidegger's work may be relevant to the exploration of apathy.

Two things must be noted here. This project is taking a pragmatic, mixed-methods approach to researching apathy. Secondly, and related to the first point, the three sections described above are split for ease of explication rather than suggesting that the concepts are opposed (as has been assumed in the past) or mutually exclusive. Discussing the relationship between the natural and social sciences, for instance, will necessitate discussing objectivity and subjectivity. All sections are interrelated and together form a cohesive exploration of the methodology used to explore apathy given the exploration undertaken in chapters one and two. The order of the sections is designed to start from the broader issues about the nature of knowledge acquisition by comparing the natural and social sciences, and gradually reduce to the specific issues about how best to approach the study of apathy. Therefore, the last section is a summary of qualitative research methods and will culminate in the justification of why interpretative phenomenological analysis (IPA) is the most appropriate tool to use to understand the experience and definition of apathy.

3.1 The emergence of psychology:

To discuss the emergence of psychology as a distinct academic field of research necessitates discussing where psychology (as a separate field) began and how the early progenitors saw this emergent field. In the early 19th century, the work being conducted under the somewhat loose field of psychology was closely aligned with philosophy and broadly interested itself in the study of the meta-physical, leaving the natural sciences to deal with physically demonstrable elements and theories. There was a clear demarcation between biology, chemistry, and physics, the three natural sciences, and psychology.

In the mid-19th century, however, psychology began to be interested in more tangible elements of human behaviour and began to develop a new area of research. Psychology moved away from meta-physical, philosophical questions and began to approach the study of human behaviour in an experimental manner, borrowing techniques usually seen in the natural sciences. Psychology was becoming a scientific pursuit, using methodologies from natural science to study human behaviour.

The renowned 'father' of experimental psychology, Wilhelm Wundt, studied medicine and was a physician and physiologist before turning his attentions to the field of human behaviour. In 1874, Wundt published his book 'Principles of Physiological Psychology' and described it as "an attempt to mark out [psychology] as a new domain of science" (Fancher, 1979, p. 126). Wundt was at the forefront of this new area of research. Psychology was now the study of human behaviour and the workings of the mind. Philosophy was interested in how the mind worked from a meta-physical perspective and natural science was interested in the physical mechanics of the brain, while Wundt was interested in studying the structure of the human mind and systematically examining how individuals perceive stimuli. Using repeated observations and carefully controlling what participants were exposed to, Wundt created an experimental form of psychology, and showed that the study of human behaviour could be approached using a rigorous methodology previously only used in the natural sciences. This new area of scientific and academic inquiry was a technique casually misunderstood, both at the time and in more recent historical reflections. While Wundt held that the brain could be structurally examined through rigorous observations, he did not believe in the idea of reductionism. To Wundt, the idea that the mind could be broken down into its constituent elements without losing its humanity was obviously fallacious. Wundt was not searching for steadfast rules or theories, or to develop the Newtonian mechanics of the mind. Rather, Wundt believed that it was possible to reveal some psychological mechanisms through observational research, but, as Kant stated some years before, large parts of human behaviour are not predictable or simply observable. He held that it was impossible and naïve to apply a perspective driven by the natural science approach when approaching the study of the mind (Wundt, 1910) because there is a fundamental and qualitative difference between naturally occurring phenomena (e.g. gravity or cause and effect between natural events) and human behaviour. Wundt stated that one of the major differences between the natural sciences and psychology was that cause and effect must be found or discovered in the natural sciences, whereas in psychology, our "inner processes" (Wundt, 1910, p109) immediately connect perception with events. His view, that human behaviour and the constituent elements of the mind were more than the sum of their parts, was in direct contrast to other psychologists and the reductionist school of thought.

One of those who Wundt fundamentally disagreed with was his former student, Edward Titchener. Titchener was a proponent of the theory of Structuralism, which sought to deconstruct the mind and consciousness into their constituent parts and then analyse the process by which these components formulated an experience. In doing this, Titchener used, among other techniques, the practice of introspection, which he learned in-part from Wundt (Schacter et al., 2011). Introspection is a form of reflection whereby a person examines their own experience using controlled, replicable observations and is still used in techniques such as the think-aloud protocol (Kuusela and Pallab, 2000), albeit in a heavily adapted form. Introspection was a key tool used by both Wundt and Titchener, but also one of the fundamental differences in their thinking. The work conducted by Titchener after he left Wundt's tutelage, and which developed into the theory of Structuralism was a major divergence from the work of Wundt. Titchener used Wundt's name to forge a successful career in America, but developed theories that Wundt did not recognise. Despite using the same methods, Wundt and Titchener fundamentally disagreed. Just as psychology was becoming an independent subject, there were schisms concerning the direction that psychology should take.

Titchener contested that the physiological make-up of a person could explain their behaviour and that therefore it was (at least theoretically) possible, given enough explanatory variables, to develop a unifying theory that would explain human behaviour. He was a reductionist; he saw the 'problem' of the brain and consciousness as a mechanical problem, believing the brain to be merely another organ that somehow generated what we call consciousness. Further, and to Wundt's displeasure, Titchener believed that introspection was a useful tool in revealing some of the mind's workings but was not a powerful enough method to explain any mental process. Comte and Durkheim, other early pioneers of the umbrella term social science (i.e. psychology, sociology, economics, etc.), both suggested that the study of human interactions and societal research could be completed with data collection techniques taken straight from the empirical methodologies developed in the natural sciences. Comte believed that the social world was as amenable to generalisable laws as the natural world and developed his positivist theories accordingly (Macionis, 2012). He drew a direct comparison between natural and social phenomena, stating that both are predictable given enough observational data. Durkheim, arguably the father of sociology, suggested that social phenomena such as crime and suicide existed independently of society and could therefore be studied in a similar manner to the objects of natural science (Yar, 2004). These reductionist positivist views that human behaviour can be simplified and predicted in the same manner as the study of gravity or other inactive phenomena, however, have been arguably under threat since Comte and Durkheim's assertions.

Wundt was staunchly against the notion of reductionism and believed that the generation of consciousness was not a simple biological mechanism, but a complex interaction of biology and environment. He described Titchener's and similar views in combative style, stating they were:

"…materialistic pseudo-science which sufficiently reveals its tendency to destroy psychology by claiming that the psychological interpretation of mental life has no relation to mental life itself as it is found in history and society." (Wundt, 1910, in Mischel, 1970)

Wundt held that physiology alone could not explain mental processes and that attempts to do so had no place in modern psychology. Resorting to explaining mental processes purely in terms of physiological make-up was, in Wundt's eyes, lazy, unscientific psychology and an attempt to subvert the promise of psychology by reverting to studying human behaviour purely in the natural sciences. Indeed, Wundt later stated that even if it were possible to see the workings of the mind "as clearly before our eyes as the mechanism of a pocket watch" (Wundt, 1910, in Mischel, 1970), the underlying mental processes would not be explained by this physiological mechanism.

It is interesting to note that the main disagreement between Wundt and Titchener is a debate which continues today. With the advent of functional Magnetic Resonance Imaging (fMRI) and increasingly accurate imaging techniques, psychology is beginning to see the brain and its inner workings in a similar fashion to Wundt's hypothetical pocket watch. We know which areas of the brain control which areas of the body and are able to see which parts of the brain are most active when we perform certain activities. Yet, despite these incredible advances, we are arguably not much further on in understanding Wundt's 'mental processes'. Perhaps Wundt's non-reductionist argument was indeed correct, and the human brain is more than merely the sum of its parts. There are those, however, who believe that more advances in brain imaging will allow us to 'see' problems in our mental processes and consequently fix them, while there are others who acknowledge that no matter how much we understand the mechanics of the brain, there will always be the elusive ghost in the machine. Both Wundt and Titchener identified a possible route for psychology if superior technology was available. Titchener believed that mapping the psyche through physiology alone was technically possible, while Wundt maintained that experimentally rigorous techniques could further our understanding of the human condition but would not fully explain or reveal consciousness. Later, Ernst Mach, the eminent physicist and philosopher, agreed with Wundt, stating that:

"…a color [sic] is a physical object as soon as we consider its dependence, for instance, upon its luminous source, upon other colors, upon temperatures, upon spaces, and so forth. When we consider, however, its dependence upon the retina ... it is a psychological object, a sensation. Not the subject matter, but the direction of our investigation, is different in the two domain[s]". (Mach, 1897)

Mach clearly defines a difference between the physical existence of an object and the psychological existence of the same object, echoing Wundt's view. Colour exists in the universe completely independently of humans (we, as humans, presume). Our experience or sensation of colour, however, does not and cannot exist without further, human mental processes. It is logical, then, to surmise that the study of the physical and the psychological are qualitatively different, as Wundt said, and should therefore be treated as such. This source of divergence between the physical and the psychological is also where the natural and the social sciences drastically, and necessarily, move away from each other. Natural science is focussed on the physically demonstrable and views colour as a permanent object; colour exists in the Universe as a certain wavelength and will remain the same whether a human is viewing it or not. Social science, on the other hand, is not interested in the physical existence of colour, but is instead interested in how humans see, interpret, and experience that wavelength in their life. For the social sciences, the existence or inexistence of colour is inconsequential; what is important is how humans see, interpret, and experience that colour.

Natural and social science, then, should perhaps be viewed as diametrically opposed. The things they explore are completely different. Dilthey (1989) and Weber (1949), suggested that there is enough of a difference between the social (Dilthey describes 'human' science) and natural sciences to warrant the use of entirely different methodologies. Ingthorsson (2013) has argued that we should change the way we understand and judge research instead of trying to design studies that replicate natural science techniques while studying a fundamentally different phenomenon. Instead of using specific methods and assumptions derived entirely from the natural sciences, we should reframe social science methodology to reflect this qualitatively different nature of the phenomena under investigation. Danziger (1998) also addresses this, taking influence from Wundt's ideas and describing the conflation of natural and social science as "the pretence that psychological experiments are not in principle different from experiments in the natural sciences." Free will in the research object of social science necessitates an acknowledgement that psychological experiments are different. Danziger continues, stating that it is surely better to study the researcher-participant relationship rather than pretend it does not exist in psychological research. In a contribution to an article, Ingthorsson (2013) states that understanding social relationships is equally as important as natural science's obsession with "merely physical, unconscious matter" (p28, 2013). Social sciences, he continues, study "meaningful phenomena" (p28, 2013). Ingthorsson points out that natural science deals with "lawful phenomena" (p31, 2013) that obey specific sets of rules; the existence of colour, independent of humans, can be determined without the necessity of human interaction. Social science deals with "meaningful phenomena" (p28, 2013) and tries to understand how humans experience colour regardless of the physical existence of colour. In other words, Ingthorsson agrees with both Wundt and Mach that the natural and social sciences should be held to account by different standards.

This simple opposition between the aims and methods of natural and social sciences, however, is somewhat reductionist (a point which Ingthorsson concedes later on, stating that he has deliberately exaggerated the differences between natural and social science to emphasise his point). It is certainly reasonable to assert that in the mid-19th century, the scientific field was not as broad as it currently is (if for no other reason than the greater amount of technology available today) and therefore the differences between the scientific fields in existence was larger. As the scientific community has grown, however, the overlap between fields has become pronounced. Neuropsychology, a field that attempts to understand how cognition and behaviour are influenced by brain physiology, is neither entirely a natural nor entirely a social science. It demonstrates that although natural and social sciences certainly investigate qualitatively different phenomena, they do not exist in isolation, and can work together. There is undeniably a significant, valuable overlap between natural and social sciences. This is both interesting and problematic. It suggests that both Wundt and Titchener's influences on the direction of psychology have remained. Neuropsychology is a melding of Wundt's staunch non-reductionist ideas about observing human behaviour and Titchener's purely reductionist ideas that we can isolate behaviour to brain physiology.

This melding of the natural and social sciences is important when thinking about the concept of apathy. The study of apathy, particularly in people with HD, is a phenomenon similar to neuropsychology and operates between the natural and social sciences. HD is a degenerative, neurological condition that affects a person's brain functioning. Through neurological research, we know (broadly speaking) how HD affects someone's brain in a similar way to Wundt's idea of revealing the mechanism of a pocket watch. We know that HD causes a multitude of neurological symptoms that have horrendous consequences for individuals and those close to them. Apathy seems to be a particularly prevalent symptom of HD with estimates of prevalence ranging from 33-76% and it seems to have a debilitating effect on a person's motivation, interest, and engagement in activities they previously enjoyed. The cause(s) of apathy is unknown and although some have used neuroimaging techniques to try and isolate where apathy may be located in the brain and gone so far as to suggest treatment implications (Thobois et al., 2017), this is a potentially problematic approach that reduces apathy to its neurological correlates. In the long term, a neurological approach may identify the systems implicated in causing and continuing apathy in a person and that may lead to useful breakthroughs for treatment and management. In the short term, however, this neurological approach is of little benefit to people currently suffering with apathy. Additionally, a solely neurological, natural science-inspired approach is bound by our lack of understanding of neurology. Until a breakthrough occurs, there is no possibility of improvement for people currently apathetic. The neurological approach should, therefore, be one part of a multifaceted approach to understanding apathy.

An approach that focusses on understanding the concept of apathy from a behavioural and experiential approach will greatly help both a short-term goal of improving treatment and management for people currently living with apathy in HD, as well as aid the long-term goal of understanding neurological correlates of apathy. Exploring people's experience of apathy, what it means to them, and how it affects their lives will help future research identify appropriate treatment and management techniques.

3.2 Objectivity and subjectivity:

Objectivity is defined in the Oxford English Dictionary (OED) as "the ability to consider or represent facts, information etc. without being influenced by personal feelings or opinions…" (OED, 2014). Its antonym, subjectivity, is described as "the quality or condition of being based on subjective consciousness [or] experience [and] the fact of existing in the mind only" (OED, 2014). Objectivity in research, therefore, is the ability for a theory or conclusion to be demonstrably unbiased and to reveal a truth about the world using a methodology informed by a validated form of inquiry. It also implies that the truth revealed exists in the world independently of the researcher, is not purely socially constructed, and that the same conclusion could have been drawn regardless of the individual conducting the research. Objective findings in research are often used to create universal statements or laws that are not broken in the natural world unless a revision is made to the statement to include any anomalies that are discovered through further research.

Objectivity in most types of scientific research is therefore typically aligned with a positivist, reductionist standpoint that seeks to make generalisable laws from observational and experimental data. As discussed above, social science, particularly experimental psychology, often uses a positivist, empiricist standpoint and employs the same methodologies as the natural sciences as this is seen as the gold standard of knowledge acquisition (Feyerabend, 1975).

In the field of psychology, however, objectivity and empiricism can be particularly troubling. Contrary to Comte and the beliefs underlying positivism, human behaviour does not follow observable, predictable natural laws. Newton's laws of motion apply universally (on this planet) to all objects; we can accurately predict how a stone will react if kicked but cannot consistently predict how a human will react. Newton's laws will dictate the direction, speed, and distance a rock will travel if we know enough about the parameters of the situation. To attempt to predict and understand a human's reaction to being kicked, we must gather information about the scenario, the people involved, and the preceding events before subjectively interpreting the situation using our acquired knowledge about the possible social, cultural and personal ramifications of the kick. Even with all of this information at our disposal, we still cannot say how an individual will react. The crux of this 'problem', and one that non-sentient objects do not possess, is that humans have agency, free-will, and the freedom to react; if gravity had free will and the freedom to choose, Newton's laws would not apply universally. The problem for psychology, however, is that society (particularly in the West) views the positivist, empiricist, reductionist scientific method as superior to other forms of knowledge acquisition, leading psychology to desire objectivity and the scientific method without being sure if it is possible within the sphere of studying human behaviour. Although Feyerabend (1975), among other great thinkers, reasonably argues that scientific knowledge acquisition is no more or less valid than other human-created forms of enquiry (such as common sense or superstition), this is not a commonly held belief.

On a basic level, natural science involves exploring non-human subject material, such as the orbit of planets or how cells divide. In this way, natural science involves looking into a system from the outside and being able (theoretically) to objectively observe all the components and establish natural laws, given the appropriate equipment. Being able to observe an object of inquiry from a neutral position makes generating truly objective, empirically sound, falsifiable theories possible. Social science, however, is a study of human behaviour and social relationships (OED, 2014) that involves looking into a system from the inside. In analysing, discussing, and generating theories in social science, we are necessarily interpreting another person's interpretation (Weber, 1949), something which cannot be done objectively but by understanding and working with the inherent subjectivity. As long as the methodology employed allows for this, there is no reason why, as Feyerabend implies, the pursuit of subjectivity in research is not as useful as the pursuit of objectivity. Instead of trying to imitate the natural sciences' positivist claim to objectivity, social science should study social constructions such as interpersonal relationships as "objectively real entities" (Ingthorsson, 2013, p25).

It is necessary to point out that although the natural sciences often lay claim to being entirely objective and without subjective bias, this is somewhat of a misnomer. Positivists claim to present the definitive search for objective reality and go further, claiming that the truth which science reveals is the only possible version and the only valid form of knowledge acquisition (Egan, 1998). Yet, in studying any subject, we are engaging in a multitude of processes that are entirely subjective. Using the English language to think about the subject, using a pen to write down ideas, or even employing the five human senses could be considered a pollutant to 'true' objectivity. To be entirely objective in one's approach to the study of anything would involve not being influenced by anything other than the object of inquiry. All received knowledge would have to be rejected and the inquirer would need to start their investigation from the very beginning, potentially by developing a new form of language or communication not necessarily based on established traditions. Evidently, any form of reasonable inquiry cannot do this. While this is exaggerating the definition of objectivity to absurdity, in line with Kant's essay on enlightenment (Kant, 1983), it also reveals the absurdity of the positivist notion that all knowledge can be treated the same. In the social sciences, for instance, this definition of objectivity does not make sense.

In a positivist sense, to be truly objective in the social sciences would involve studying humanity from an objective, removed position, influenced only by reason and logic. As previously stated, it is arguably plausible to study some parts of natural science objectively as they involve looking into a system from the outside; e.g. as a human it is possible to objectively examine a rock and postulate how it was formed without needing to rely on the cultural or behavioural characteristics of the rock, since it has none. It is not, however, possible to examine either oneself or other humans without already having some level of expectation or received knowledge that influences one's judgement and renders the research, to some extent, subjective. (Although it must be pointed out that the same problem can be levied at natural science, as objectivity is impossible without using received knowledge, which arguably renders research subjective). Merleau-Ponty (p. 139, 1968) identified this problem, stating:

"Thus, since the seer is caught up in what he sees, it is still himself he sees: there is a fundamental narcissism of all vision."

It is impossible, as a human, to step outside human experience and objectively view one's life and behaviour; we are always influenced by the social world that we exist in and we cannot step outside the system. Indeed, Bhaskar and Callinicos' (2003) critical realism suggests that not only are our interpretations of the world entirely subjective, but there is likely a reality which does exist 'out there' in the world, but we are unable to accurately see this reality without viewing it through our own subjective lens. This idea, that an external reality exists but we are not able to see it, is particularly interesting. Bhaskar and Callinicos' critical realism stems from the same positivist standpoint that it is easy, as a psychologist employing a Rogerian, person-centred approach, to reject outright. This rejection, however, is unfair. Critical realism is what may be termed a softer positivism. Given the progression described above in both the natural and social sciences and the manner in which subjects such as neuropsychology work towards an understanding of the human condition by using both paradigms, a critical realist approach makes logical, intuitive sense. While denying a purely positivist outlook on knowledge acquisition, it is tempting to defend experiential data over observational data and become convinced that the answer to our question lies purely in examining the human condition. A combination of critical realism and hermeneutic reflection (further examined in the section below on interpretative phenomenological analysis) should remind us that it is important to be able to step back and take note of our own assumptions while remembering that we may not be able to see the reality that exists independently of human experience. A purely positivist, objective approach to knowledge acquisition suggests that the correct methodology can identify this reality, while an experiential, social science approach would suggest that this independent reality is not important since we cannot identify it. Both of these approaches are overly reductionist.

Despite these seemingly insurmountable problems that lie at the heart of assumptions behind psychological research, and likely due to the import that society places on the notion of objective research, psychology employs predominantly positivist assumptions and uses the scientific method to answer most research questions. The gold standard of research in various fields is often understood to be the randomised controlled trial (RCT; Akobeng, 2005), a method used to test a range of things from pharmaceutical interventions to psychological theories. This method aims to observe objectively the behaviour and reaction of two or more groups of randomly selected people. RCTs are viewed as an extremely rigorous methodology that tries to eliminate bias and allow the researcher to control as many variables as possible and identify the active ingredient in a treatment, be it pharmacological or psychological. The scientific method is laudable in its attempt to exert control on whatever the subject matter is, but when human behaviour is involved in the investigation, objectivity and control are rarely possible.

The positivist, empiricist methodologies used in social science, however, do produce a large volume of excellent quality research. These scientific, objective methods provide testable, falsifiable conclusions that satisfy a lot of our assumptions about whether the ideas generated are high quality. As stated above, however, the assumption that scientific, objective methods provide high quality evidence in every field are not necessarily accurate. Again, Feyerabend's supposition that scientific knowledge acquisition should not be valued over other forms of knowledge is key. Kant's (1983) enlightenment essay, originally published in 1784, also disagrees with the current high regard for this form of knowledge acquisition, instead arguing for the rejection of all received knowledge. While this is an entirely impractical idea, it does provide a useful explanation of why the methods and approach typically aligned with natural sciences are so highly praised. The notion of the economy of thought is important here in respectfully refuting Kant. Firstly, as described by Jourdain (1914), economy of thought refers to the notion that since there have been so many breakthroughs in scientific discovery, it would be a ridiculous and impractical notion to begin a process of rediscovery in rejecting all received knowledge (with the possible exceptions of quantum mechanics and other slightly esoteric fields of enquiry). In receiving knowledge from those who have come before, the process of discovery continues forward, rather than stagnating as every enquirer wonders why the apple fell from the tree. Secondly, there are human limitations that impose a restrictive economy of thought. Although ideally, we may want to understand and study everything, we do not have the mental capacity to do so (Argyrou, 2013). We need to concentrate on using our limited time and mental resource in an economical, pragmatic manner. Employing an economy of thought, then, means that we can progress in research and knowledge without, necessarily, questioning everything we do. It is a somewhat contradictory state of affairs but one that is borne of necessity and pragmatism, rather than employing a purely Kantian set of methodologies that require every researcher to start from scratch. Social science, like natural science, employs an economy of thought by using taken for granted assumptions so that it may continue to research previously unstudied fields rather than reinventing previously determined elements. Therefore, the use of an economy of thought can be seen to be contributing to social science employing and valuing the scientific method over all others. Instead of reinventing the wheel, methodologically speaking, it is easier to continue using a method that has been seen to work.

When it comes to studying apathy in HD, adopting the middle ground between objectivity and subjectivity, and between the natural and social sciences, is a necessary starting point. As a concept, apathy is extremely loosely defined, as demonstrated in the systematic review in chapter two. The current literature does not provide a standardised definition of apathy. Instead, each paper seems to have its own definition of apathy, which may or may not be explicitly stated. This has created a slightly odd situation. There are a large number of measures that purport to measure apathy, but it is likely that each of these measures has validated a unique concept that may or may not be apathy. All of the measures have used a quantitative, positivist-inspired methodology to develop the measures, and have done so extremely well. Clarke et al. (2011), in their review of the measures of apathy, concluded that each measure was relatively good and well developed, but without clarity over the term apathy, they were difficult to corroborate as a group. This lack of understanding must be addressed in order for us to be able to accurately assess and eventually treat apathy.

3.3 Methods and apathy:

Research is often split into two methods; qualitative and quantitative. It is now common practice to employ a mixed-methods study wherever possible, whereby one part of the data collection will use a quantitative technique, and the second a qualitative technique. Mixed-methods studies overcome a lot of the disadvantages of using just one method and are a more complete investigatory tool. In a general, simplistic sense, qualitative studies engage in subjectively analysing the research object, while quantitative studies try to objectively separate the researcher and research object. There is, equally however, objectivity and subjectivity in both methodologies. Some qualitative studies look for causal relationships, while quantitative studies can involve the subjective interpretation of statistical results. It is also the case that both quantitative and qualitative methods are not homogenous groups. The underlying assumptions behind a methodology often differ in qualitative techniques (as will be discussed later), and the assumptions held in predictive and Gaussian statistics, for instance, are entirely different.

As Bryman (2006) states, the war between staunch proponents of quantitative, empirical work and qualitative, inductive work has "largely subsided" (p111). Qualitative and quantitative methods are no longer opposed epistemological approaches but merely different tools that answer different questions and are designed to collect, analyse, and report different datasets. This pragmatic approach to research design is a useful step forward in understanding the world around us and is particularly useful for the social sciences to be able to choose one or more appropriate methodologies for the study of a chosen research topic.

The concept of apathy, as discussed above, is a complex phenomenon and one that requires careful investigation. The current apathy literature has taken a predominantly quantitative approach. The literature has attempted to identify a group of patients who are diagnosed with, or at risk of, developing apathy and then devised a measure of apathy to confirm or refute a diagnosis. The focus has been on identification of apathy through rigorous quantitative testing and using statistical analysis to validate the developed measures. Marin (1990, p143) describes apathy as a:

"…lack of motivation that is not attributable to diminished level of consciousness, cognitive impairment, or emotional distress".

There is no discussion, however, about what this definition means to patients or health care professionals. None of the three diagnostic criteria for apathy have been adopted by either the Diagnostic and Statistical Manual V (DSM-V; APA, 2013) or International Classification of Disorders 11 (ICD-11; World Health Organisation, 2018).

Apathy, then, is in its conceptual infancy and needs to be treated as a relatively unknown concept. Qualitative research allows for an exploratory method better than quantitative. Qualitative research allows for an open framework in which the participants can guide the data and tell the research what they think apathy comprises. While it is theoretically possible to do this with a quantitative method (for instance using a think-aloud method; see van Oort, et al., 2011), the iterative and prescriptive nature of quantitative research required to produce replicable, reliable, and objectively valid findings would stifle participants' thoughts. If, for instance, a questionnaire was sent out to potential participants who were diagnosed with apathy asking them to answer a set of questions, the questions would have to be prescriptive and implicitly test a set definition of apathy. Alternatively, an open-ended questionnaire could be sent out to participants, allowing participants to provide their own answers, unguided by the researcher's assumptions. These questions can then be quantitatively coded and fed back into the statistical analysis of the data corpus. This method of quantitative surveying, however, immediately introduces an extremely subjective element to an otherwise objective process, leaving the results and analysis difficult to validate.

A qualitative, semi-structured interview, however, allows the participants to inform and guide the research, almost entirely subjectively, based on their personal experience of apathy. As Labuschagne (2003) points out, qualitative research is fundamental in developing a concept from a stakeholder's perspective. Quantitative research allows for rigorous testing and quantifying of a problem or concept, while qualitative research allows an in-depth analysis of meaning and detailed description of the elements that constitute a problem or concept. While quantitative research requires a reductionist view of the data under investigation, qualitative techniques require an exploding of the data in order to view the concept in its entirety (Smith and Osborn, 2008).

3.4 Methods:

This section will build on the above exploration of scientific inquiry and discuss the main methods of qualitative inquiry and an outline of interpretative phenomenological analysis (IPA), the method which will be used to analyse the data resulting from semi-structured interviews with patients and carers.

3.4.1 Discourse analysis:

Discourse analysis (DA) is a form of qualitative data analysis that concerns itself with the way in which we create social reality through the language we use. DA is not as interested in the individual behind the language used but more in how the language itself creates a description of the subject under investigation. Potter and Wetherell (1987) offer perhaps the most well-known and frequently used version of DA, which is often labelled discursive psychology. Discursive psychology takes issue with the assumptions underlying cognition theory in mainstream psychology and does not subscribe to the notion that language is necessarily a route to being able to 'see' cognition. Instead, the discursive approach supposes that people use language to present the best version of themselves in any given context. If a person is asked the same question by different people in different scenarios, they are likely to give a different answer. DA is an extremely useful method for exploring 'the why' behind language used. It is commonly used in gender studies, for instance, or for analysing the way in which two people interact in a certain scenario. It is not appropriate for the current study exploring the definition and experience of apathy in HD, as the focus of the investigation is the experience of the phenomena of apathy rather than (as would be more appropriate when using DA) the relationship between two people, one of whom has been diagnosed with apathy.

3.4.2 Thematic analysis:

Thematic analysis (TA) is a broad, qualitative analytic method that is predominantly used in psychology and is often used as an "accessible and theoretically flexible approach" (Braun and Clarke, 2006). TA is a descriptive technique which offers little to no theoretical grounding and can therefore be used in almost any research situation to give a quick analysis of the data under investigation and identify patterns within the data. The process of conducting TA is like other, more complex and theoretically grounded methods such as IPA. Researchers must read through the dataset and take note of any major themes that run through all the data. These themes are then used as the subheadings for the analysis which consists predominantly of describing participants' words and experience. Braun and Clarke (2006) offer perhaps the best practical explanation of TA, although it must be noted that they present a form of TA which is overly inclusive and capable of handling any data. While TA can be used to analyse a lot of subject areas, it is also important to note that its lack of theoretical grounding limits its usefulness when attempting to interpret a dataset. TA does not posit any position (like DA firmly standing against cognitivist assumptions) and therefore cannot claim to interpret a participant's thoughts or intentions that lie behind his or her language use. TA is inappropriate to use for this study for that very reason; when examining a person's experience of an understudied concept, it is important to be able to use appropriate and complex qualitative tools to frame the study within a theoretical framework. Using interpretation or hermeneutic reflection in TA, for instance, is not desirable as this necessitates clarifying the researcher's position within the data.

3.4.3 Narrative analysis:

Narrative analysis (NA; Cortazzi, 1993) is a form of qualitative data analysis often used in psychology, but also used in other social sciences. NA refers to a way of analysing data and creating a story based on the experience and description provided by the participants. NA will often present the data analysis in a traditional 'storied', chronological manner with a distinct beginning, middle, and end. Based on the premise that humans are by nature social animals who use language, words, pictures, and art to tell a story that will help future generations, narrative psychology is a way of making story telling scientifically rigorous (in the sense that it has a defined methodology). In addition, NA often gives voice to individuals who might otherwise not be heard by society. NA can be effectively used to tell the story of a disenfranchised part of society. Although this method would create an interesting interpretation of apathy in HD, it would be focussed on the story of apathy rather than the experience of apathy. While these are very semantically similar concepts, the story of apathy and an individual's experience of apathy are two separate entities. Telling the story of apathy would involve concentrating on a sequence of events that led up to a conclusion or ending of the story. When exploring a concept such as apathy, there is no ending, but rather a continuing description of experience. IPA is better suited to presenting and analysing an individual's experience of apathy and not necessarily having to form links between all the items of experience a person may describe to form a cohesive story.

3.4.4 Grounded theory:

Grounded theory (GT; Charmaz and Belgrave, 2015) is a novel, complex inductive method of data collection and analysis. It is not purely a qualitative methodology, but rather a method of conducting research. It is similar in approach and assumption to IPA but is perhaps what may be termed a 'purer' method in that it attempts to be entirely data-led and without agenda. GT is a method that is focussed on directly 'grounding' all research conclusions or theories in the data collected. That is to say that research conclusions or theories must be explicitly demonstrable in the data. When using GT, the researcher is interested in understanding a participant's meanings, intentions, and interpretations of events that have occurred to him or her. A novel part of GT's method is that the direction of the project is often led by the data, rather than (as in hypothesis-driven research) being led by one question that is laid out from the start. This data-led approach is a concrete way for GT to ground itself in the participants' experience.

GT has the potential to be a useful method for exploring apathy but could also present some difficulties that the project would not overcome. The data-led approach that GT necessitates mean that the qualitative interview data collected would not, definitively, be about apathy. Although the researcher would (presumably) ask questions relating to apathy, the participants may not have thought about the relatively abstract and unusual concept of apathy before and thus would not talk about it unless explicitly asked. One of the main problems that this thesis is attempting to address is that there is no clear definition of apathy in the literature, and there is no description of apathy from a patient's perspective. This suggests that a GT methodology that would necessarily start from a broad perspective and later move on to more specific lines of inquiry led by the data is unlikely to end up discussing apathy. In addition, the tradition in GT to delay the literature search until after analysing interview data would preclude this method. While GT and IPA share some common traits in their approach to prioritising the participants' experience over hypotheses, IPA remains a more appropriate, practical way of exploring an ill-defined concept such as apathy.

3.4.5 Interpretative Phenomenological Analysis:

This part of the chapter will explain why using IPA is an appropriate and useful methodology for exploring the concept of apathy and understanding how people experience apathy. An explanation of what IPA is will clarify the methodology. An explanation of some of the important philosophical issues related to IPA will follow in order to give context to the methodology. An explanation of how these issues will help to uncover the concept of apathy will also be integrated into these sections.

Interpretative Phenomenological Analysis (IPA; Smith, Flowers, and Larkin, 2009) is an inductive qualitative research tool that is used to explore how people make sense of an event or phenomenon in their lifeworld (Smith and Osborn, 2009). It is primarily interested in how people interpret their experience and how they integrate that experience into their life. IPA is not interested in an objective account of an event, but instead is interested in understanding a person's experience and what it means for them. In analysing data, an IPA study will ask critical, interpretative questions of a person's account of their experience to try and understand experiential elements that may be hidden (e.g. attempting to interpret motive behind a word or a phrase that a participant uses). Developed by Jonathan Smith and colleagues, IPA's emphasis on making sense of experience lends the method to exploring people's experience of healthcare in human, health, and social sciences. While much of the practicalities of conducting a study using IPA are procedurally similar to the majority of qualitative research tools, IPA's development and roots in phenomenology, hermeneutics (the study, theory and practice of interpretation) and critical realism (outlined above) are unique. The philosophical elements that have been used in the development of IPA provide a theoretical and practical framework for conducting research and for interpreting an individual's experience.

IPA takes a phenomenological approach to answering research questions and is heavily influenced by the field of phenomenology (literally 'the science of phenomena'; Moran &

Mooney, 2002, p5). Championed by Husserl, Heidegger, and Merleau-Ponty (and often with reference to earlier work by Hume and Kant), phenomenology is the study of subjective human experience. Phenomenology is interested in what and how an individual person experiences, and how this contributes towards their understanding of their lifeworld. Thus, IPA aims to interpret experience and examine how experience reveals the phenomenon under investigation.

3.4.5.1 Husserlian phenomenology:

Husserl (2001) famously advocated that phenomenologists go "back to the things themselves" (p168) when engaging in phenomenological inquiry. By this, Husserl means that experience should be reduced into its constituent elements (by performing what he termed an eidetic reduction) and the essence of the experience understood. Further, when trying to interpret experience and its elements, Husserl states that the phenomenologist must be able to distinguish between the experience itself, and one's own interpretation or preconception of how a person experienced an event. A researcher's previous assumptions or interpretations of the phenomenon under investigation should therefore be identified and ignored (often referred to as bracketing off). In looking to explain experience, it can be all too easy to jump on what may be a familiar phrase or tone and immediately assume that we know what and how the person is experiencing the phenomenon under investigation. Husserl's eidetic reduction, however, pulls us back from immediately explaining or interpreting a described experience and insists that we first identify and rid ourselves of any preconceptions. This will ensure that our own preconceptions cannot interfere with our understanding of another person's experience.

Husserl is equally vehement in his rejection of the notion that knowledge gathered through natural science techniques is any more worthwhile or valid than our everyday understanding of the world. Husserl's phenomenology stipulates that our subjective, everyday understanding of our experience is first-order knowledge (loosely 'common sense'). This therefore must mean that knowledge gathered with an objective natural science method is second-order because it is predicated on our first-order experience. In short, our understanding of the scientific world is contingent on our experience of the world around us that we take for granted. Without common sense, we would not be able to take the ontological leap to developing a perceived higher-level of 'scientific' knowledge. For Husserl, then, the process of phenomenologically revealing subjective experience is first-order knowledge and is able to accurately give an authentic, real description of the 'what' and the 'how' of a person's experience without being contaminated by a researcher's preconceptions.

3.4.5.2 Heideggerian phenomenology:

In both assertions above, Husserl is describing a theoretical, conceptual process that places the natural sciences and phenomenological inquiry as directly opposing. As described in the previous sections, this notion of the "paradigm wars" (Bryman, 2006, p113) is no longer a useful way of describing or thinking about practical research.

Heidegger, in comparison, offers a more practicable version of phenomenology that is not so extreme in its positioning. Heidegger intended his ideas to be an antidote to Husserlian conceptual phenomenology and provide a practical guide to conducting phenomenological inquiry. One of his most useful suggestions was the ridding of Husserl's idea that it is possible or desirable to bracket off our own preconceptions. Heidegger starts his phenomenological inquiry from the position that understanding experience is a constant and often inactive interpretative process. It is therefore impossible to not interpret and so aiming to bracket off our own experience is impractical. We cannot experience the world without engaging in a constant interpretative process; thus, aiming to inhabit an objective realm in which we do not interpret or use our preconceptions, as we must if we are able to perform Husserl's eidetic reduction, is unreasonable. Instead, Heidegger moves away from Husserl's reductions and instead focusses his phenomenology on existential questioning of our experience and what it is to exist.

In Being and Time (1962), Heidegger delves into how it is that we, as humans, existentially come to be and to experience the world and explores perhaps his most well-known, if difficult, concept, '*Dasein'* (literally 'there-being' or 'existence'). For Heidegger, *Dasein* can be seen as the questioning of the kind of being we are that is exclusive to us as humans and can be thought of as a means to understanding our '*In-der-welt-sein'* or Being-in-the-world. *Dasein* is Heidegger's explanation and interpretation of the uniqueness of being human, of being an interpretative being, and encapsulates the shared experience of being human in a social world (see Haugeland, 2005). We, as humans, are unable to experience not existing (we do not choose to be born) and must engage in interpreting the world in order to understand it, at least on a basic level. *Dasein* can perhaps be thought of as the existence of the social world of humans. Individually, humans must interpret the world to exist in it; we make choices based on what is in front of us, our experience past and present, and how we wish to exist in this world. These choices are wrapped up in the social world in that our choices have ramifications on that world, but also the social element of the world is made up of those individual choices of *Dasein*. This slightly tautological explication also means that if we did engage in Husserl's bracketing of our preconceptions and interpretations, we would not be able to understand the social world we are trying to explore. Related to the concept of apathy here, Heidegger's *Dasein* should be seen as the way in which apathy may mediate or alter people's relationship with the world around them (if at all). Has people's understanding of themselves and the world around them, social and otherwise, significantly changed or does there seem to be a perhaps equally surprising lack of change? Are there elements of the world, of people's Being-in-the-world and *Dasein*, that have fundamentally changed? These are important questions that Heideggerian phenomenology suggests will represent a significant change in people's lifeworld.

Related to *Dasein* and understanding the social world within which we exist and create, Heidegger compares two modes of Being-in-the-world; '*Eigentlichkeit*' or authenticity, and '*Uneigentlichkeit*' or inauthenticity. Authenticity is being true to one's own self in the decisions that one makes, while inauthenticity is doing things because they are done, thereby being passively part of the 'they' self that exists in the social world. Neither are necessarily a positive or negative state, although Heidegger does extol leading a good, authentic life and appreciating the finiteness of our being, particularly in relation to Being-towards-death, as Carel (2008) in particular discusses in her exploration of illness and ill-health. Inauthenticity can be a useful part of the social world that allows us to follow social norms, while authenticity allows us to consider our *Dasein* and try to act in a way that is true to our self, in whatever form one decides that self exists. It is perhaps ownership of our decisions and choices that fundamentally splits these two modes of Being-in-the-world; is it the choice of 'my' self to act in a certain, authentic way or have I ceded my inauthentic choice to the 'they' self of the social world? In this way, the social world also not only exists 'out there' but also within each person. We form the social world by interacting with others but also by interacting with our own self and our authentic or inauthentic choices. Perhaps another way of thinking about these two modes of being is also in terms of a passive and active state. For the most part, our *Dasein* is passive; we exist in the social world, passively interpreting, and make many decisions that are inauthentic, ceding to the 'they' self in a similar way to the notion of the economy of thought discussed earlier in this chapter. Many choices we make in everyday-life can be inauthentic because it is a pragmatic use of our time to economise thought in this area. Inauthenticity is therefore a necessity of authenticity where authenticity is an active process of questioning one's *Dasein*. Again, these concepts will be particularly interesting when applying them to people's experience of apathy. Do people's experiences of apathy alter their authentic and inauthentic modes of Being-in-the-world? Do they demonstrate ownership of their behaviour or is the they-self of Heidegger's '*Uneigentlichkeit*' more powerful in their post- or during-apathy time? Perhaps apathy allows elements of increased authenticity in their lives as people care less about social norms or cues. These are questions that Heideggerian phenomenology asks here when exploring the concept of apathy.

Heidegger also spent a large portion of particularly his later writing exploring the concept of boredom, which may be of use here when considering the concept of apathy; at a superficial level, and perhaps more, boredom and apathy could be similar notions. For Heidegger, boredom (or '*Langeweile*') is one of the main attunements or moods of *Dasein* and exists in three distinct states with distinct relations to the experience of time and *Dasein* (see Hammer, 2004, for a full evaluation of the three states of boredom). Heidegger seems to suggest that boredom is a state in which some of *Dasein* is suspended; our meaning-making (or our illusion of meaning-making), that is normally a fundamental part of Being-in-the-world, is removed, stripping away part of our interpretation of phenomenon. When we are fundamentally bored, in the sense that Heidegger means, we see what may be viewed as perhaps a purer version of phenomenon. Our boredom allows an existential interpretation of *Dasein* (Slaby, 2010) and is therefore a gateway to authenticity. Heidegger's concept of boredom is reminiscent of the Stoic philosopher's state of apatheia in its postulation of an altered state of being that allows for a different, perhaps greater understanding of the world around us. This idea may be useful as an additional interpretation of how people experience apathy.

Heidegger adopted an interpretative form of phenomenology that accepted that Husserl's idea of bracketing was not possible because *Dasein* forces us to interpret the social world we exist in, even if only passively, and seeks meaning in phenomena on top of a descriptive explanation. Heidegger's two modes of being, authenticity and inauthenticity, describe two ways of Being-in-the-world and of understanding one's own self. When exploring another person's experience, these concepts are key. What interpretative processes are on-going in a person's experience and are they active or passive choices?

3.4.5.3 Practicalities of interpretation:

When using IPA, a researcher must engage with these philosophical debates and decide how they apply, practically, to a research project. The 'I' of IPA is naturally more aligned with Heidegger's interpretative phenomenology, but this alone does not provide firm advice on how to use IPA. Additionally, Heidegger's interpretative stance seems to make more logical sense than Husserl's claim that the world is describable without using any personal preconceptions. Some other concepts are needed to further explain the practical issues related to using IPA as a research approach.

During the analysis process, the researcher is attempting to make sense of how the participant makes sense of their world (Smith and Osborn, 2008). This process, a double hermeneutic (Giddens, 1984), is a key assumption of IPA. Hermeneutics is the theory of understanding and interpreting phenomena and is often associated with the interpretation of religious and philosophical texts; to engage in the practice of hermeneutics is therefore to attempt to understand and interpret a phenomenon. This process involves one individual using their experience, intelligence, and prior knowledge of a subject area to gain a novel understanding. In this sense, hermeneutics concentrates on one active agent (the individual) trying to uncover an understanding in an inactive subject matter (the religious or philosophical text). Thus, a double hermeneutic is the theory, expounded by the sociologist Anthony Giddens, that explains one of the key differences between the natural and social sciences. The natural sciences can be described as utilising a single hermeneutic as a researcher often attempts to understand an inactive event (in that the event or phenomenon under investigation does not have free will). In contrast, the social sciences are subject to Giddens' double hermeneutic, whereby the researcher is attempting to understand and interpret the experience of an active subject matter. In IPA, this double hermeneutic is immediately evident. Adopting a Heideggerian position, we can assume that participants are trying to make sense of the world around them and interpret how their experience relates to the world. During the research process, the participant attempts to tell the researcher about their experience. The researcher is therefore engaging in a double hermeneutic, attempting to understand an already interpreted understanding of the world. At the same time, the researcher must be aware that they are also engaging in several interpretative processes; interpreting the world around them, as the participant is doing, but also trying to understand how the participant is experiencing that interpretative process. While being aware of this ongoing interpretative process does not necessarily change how a researcher conducts the research, when analysing the data, it is particularly useful to remember that what is being analysed is an interpretation of a phenomenon. This is useful as it reminds the researcher to analyse this subjective data carefully and not to over-interpret.

The phenomenologist van Manen describes a useful process for understanding a concept or phenomenon without over-interpreting the data available. As described above, existing in the world involves a constant interpretative process whereby we are forced to make assumptions or draw conclusions from the data available. When meeting people in certain situations, for instance, social interaction is easier and quicker if we assume common roles –

when walking into a shop as a customer, we immediately take on the role of the customer and expect to be treated as such by those we assume to be staff. When we do this, we assume and take-for-granted the roles of 'customer' and 'staff' and know what to expect. We do not question those assumptions as it is not useful. When engaging in phenomenological inquiry, however, we are questioning those assumptions and trying to find out what we mean by 'customer', what our expectations are, and what makes us a 'customer'. We are trying to uncover the 'essence' of the phenomenon we are investigating to understand the constituent elements that make up that phenomenon (van Manen, 2017). Phenomenological inquiry involves exploring the essence of something to be able to define it. Thinking about the essence of something helps us to stay grounded in the data available to us and not jump to conclusions by over-interpreting and taking-for-granted what we are studying.

Uncovering the essence of apathy involves asking participants what it is that they experience when they are apathetic and what element of that experience might be taken away for them to no longer experience apathy. To understand how apathy is experienced, we must reduce individuals' experience into manageable parts while maintaining an understanding of how that experience is part of participants' lifeworld. The analysis must always be able to explain how it is relevant to participants' lifeworld and how apathy is affecting them, and where precisely that interpretation is rooted in the data. In a similar manner to Husserl's mantra, van Manen encourages the phenomenologist to reduce concepts to better understand their essence and therefore what makes them themselves.

The final concept of particular use to this discussion of phenomenology, IPA, and apathy further addresses the difficult issue of bracketing. We can see that Husserl's bracketing was too extreme, while Heidegger's, despite claiming to be practical, offers little to the design of a project. The essence of bracketing, however, remains useful. When engaging in an interpretative process, understanding one's own preconceptions before engaging in interpreting someone else's experience must be useful. Finlay (2003) argues that understanding one's own experience and preconceptions is essential to fully engaging with any data gathered through IPA research. This awareness of one's own preconceptions allows the researcher to discover their unique position and allows for scrutiny of decisions made during the research. This, in turn, allows for further introspection and thus further analysis of the data gathered. Dahlberg (2006) suggests using the phrase 'bridling' (as in to bridle a horse) instead of the term 'bracketing'. Therefore, when we engage in the process of recognising and actively using our preconceptions to improve the analytic, interpretative parts of research, we take control of, or bridle, those preconceptions. We recognise them and keep them in check while interpreting the data available. We are unable to avoid interpreting the world when conducting IPA and must engage in subjective interpretation that relies on understanding our participants. We must take hold of our ideas about the phenomenon under investigation and make sure we control them during the research process. In this way, we can "reflexively uncover" (Finlay, 2003) the phenomenon while being able to identify our involvement in the research.

3.4.6 The difference between IPA and other qualitative techniques:

IPA is, at first glance, practically similar to other qualitative techniques such as thematic analysis. The main difference between IPA and other qualitative techniques lies not in the practical approach to transcription, coding, or analysis (although there are some), but in the approach to considering the topic under investigation. The reliance and insistence on understanding a range of philosophical subjects (briefly explored above) and how they interact is an integral part of conducting a good quality IPA study. IPA involves thinking about one's subject of investigation in a specific, rational, explorative, open manner. In using IPA, the researcher is committed to exploring a phenomenon in its entirety and understanding both the participant's and the researcher's relationship with the phenomenon.

IPA is both an approach to research, a philosophical position, and a systematic research tool. IPA places emphasis on an individual's account of an experience as revealing, explaining, and defining a phenomenon when a researcher engages with an interpretative process. Where other methodologies may place greater importance on simple description of events or experiences, IPA aims to take analysis a step further and encourages the researcher to engage in an in-depth interpretative process. IPA is not particularly interested in revealing an exact retelling of an event or experience (as may be the case if one were engaging in classic Husserlian phenomenology – see below) but is more interested in how an individual makes sense of the experience. The retelling of an experience may differ substantially from the precise order of things in reality, but this is relatively unimportant.

3.4.7 Application of IPA to the study of apathy:

The above briefly outlines the field of phenomenology, presents descriptive (Husserlian) and interpretive (Heideggerian) phenomenology, presents Giddens' double hermeneutic, van Manen's notion of the 'essence' of a phenomenon, and the concept of bridling. These are all important when applied to the study of apathy in people with Huntington's disease. We are trying to understand what the concept of apathy is and how participants experience it. In order to do this, we are asking participants to define apathy and give us an account of their experience of apathy.

We can see above that if we were to adopt a pure Husserlian approach, we would assume that the researcher was able to bracket off any and all of their personal experience and evaluate the participant's account from what may be termed an objective standpoint. This, however, is considered implausible. Instead, a Heideggerian approach will be adopted, meaning that the researcher will acknowledge all preconceptions of apathy. Moreover, the data collection process will be viewed not as a one-way flow of information from participant to researcher, but as a collaborative process of creating the data between participant and researcher. Given that the participant group will be experiencing apathy, a concept that seems to have a lack of motivation at its core, seeing data collection as collaborative is particularly important in ensuring that the data created is of a good quality. Building a rapport between participant and researcher and remaining adaptable throughout the process (e.g. allowing for changes to the protocol if it will aid a participant) will also help ensure the data collection and subsequent analysis is completed successfully.

Giddens' double hermeneutic is also a useful way of imagining the process that will occur throughout the research. The researcher will be trying to understand how a person understands the concept of apathy and how they experience their specific apathy. It is likely that some participants will have considered their apathy, while others will not have thought about it at all, and others do not feel themselves to be apathetic despite what others around them may say. This means that the research interview could be a particularly difficult interview, potentially in all cases. Understanding one's own motivation or will is not an easy concept to explore within one's self. The interviews between the researcher and person with apathy are asking the participant to uncover very personal feelings. It is important that the researcher takes time and effort to treat the interview process with care and to respect the participant.

The concepts of uncovering the essence of apathy and Dahlberg's notion of bridling are closely interrelated. Without bridling our preconceptions, it is easy to quickly and potentially erroneously misinterpret a participant's words simply because we think we have spotted the essence of apathy. This desire, to quickly explain, is something we are keen to do, particularly in the context of research where one of the main purposes is explaining a phenomenon. In addition, exploring the concept of apathy, it could be very tempting to put words in a participant's mouth or over interpret the data. We must, however, pull back on the reins and bridle ourselves until we have a clear, evinced interpretation of apathy.

3.5 Studying apathy:

What must be remembered throughout this study is that we know relatively little about the concept of apathy in practical, usable terms. The research conducted so far, mainly looking at measuring apathy, has been of high quality but does not seem to have uncovered the phenomenon of apathy. We do not know what the essence of apathy is, or how people with apathy experience and interpret apathy. We do not know how a researcher can actively interpret and reveal the concept of apathy through a person's account of their experience. We do know that it seems likely that apathy occurs relatively frequently among people with HD (based on the estimate of prevalence of 33-76% reported by van Duijn, Kingma, & van der Mast, 2007) but there are no empirical studies that detail people's experience in HD. An IPA study asking people with apathy in HD to explore their apathy will help to answer these questions and start the process of uncovering the phenomenon of apathy.

Apathy is an extremely complex idea that thus far has not been explored from a patient's perspective. This study will use patient experience, gathered and analysed in semi-structured interviews, as the main data in understanding apathy. In addition, by interviewing a relatively small number of participants (including patients and their primary caregivers), this will enable the data analysis to be extremely detailed, focussing on individual's experience and revealing how they are able to make sense of apathy.

IPA is considered most appropriate for studying the concept of apathy as one of the aims of IPA is to examine how people comprehend and make sense of their experience (Smith, et al., 2009). IPA helps to piece together the units of phenomena that make up participants' experience (Smith et al., 2009). The participants' individual experience of apathy, specifically the problems they identify, language they use to define apathy, and changes they or their carer recognise are all individual and must be treated as such. As IPA is non-prescriptive, and can be described more as an approach to doing research (Smith, Flowers, Larkin, 2009, p40), this method will give different results to the predominantly quantitative methods previously employed in the field, and will provide a new perspective on what it is to have apathy and be apathetic. IPA allows the research "to go back to the things themselves" (Husserl, 2001, p168) meaning that participants' experience is the key focus of the research.

4. Chapter Four – Methods

This chapter presents the methods used in conducting the data collection, including discussion of the validity of the qualitative methodology analysis.

4.1 Aim of the research:

The aim of this research was to understand how people with apathy and HD understand and experience apathy. Where possible, primary caregivers were also interviewed to elicit their understanding of apathy and to cross-reference the patient's and caregiver's experience of the patient's apathy.

4.1.1 Research questions:

The primary research questions were:

- How do people with apathy in Huntington's disease define and experience apathy?
- What impact does apathy have on participants' lives?

The secondary research question was:

- How does patients' apathy impact family members and/or carers?

4.2 Design:

The study used a mixed-methods approach. Qualitative analysis of semi-structured interview data was combined with analysis of three measures of apathy.

The semi-structured interviews asked people with apathy and HD, and their primary caregivers (where possible), to explore the concept of apathy. The interviews focussed on asking participants to define apathy and talk about their experience of living with apathy. As there are no interview data with this population currently in the literature, these data give voice to a previously unheard population. The schedule was relatively detailed (compared to other semi-structured interview schedules) as it was expected that participants would have difficulty answering questions. Some of the questions used were therefore very specific to facilitate a longer interview and to give the participant a chance to become as engaged as possible with the interview.

Following the semi-structured interviews, three validated empirical measures of apathy were administered with participants with HD and apathy. All the participants who consented to be interviewed wished to be interviewed at the same time, i.e. as a couple. This seemed to be partially because the caregiver was concerned for the overall wellbeing of the person with apathy, and also that the person with apathy would not be able to provide any answers on their own. From a methodological perspective, this was a limitation, but was also acknowledged as a possibility in the design of the study and did allow for useful direct comparison between the thoughts of both participants. The AES-C, LARS, and DAS were used with patients, with the caregiver present at the time and helping where needed. Notes of any conflict between the patients' and caregivers' responses were noted. Combined with the semi-structured interviews that were also conducted with both patient and caregiver present, this provided the study with the cross-referencing data that the separate interviews would have provided. It must be acknowledged that conducting the interviews with both people present may have led the caregiver to temper their answers somewhat. There were, however, numerous examples of the caregiver openly contradicting the patient (although always politely).

Interviews were conducted with eight participants with HD and three primary caregivers. Given the population, it proved difficult to get as many 'pairs' of participants (people with HD and their primary caregivers) as individuals. The participants who were still apathetic were particularly challenging to interview, thus the relatively large number of interviews conducted, as IPA is normally conducted with five or fewer participants. This has made the analysis process longer than is normal for IPA, but this is an accurate reflection of the complex nature of trying to understand apathy.

4.2.1 Population:

Participants were people with HD, either currently or recently believed to be apathetic by the clinical team, and their primary caregivers (where possible).

4.2.2 Eligibility criteria:

Two groups of participants were recruited; patients and their primary caregivers.

Patients had:

- A diagnosis of HD; and

- Were currently or recently apathetic (as determined by the clinical team)
    - Consensus among the clinical team was a prerequisite for stating that participants were apathetic. Any dissent was discussed in a multi-disciplinary team meeting – if no consensus could be reached that a participant had or were experiencing apathy, that participant was not recruited to the study;
    - All participants were aware that clinicians considered them apathetic at some stage, and had discussed this issue with their clinician prior to being involved in the research.

Carers were defined as:

- The primary caregiver for someone with HD (n.b. we expected this person to be a spouse or close relative but could also have been a person who cares for a person with HD full-time, non-professionally);

All participants had to:

- Verbally communicate in English; and
- Have capacity to provide informed consent.

4.2.3 Exclusion criteria:

Participants who exhibited the following were excluded:

- Severe dementia;
- Severe communication difficulty;
- Physically or mentally unwell to the extent that taking part in the research may have worsened their condition.

4.2.4 Recruitment:

In the original protocol, participants were going to be recruited through two centres; the clinical HD team and the Enroll-HD clinic at Castle Hill Hospital. Recruitment began with the clinical team who successfully recruited sufficient participants, therefore recruitment via the second centre was not needed

The clinical HD team, headed by my supervisor, Dr Markova, sees patients with HD in the Hull and East Riding geographical area. The clinical team identified patients with apathy and told them about the project. If the person was interested, the clinical team gave them a Participant Information Sheet (PIS) and asked if they could give the person's contact details to me. I then telephoned them at least 48 hours later (to give them a chance to read the PIS) and asked if they wanted to take part. If a person had a lot of questions or would rather meet in person to talk about the research, this was arranged, although this was not necessary with any of the participants.

4.2.5 Procedure:

The research procedure was relatively simple. All participants wished to conduct the interviews in their homes, with the exception of Daniel, who wished to be interviewed on hospital grounds. When meeting the participants, the researcher went through the PIS and consent forms with them, ensuring participants understood what the research entailed. Pre-advice was only given to participants if they stated they were unsure what apathy was. In this case, participants were read a brief definition of apathy that used standardised dictionary definitions of apathy. This was only necessary with one participant, Mary. The semi-structured interview then took between 15-60 minutes and was followed by a short debrief. Interviews were not a set length and they varied greatly in length, with, as expected, interviews with more apathetic patients taking considerably less time. A second meeting was then arranged for around three weeks later during which participants completed three measures of apathy. This took between 10-30 minutes. Although this was not advised for the measures, it was the best possible way to conduct the research given the wishes of the participants.

4.2.6 Data collection:

There were two stages of data collection, the first involving semi-structured interviews to explore participants own understanding of apathy, and the second involving using three validated measures of apathy to explore both the measures themselves and as a comparator to the semi-structured interview data. Both forms of data were then analysed, first separately and then comparatively, to answer the research aims.

4.2.6.1 Stage one – Semi-structured interviews:

Semi-structured interviews with patients and caregivers were conducted in which all participants were asked to discuss their experience of apathy. Patients were asked about their understanding of apathy, their symptoms, and to describe any changes in their lives. Caregivers were asked similar questions about apathy and how this affects both their and the patients' life. Caregivers generally seemed open and willing to politely contradict the patient during the interviews if they believed the patient was wrong or falsely remembering something. The semi-structured interviews with both patients and caregivers were audio recorded and transcribed verbatim, including notation of inflection, short and long pauses, and speech difficulties (e.g. stuttering).

In IPA studies, a relatively sparse interview schedule is common. This usually entails broad areas of inquiry that an interview should follow rather than detailed questions with follow-up items. A relatively detailed semi-structured interview schedule was used in every interview (Appendix 10.1). This was primarily to encourage the interviews to cover a wide variety of areas, where possible. In all interviews, as few prompts as possible were used and in no interview were all prompts used. In-line with accepted methodology, all interviews opened with broad, open-ended questions and came down to narrower, occasionally closed questions if necessary and much later on in the interview. This was necessary in, for instance, Mary's short interview as she often struggled to think of anything to say for broader questions. While this is not the norm for IPA, it reflects the cognitive difficulties of the population under investigation and is considered a necessary and appropriate adjustment for working with this under-researched group.

The semi-structured interview schedule was not adhered to when participants became particularly upset or struggled excessively to give an answer. Where possible, the researcher tried to minimise the impact and burden of taking part on participants.

4.2.6.2 Stage two – Three measures of apathy:

Three validated measures of apathy were administered with the patients, and all three of the caregivers wished to remain present. The measures used were Marin et al.'s (1991) Apathy Evaluation Scale-Clinician (AES-C), Radakovic and Abrahams' (2014) Dimensional Apathy Scale (DAS), and Dujardin et al.'s (2013) Lille Apathy Rating Scale (short form; LARS-SF). These measures consist of questions about patients' apathy, producing an apathy score. This second stage was generally conducted between two and three weeks after the first. This delay did seem to help in firstly reducing burden on participants, and secondly, ensured that the topics discussed during the semi-structured interview were not fresh in their mind.

These three measures were chosen after the exploration of measures conducted in the systematic review (chapter two) as they were all validated measures that were respected in the literature. Additionally, the measures were all relatively different from each other, thereby offering comparative value both to the semi-structured interview data and also to each other. Marin's (1991) AES-C uses 18 items to produce a global sum-score of apathy. A guideline for administering the AES-C was obtained and followed. Full details of reliability and validity of the AES-C is reported in Marin, Biedrzycki, and Firinciogullari (1991). For the purposes of their use here, it should be noted that Cronbach's alpha for internal consistency is 0.90 and Pearson's *r* is 0.88 for test-retest, representing good test scores for these statistical analyses.

Radakovic and Abrahams' DAS consists 24 items across three sub-scales, and does not produce a global sum-score. As the DAS was designed for both clinician and 'self' testing, very few guidelines are provided or necessary due to the relatively simple design that consists of a series of statements that participants agree or disagree with. Full details of reliability and validity are available in Radakovic and Abrahams' (2014) paper. Cronbach's alpha for internal consistency was a reasonable 0.798. It should be noted that there now exists a short form of the DAS, but this was not available when data collection was completed.

Dujardin et al.'s (2013) LARS-SF is based on the Lille Apathy Rating Scale (Sockeel et al., 2006). The short form was used here as the authors suggest that it is more reliable and easier to administer than the full scale. Full details of reliability and validity are available across the two papers mentioned here. The short form consists nine items, each with multiple sub-items. These then load on to seven areas that include emotional responses, motivation, and interests, all of which sum to a global sum-score of apathy. Cronbach's alpha for internal consistency was a reasonable 0.73.

4.3 Data analysis:

The semi-structured interviews were analysed using Interpretative Phenomenological Analysis (IPA). IPA was considered most appropriate as it seeks to examine how people comprehend and make sense of their experience and helps to piece together the units of phenomena that make up participants' experience (Smith, et al., 2009). IPA allows the research "to go back to the things themselves" (Husserl, 2001) meaning that participants' experience and interpretation of phenomena is the analytical focus.

The data collected from the validated measures of apathy were analysed using descriptive statistics based on the authors' guidelines. These data were then used alongside the semi-structured interview data to build up a fuller picture of each participant's experience. It was not appropriate or useful to conduct full statistical analysis on these data as there were no groups to compare or effects to analyse. In addition, as the sample size was 11 participants (eight patients and three caregivers), power or effect size calculations would be inappropriate.

4.4 Analysis validity:

The validity of qualitative data analysis has long been the source of debate. IPA acknowledges its reliance on Giddens (1984) double hermeneutic, which is an inherently subjective process; the researcher is trying to understand the participant who is trying to understand their lifeworld. Thus, the explanation of a participant's lifeworld that appears in the write-up of a data analysis has gone through a lengthy process of interpretation and reinterpretation.

To ensure that the data analysis was performed appropriately and rigorously, Yardley's (2000, 2017) validity criteria, as presented by Smith, Flowers, and Larkin (2009) in response to concerns about the potential for misinterpretation of IPA data, was used to assess the validity of the qualitative data analysis. Yardley's (2008) criteria were considered particularly relevant as they suggest assessing validity from the outset, throughout the research process. Thus, the quality of the data analysis was dependent on the quality of the data collection as well as the design of the research. There are four criteria for assessing validity, presented below with a brief explanation of how this research addressed the criteria:

1. Sensitivity to context – to achieve this, the context of the research must be clearly assessed and thoroughly developed through engagement with current literature and a careful, considered approach to data analysis.
    - A systematic review of the concept of apathy took place alongside the semi-structured interviews to ensure constant awareness of how apathy is viewed and that this was considered when analysing data from participants. This systematic review of how apathy is defined and the exploration of the experience of people with apathy and their primary caregivers gave the research a variety of information sources to use when considering the concept of apathy.
2. Commitment and rigour – the research must be committed to providing good quality, clear results and is performed in a rigorous, scientifically-valid manner.
    - The design of this study has been carefully constructed based on findings from a literature review, discussion with the supervisory team, and advice from the thesis advisory panel. The protocol was reviewed and approved by the Hull York Medical School (HYMS) and National Health Service (NHS) research ethics committees and supported by local NHS Research and Development offices. The combination of a systematic literature review, an exploration of the

    etymology of the concept of apathy, semi-structured interviews, completion of several measures of apathy with people with apathy and their primary caregivers has provided a cohesive, rigorous picture of apathy and enabled this research to further our understanding. The supervisory team and thesis advisory panel continuously monitored and supported the research to maintain this rigour.
    - Data collection (or the process of data collaboration) is a vital stage of any research. Particularly in IPA, care must be taken to ensure that a participant can trust and quickly build a rapport with the researcher. In this study, where the participants with apathy and HD were already dealing with a lot of clinical problems, a commitment to providing good quality research was both an ethical and moral imperative. As part of ensuring this commitment and rigour, I attended an advanced IPA training workshop prior to data analysis. This provided enhanced understanding of the necessity to collaborate with a participant to understand their experience. Data collection is not a simple question-and-answer exercise but a personal, reflexive, and often emotionally difficult process for both participant and researcher.
    - Cross-checking of data was performed by the researcher and project supervisor. One section of data was coded together to ensure our approaches to coding were consistent. Any differences in coding were discussed and appropriate changes made to the data analysis.
3. Transparency and coherence – the research process must be clear and understandable.
    - This chapter provides a comprehensive record of all stages of the research process.
    - This thesis has presented the current understanding of the concept of apathy, supplemented with data from participants, and suggests how this thesis may develop the concept of apathy. This runs throughout the thesis to present a clear and coherent narrative.
4. Impact and importance – perhaps the most important assessment of validity, this asks if the research tells the reader something interesting, important, or useful.
    - The concept of apathy is an important object of inquiry that this thesis has set out to explore and better understand. The three criteria above must be achieved for this goal to be met and for the thesis to give the reader a better understanding of apathy. Chapter seven, the discussion, ties together all aspects of this thesis and provides a novel description of the concept of apathy based on the previous chapters.

4.5 Researcher biography:

The researcher is in his early 30s, male, mixed race, has no known neurological conditions or adverse health needs other than being slightly hard of hearing, and is interested in people's experience of illness. He is experienced in healthcare research, particularly using qualitative methodologies, and has worked with people with various chronic illnesses, including Parkinson's disease, chronic kidney disease, and arthritis. He had not previously worked with people with Huntington's disease until starting this research.

In line with the reflexive, phenomenological approach used throughout this study, the researcher kept a short diary documenting his thoughts and reactions and has used this to inform the analysis. It was particularly useful to read back through the notes after the emotionally difficult first interview with the first participant and reflect on why that was so troubling. The use of the reflective diary led to several discussions with the PhD supervisory team about the potential for describing different types of apathy, particularly related to a participants' level of insight into their own behaviour, that are discussed later in this thesis.

4.6 Conducting research in a sensitive area:

There are several potential issues that must be considered when conducting research. The participants in this research were living with HD, or with someone with HD, at the time of interview and were potentially vulnerable and this was important to remember when conducting the interviews and subsequently analysing the data. It must be noted that while the aim of this research was to develop a better understanding of apathy, this was not a priority over the participants' dignity or consent. The researcher made every effort to ensure that all participants understood and were comfortable with the research process, and were respected and considered throughout the research, including any potential publications, presentations, or further research results.

4.6.1 Communication and HD:

There are several symptoms of HD that can affect a person's ability to communicate effectively or clearly. There are several physical symptoms that can directly affect a person's speech making it physically difficult for a person to speak. Cognitive symptoms can induce memory loss or difficulty accessing memories, which can reduce people's ability to express themselves. Apathy itself could also present a communication barrier. Particularly if patients lacked insight into their apathy, they could find it difficult to talk about their experience.

These potential communication difficulties could have meant that people were not able, or were unwilling, to properly articulate their feelings to the researcher.

The inclusion criteria above stipulated that participants must be able to verbally communicate and be able to provide informed consent. This would preclude any participants whose memory loss was so severe that they could not remember large portions of their experience. The other communication difficulties mentioned, however, particularly the physical symptoms related to speech difficulty, did have an impact on the research.

Any communication difficulties between researcher and participant were dealt with on an individual basis. The clinical staff recruiting for the study were aware that participants needed to be able to communicate in English and relatively well.

Another communication issue, touched on above, concerned the problem of participants' insight into their apathy. People who clinical staff identified as apathetic, but who did not themselves acknowledge their apathy, or actively argued they were not apathetic, were considered lacking insight into their apathy for the purposes of this research. Some participants stated that they used to be apathetic but are not anymore, despite, from a clinician's perspective, no change in motivation or activity level. In these cases, when interviewing a patient who does not have insight into their apathy, there were several ethical and moral issues that were carefully considered. Firstly, the interviewer did not imply that the patient was apathetic unless the clinical team openly used this terminology when talking to the participant (and this was clarified before recruitment). Secondly, any suggestion in the analysis that the patient was apathetic when they stated that they are not, was done with extreme care, stressing that this was the researcher's opinion.

4.6.2 Coping with HD:

Living with HD is extremely difficult and distressing from both a physical and emotional perspective. One of the potential problems of in-depth qualitative interviews is that they ask a participant to actively engage in reflecting on the difficult parts of their lifeworld. During this project, the researcher asked participants to reflect on their apathy in the context of HD. This was a potentially very upsetting topic that some may not have thought about before or may not have articulated before. This put the researcher in the highly privileged position of being allowed access to a very personal part of a person's lifeworld. This position of privilege comes with responsibility. At all times, the researcher was prepared to suspend or stop the research and provide the participant with as much support as was appropriate, be that informing their family and friends, referring them to their clinical team, or providing professional help. This was only necessary in one interview, as reported in chapter five.

4.6.3 Coping with apathy:

As soon as the recruitment process began, one of the clinical staff had trouble in interesting a potential participant. One person was asked if he wanted to take part in the research, and he replied that he, "couldn't be bothered" (or words to that effect). The particularly frustrating aspect of this recruitment difficulty is that this person may have been a particularly good participant to interview.

Methodologically, interviewing people with apathy was extremely difficult. When conducting semi-structured interviews, maintaining appropriate methodological standards is important. Where possible, the interviewer must not ask leading questions, must not suggest an answer to a participant, and must not prompt too many times or push a participant into answering a question. During these interviews, however, it was necessary to use some leading questions or suggest answers. Using these techniques led to a better rapport being built with the few participants who this was necessary with and was methodologically appropriate given the phenomenon under investigation. Other methodologies were considered, particularly when the first interview was relatively short and did not produce the best data. Elicitation techniques such as asking participants to keep a short diary (verbally or writing one down), or photographic representations were considered. All other methods, however, were rejected in favour of conducting semi-structured interviews as this was felt to offer the best compromise between getting good data and not overly burdening participants.

4.6.4 Interviewing primary caregivers:

The term primary caregiver or simply carer is used in this project to refer to the participants who cared for someone with apathy and HD. For this project, these participants were either someone's spouse or parent.

There are a few potential issues that arise from interviewing a primary caregiver. All participants preferred to be interviewed together as a dyad rather than individually. This was expected and participants were given the choice to either conduct the interviews together or separately. Confidentiality is also difficult to maintain when interviewing a pair of participants. Part of the interpretative process of conducting an IPA study involves interpreting the words a participant used and giving them meaning that perhaps the participant would rather the researcher did not know. In addition, and possibly more importantly, a primary caregiver may say something during the interview that they did not wish to share with the person they care for. This happened in one interview where the patient's spouse said a few things that the patient was not expecting. This was explored during and after the interview and both participants were happy for the disagreements to be included in the transcript.

5. Chapter Five – Results

This chapter reports descriptions of semi-structured interviews, including background information on each participant and a report on salient information from the interview. Additionally, the results from the measures of apathy are reported and discussed. Chapter six then analyses these results in more detail, including a thematic analysis of the interpretative phenomenological analysis performed.

5.1 Introduction:

In analysing the data from the eight participants with HD and apathy and three of their primary caregivers, several steps were performed, in line with accepted methodology for conducting an IPA. These steps are described in more detail in the previous protocol chapter, chapter four. These are not distinct, separate processes, but on-going elements that help to shape the researcher's understanding of each participant's lived experience. Each transcript was analysed individually, with repeated readings of the transcripts helping the researcher process what was discussed during the interviews. In respecting the participants' stories, it was important to understand and tell each participants' story before moving on to collate the findings and group them into overarching themes. In this chapter, each participant's story is outlined, while chapter six prioritised interpreting the experiences of apathy and HD to shed light on what it is to experience apathy.

The reports in this chapter give an overview of each participant, detailing some of their biographic information, and briefly outlines and gives context for the interview. Within these introductions to the participants, the researcher's impression of each is given. This is important as some subjective interpretation of each participant's apathy, even very simply whether they presented as apathetic, is necessary to aid discussion of the interview and measures data (e.g. if there is a mismatch between the interview data and the results of the measures of apathy, the researcher's impression of the participant can be used to contextualise this mismatch).

The results from the validated measures of apathy are also reported in this chapter. The measures provide further context for each participant's experience of apathy and a brief commentary of the measures is included. Descriptive and psychometric information regarding the measures is reported in chapter four. It must be noted here that the data gained from the measures do not lend themselves to statistical analysis or comparison; no power calculations or sample size homologation were appropriate or necessary for this study. Rather, the purpose of using the measures in this study was to provide further context to the participants' experience. These data are then used to differentiate between participants' experience of apathy by using individual items to show clear differences in how participants reacted to some situations. Chapter seven then continues some of this discussion.

5.1.1 P1 – Mary:

Mary is in her mid-60s, white British, and was a nurse before retiring when her symptoms of HD became unmanageable with work. Mary is married with several children and grandchildren and now lives in a care home. Her husband is also unwell and lives in a different care home, which causes Mary great distress. Some of her family live close and visit when possible, while some of her family live abroad. She maintains contact with some friends but sees them rarely because most still work, and Mary's health problems make communication difficult. Mary finds walking very difficult and uses a wheelchair (although she does not like it) when she leaves the care home. During the relatively short 16-minute interview, her posture became increasingly slumped, with a pronounced arch in her back becoming worse. Mary's speech is quite badly impaired and becomes more difficult to understand when she is upset or stressed. Similarly, she seems to suffer from increased rigidity and muscle spasms when stressed. Mary's cognitive ability was also impaired but did not seem to be as pronounced as her deteriorating physical function. She found some of the questions difficult to follow, particularly if there were more than one clause, and most of her answers were extremely short. This was likely due to a combination of the physical effort it seemed to take for her to speak, the frustration at having to repeat things if the interviewer did not understand, and a general paucity of thought that left her unable to think of anything to say. She also struggled with the meaning of some words. Initially, Mary said that she did not understand what the word 'apathy' meant but then said she did after a short explanation.

The interview with Mary was the most difficult of the eight interviews, both methodologically and emotionally. Mary became very upset throughout the interview and struggled to give any definition of apathy or talk for more than five words without verbally stumbling or breaking down in tears. The researcher suggested a definition of apathy (as laid out in the interview schedule when a participant said they did not understand the word) and added an anecdotal definition; Mary answered 'yes' to the definition when asked if it made sense but made no further comment. During the short interview, apathy was revisited but Mary did not have anything to add. Due to her physical limitations, Mary has a professional carer (one of the members of staff from the care home) with her almost all the time. Throughout the interview, Mary's carer that day, Clare, was present and helped to console Mary when she became upset.

It was a particularly stressful time for Mary, which may have worsened her comprehension, speech, and mood. A few days prior to the interview, Mary's best friend in the care home passed away. Mary was generally unhappy about living in a care home and being away from her husband. Most of the other interviews were around 40 minutes in duration, but the interviewer stopped the interview after 16 minutes as Mary was too upset to continue. Mary wanted to continue, but the interviewer did not feel it was in her best interest to continue and, additionally, she had probably said as much as she was able about apathy. After the Dictaphone was switched off, the interviewer stayed with Mary to debrief and ensure she was not overly distressed by the process. The researcher was upset by how distraught Mary seemed by her living situation and dwelled on the interview for some time afterwards. This was discussed during the next supervision meeting and Mary's care team were immediately informed that she became upset during the interview (in line with the protocol).

Mary seemed depressed during the interview. She was mourning the loss of a good friend, as well as the separation from her husband, and found the general lack of contact with her family difficult (her family did try to see her as often as possible, but Mary felt lonely without daily contact). Her health did not seem to be a major cause of sadness, but of frustration. She did display some symptoms of apathy, including not wanting to get out of bed, a lack of interest in anything, and poor motivation, but these could also be signs of depression. In the researcher's opinion, Mary was suffering from both apathy and depression, concurrently, and it was difficult to pull them apart. Without the loss of her friend a few days before, it seems likely that Mary would have seemed more apathetic than depressed; the loss seemed to worsen her symptoms of depression and emphasised her sadness.

5.1.2 P2 – Daniel:

During the briefing for his interview, and before the Dictaphone was switched on, Daniel asked what apathy was and the interviewer responded with the wording laid out in the semi-structured schedule. Daniel responded that he had experienced apathy before but did not anymore. As the consent form had not been completed while this conversation occurred, this brief interaction does not appear in the interview transcript. Additionally, as Daniel had already stated that he did not know what apathy was, he was not asked to define apathy during the interview. Instead, the interview focussed on eliciting as much detail as possible from Daniel in terms of his daily activities, general well-being, and following the few lines of conversation he initiated.

Daniel is in his mid-50s, white British, and used to work as a paramedic for the ambulance service. He stopped working in 2012, five years prior to the interview in 2017, when his symptoms related to HD became too severe. Daniel lives on his own but regularly sees his partner. His sister, who has also tested positive for HD but is not symptomatic, lives over 250 miles away but visits at least twice a year.

Daniel had pronounced chorea characterised by frequent erratic movements during the interview and had some difficulty speaking, particularly at any length. He was also having some breathing issues that made his speech difficult to understand at times. Having met Daniel before and meeting him again after the initial interview, almost everything was able to be transcribed. Occasionally, Daniel made small noises that sounded like he was saying 'yeh' but were him catching his breath in his throat or gasping slightly. Daniel struggled to talk about his emotions or feelings, possibly due to a combination of his cognitive difficulties due to HD, his general personality, and the interview being an unusual situation. It seems likely that Daniel does not talk about his emotions or feelings on a regular basis.

During the interview, Daniel tended to answer questions with brief responses and after a small pause. It was very difficult to engage him in any discussion of his experiences or feelings. The interview itself was difficult to conduct; unlike the interview with Mary, it was not emotionally difficult, but Daniel's short answers left little room for development. Additionally, he was happy to sit in silence while the interviewer left long pauses. This is relatively unusual in interviews, with most participants feeling something akin to social anxiety and needing to fill the silence to avoid any awkwardness; this was not the case for Daniel. This general paucity of reply was probably a function of several factors that will be further discussed below. Daniel did talk more openly and at much greater length than any other topic about his family history of HD, specifically his paternal grandfather, father, and sister. Perhaps this also points towards a degree of memory and personal insight problems in that he can talk about other people, particularly in the past tense, but not about the present or recent past, including his own problems.

Daniel did seem to suffer from a degree of apathy, mainly characterised by a lack of insight and lack of interest. He claimed to have suffered from apathy in the past but had not done for some time. When pushed to explain the difference between then, when he experienced apathy, and now, where he was not apathetic, he could give no differences. With an appropriate level of insight, Daniel should have been able to come up with a few differences, such as he is interested in hobbies he used to have now or seeing friends. The most difficult aspect of Daniel's presentation was that he was seemingly content doing relatively little; this is perhaps symbolic of apathy in general in that, by its nature as a problem that causes severe emotional blunting, suffering from apathy does not cause grief. Interestingly, Jean does seem to experience a degree of grief, possibly because she still also has insight into her apathy, an interesting comparator to Daniel's experience.

It should be noted that due to his short answers, the interviewer did ask some leading questions while trying to elicit more responses from Daniel. Some of these leading questions appear as quotations in the text and caution has been taken when drawing any conclusions from these data.

5.1.3 P3 – Jane:

Jane is in her early-40s, white British, and has worked in various places including a hospital and a horse-riding school. She does not work currently due to her worsening HD. She lives with her young daughter and her ex-partner (her daughter's father) lives around 100 miles away but visits frequently. She has other family and friends within a few miles.

Jane's interview is unusual in that she talks a lot, often without prompt. In the transcript, the interviewer does not ask a question until a couple of minutes into the audio. Keeping Jane on-topic was reasonably difficult as she seemed keen to talk about the things on her mind rather than necessarily what the interviewer was asking. This was perhaps a coping mechanism as Jane seemed to struggle with thinking and often talked over the interviewer. Maintaining her own conversational flow may have been easier for her, rather than following the interviewer questioning. Jane always apologised for speaking over the interviewer, but it reminded the interviewer of people with hearing problems or social anxiety who talk over others to keep control of a conversation.

The transcript starts with Jane already talking about apathy before the first question has been asked (to such an extent that the interviewer rushed to turn on the Dictaphone). The difficultly keeping Jane on one topic became pronounced when she became fixated on a few anecdotes. On two occasions, she became agitated when recalling other people being impolite (queue-jumping) or exhibiting dangerous, aggressive behaviour when around children.

Jane showed very few physical symptoms of HD. When walking around her house, she was relatively slow and cautious in her movements, but showed no chorea or muscle spasms. She smoked a few cigarettes during the interview and was dexterous enough to roll her own and smoke them without any trouble. She frequently had to relight the cigarettes when they went out because she had either forgotten about them or had been speaking for too long. Jane did complain of several problems related to HD, usually behavioural or emotional problems such as anxiety, anger, or apathy. Of all the participants, Jane was the most aware of problems such as apathy and was keen to avoid her daughter noticing Jane's apathy but struggled to control her behaviour.

Jane seemed to describe her condition reasonably well and did seem both apathetic and anxious, as well as occasionally angry. Her insight into her behaviour was superficially good, but she seemed unable to control those emotions and behaviours sufficiently. Her insight also seemed compromised in that she could identify problems but was then unable to think of solutions or ask for appropriate help. She seemed to exhibit normal behaviour, such as being angry when her young daughter did something naughty, but then excessively dwelled on both her daughter's behaviour and her own reaction to that. Her emotional responses seemed dulled to the extent that she was performing emotion rather than feeling it; these are perhaps all signs of apathy.

5.1.4 P4 – Jean:

Jean is in her late-60s, white British, and was a schoolteacher before retiring due to stress. Jean attributes the stress that caused her to retire to several changes in school management and increased pressures on the teachers. Jean has three children, two of whom visit Jean regularly, while one lives in east Asia. Jean was married and now lives alone.

The interview was relatively long and was much more ordered than the other interviews; Jean seemed to be able to hold her short-term memory better than other participants, perhaps demonstrating less advanced decline related to HD. Jean did not complain of apathy, but was reported by her clinician to have spent weeks at a time in bed without getting up. She did not mention this during the interview, even when asked specifically about any apathetic feelings. Jean did not seem to think she was currently or had suffered from apathy, and perhaps did not remember the time when she was essentially bed-ridden.

When interviewed, Jean was recovering from a serious infection which resulted in her being hospitalised for several weeks. This also coincided with a planned holiday, which she was forced to miss. Jean's mobility was not particularly good, but Jean and her family made alterations to her house (such as levelling the floors) to ensure that she can get around safely. She has several friends close by who she goes out with sporadically and recently used to attend 'keep-fit' classes with one friend. She was waiting until fully recovered from the virus before returning.

Jean exhibited few symptoms of HD. She did complain of a few problems, mainly around mobility, but stated that most of them were manageable if she had enough time to organise things. Jean's mother had HD and followed a different symptom-pattern to Jean, and this was something that seemed to prey on her mind; Jean is waiting to see what her symptoms will be like and if she will take after her mother. Jean did not want to emulate her mother who seemed to have problems with anger and isolated herself.

Jean did not seem apathetic and was able to talk about the behaviours that may have seemed apathetic and explain why they happened such as being bed-ridden due to the infection. After talking to Jean's consultant (who is also part of the PhD supervisory team) after the interview, there seemed to be a longer period where Jean stayed in bed without the underlying reason of being ill, but Jean did not mention this during the interview. It seems plausible that Jean suffered from a period of apathy, but this passed without Jean recognising it as such. During the interview, Jean does talk about apathy, defining it as a form of tiredness where one is unwilling to do anything, and perhaps this is when she suffered from apathy; she felt tired and disinterested, hence staying in bed, and this may have been a period of apathy. Jean may not have remembered her period of apathy or hidden some details from the interviewer and not wished to talk about feeling apathetic but given how open Jean was about the rest of the topics covered in the interview, it seems relatively unlikely that she hid her feelings. Her consultant also says that Jean reports not having experienced apathy.

5.1.5 P5 – Sylvia and William:

Sylvia and William are both in their early-40s, white British, and not currently working. Sylvia used to work in a factory but stopped in around 2011 when she became too unwell with HD. William stopped working soon after this to care for Sylvia. They used to be married and now live together; William is Sylvia's full-time carer. They live close to friends and relatives who they see frequently. They look after two young children for a few days every week, something they both look forward to immensely.

Sylvia and William described Sylvia's physical state as very good since she started on B12 injections to treat anaemia. Before Sylvia had injections of B12 every three months, she seemed to be very lethargic. Since she has had the injections, both describe a huge increase in her activity levels, although she still seemed to be doing relatively little. Sylvia found it difficult to follow some of the interview, particularly when asked questions comparing her current physical or emotional state with a previous state. This seemed to demonstrate some cognitive decline. She became angry and upset when recounting some stories about work-colleagues or when William said something she disagreed with and seemed happy when talking about the young children they look after, but otherwise expressed few emotions. Her answers to questions were mostly brief and she looked to William to help her with questions. William would only interject when Sylvia asked him to and there were a few occasions when William's version of events would differ from Sylvia's; this confused (and occasionally angered) Sylvia who then struggled to keep up with the conversation and would contradict William or ask him to clarify what he meant.

Sylvia seemed apathetic in some ways and seemed, like Daniel, to exhibit some behaviours that could be a psychological reaction to HD that exhibited as apathy (e.g. losing interest in things because they were more difficult), and some apathy related to the cognitive decline associated with HD (e.g. confusion). This conflict, between apathy induced by a neurological impact and apathy induced by a psychological reaction, will be discussed later in the thesis. She found it difficult and confusing to talk about what she liked to do and her reactions to most questions were blunted, pointing towards her being apathetic.

5.1.6 P6 – Philip:

Philip is in his late-40s, is white British, and lives alone with his small dog who Philip speaks to in Danish. Philip was born in England and spent several years in Denmark where his two daughters grew up. He then returned to England with his daughters for them to attend further education in 2013. On his return, Philip lived in London where his daughters were at college. A couple of years ago, when Philip's symptoms related to HD worsened, he came to live near Hull to be closer to his brother – Philip and his brother see each other most weeks. Recently, however, Philip's increasing anxiety has reduced the amount he feels capable of going out. Previously, the two of them would go out every week to walk his dog and go to social events at a pub or music club. Philip was physically unsteady during the interview (he stood up a few times to get dog treats from the kitchen), although manages without any aids and did say that he takes the dog out walking with his brother when he feels able. Recently, his physical dexterity and social anxiety seem to have worsened and he takes the dog out walking less and less – this seems to be a catch-22; as his physical ability worsens, so does his social anxiety and the two seem to have become entwined. Philip describes social activities, including this interview, as being 'very strenuous' and this seems to be where a lot of the social anxiety stems from; for instance, Philip described being nervous about handling loose change when shopping and feels that his concentration and cognitive ability impairs his ability to perform otherwise simple activities of daily living. These symptoms have been getting worse steadily over time, in-line with what may be expected because of HD.

During the interview, Philip was distracted throughout as his dog was frequently barking. When the dog barked, Philip would talk to it and try to calm it down. Mostly, this did not work, and Philip would give the dog a treat or some food to try and keep it quiet. Unfortunately, this seemed to encourage the dog to constantly bark and be rewarded for doing so. Philip said that maintaining concentration during conversations was difficult for him and that this often led to him being confused. He mostly followed the discussion during the interview but seemed nervous, particularly when his attention was divided between the dog and the interviewer.

Philip seemed like Jane in that he was both apathetic and anxious, although exhibited both in a different manner. Like Jane, Philip was able to describe himself with a good degree of accuracy as anxious in social situations (he did seem anxious during the interview and said this) and also described himself as apathetic towards other people's problems because of the severity of his own problems. This definition of apathy is, in the researcher's opinion, closer to perhaps anger than apathy, but is an understandable definition centring around a lack of empathy. Philip describes a lack of interest in things he used to like to do, and an inability to do some social events that he used to be able to do. These seem to point towards a degree of apathy but combined with anxiety.

5.1.7 P7 – Emma and Liz:

Emma is in her mid-20s, and Liz, Emma's mother, is in her early-50s; both are white British. Emma has been symptomatic with HD for around five years. Emma's father also lives in the house and has advanced HD. Liz is a full-time carer for Emma and her father. Emma has an older brother who lives away from the family home.

Emma used to work in retail but had to stop when her symptoms worsened. She now uses a wheelchair most of the time. During the interview, she was very quiet and softly spoken, often turning to Liz (who remained for the entire interview) for advice or help answering questions. Emma's answers were usually monosyllabic or very short. It was very seldom that Emma would elaborate on any answers, focussing on directly answering the question. Liz would prompt Emma about a certain topic, and this would elicit some detail about recent activities for approximately half of the questions. For the other half, Liz would provide the detail that she had been probing Emma towards. When unprompted, Emma visibly struggled to come up with any detail, although did occasionally. She was excited by a few topics of conversation, particularly when talking about football or some activities that she did with a carer (including cycling around the local park), and this excitement tended to elicit more detail and longer answers. She seemed to find it difficult to think or talk about her emotions and questions that asked her to consider how she may have changed over time confused her. In general, the longer the question, the more Emma struggled to understand and answer, in a similar fashion to Sylvia. These elements of confusion seemed to demonstrate at least some decline in cognitive function.

The bond between Emma and Liz was clearly loving and both wanted to help the other. Liz prompted Emma frequently and, prior to the interview, told the interviewer that this would probably happen due to Emma's quietness combined with her generally reserved nature and consequent struggle to answer questions. Liz did, however, let Emma think and try to answer questions before offering prompts.

Emma did seem apathetic during the interview, but it seemed to be mostly due to her cognitive deficits related to HD. When prompted by Liz, Emma was able to talk about things she enjoyed (watching football, doing novel thing with her professional carer), but unprompted, she found it very difficult to think of a reply to a question that was not a simple yes or no. Liz described being able to put Emma in front of the television and Emma would watch it, without question. Emma was not actively watching television but would do so out of habit and because it was there. This did not seem to bother Emma and seemed to be one of the best descriptors of what it is like to experience apathy related to cognitive decline.

5.1.8 P8 – Sophia and Luke:

Sophia is in her early-70s, is white British, and lives with her husband, Luke, who is in his late-60s and was present during the interview. Sophia was a seamstress but retired and has now lost interest in it altogether but is cajoled by family to make or alter things occasionally. Sophia and Luke's son lives very close with his partner and two young children. Sophia and Luke's daughter passed away suddenly from a blood clot and is buried around 150 miles away. Sophia and Luke moved nearer to their son when Sophia's health deteriorated, and this meant moving away from their daughter. They have only recently moved to their new house, but Sophia is active in a local church group and they seem active socially. Luke has prostate cancer and was recently given a five-year prognosis, and this was discussed briefly during the interview.

Sophia was able to talk at length about her experience of apathy and other problems, as well as more general issues. She did, however, find it difficult to keep on one topic and would wander between topics as they occurred to her, often mid-sentence. Luke would interject occasionally to put her back on-track if she forgot, but mostly Sophia continued until the interviewer intervened or asked another question. Sophia was a bit unsteady when walking and uses a mobility scooter to get around the local area. She feels that she is more cautious and impatient with people than a few years ago. As the interview progressed, Sophia became increasingly agitated and had to make increasingly frequent visits to the toilet. After around 40 minutes, Luke quietly suggested the interview should pause and continue another day. Sophia seemed somewhat relieved at this suggestion and so the interview was conducted over two sessions. This nervous reaction to unusual situations seemed to be a relatively new phenomenon for Sophia.

Sophia seemed to exhibit some elements of apathy and was increasingly disinterested in things which were becoming more difficult for her to do due to the symptoms of HD she was experiencing. In relation to housework, Sophia let Luke do most of those tasks because he was able to do it quicker. She seemed to struggle with this slightly in terms of feeling guilty about Luke doing the majority of the housework, but almost seemed pragmatically apathetic about it. Sophia was herself unsure if she suffered from apathy; the fact that she could have this debate with herself suggests that she still had good behavioural insight, like Jane and Philip, but was also incapable or unwilling to change.

5.2 Measures data:

Table 4, below, shows the results of the three validated measures used with each participant. Unfortunately, due to a family bereavement, Emma was unable to complete the measures. The three measures used were the Apathy Evaluation Scale-Clinician (AES-C; Marin et al, 1991), the Lille Apathy Rating Scale-short form (LARS-SF; Sockeel et al., 2006 and Dujardin et al., 2013), and the Dimensional Apathy Scale (DAS; Radakovic and Abrahams, 2014).

A few things must be noted here. First, these measures were completed with a carer present in the cases where a carer was also interviewed (Sylvia and William, and Sophia and Luke). The researcher tried to ensure that the participants did not confer with their carer too much, although this was usually difficult to maintain. It is possible that participants changed their answers because their carers were present, but this did not seem to be the case as both couples disagreed on some points throughout the interview and were not coy in doing so, as mentioned in the previous chapter.

Second, these measures were chosen partly because they were a mix of self-report and administered by a clinician. This allowed the measures to assess both participants' insight into their state of mind and behaviour and the researcher's interpretation of the same. All three measures were administered at the same time, one after another, and the sessions audio-recorded in order to allow for re-scoring afterwards if the researcher needed more time to deliberate an answer. While the researcher is not a clinician, the same assumptions were used at each juncture when administering the measures to try and standardise the answers across the participants, particularly in the AES-C where clinician-judgement is relied upon relatively heavily.

Third, none of these measures has been validated for use specifically in HD. The DAS has been validated for use in various conditions but not HD, the AES-C was validated for general use although specifically with patients with motor dysfunction, and the LARS-SF was validated for use in people with probable (rather than diagnostically confirmed) PD. Here, the measures serve as a guide that, when combined with the data from the interviews, provide numerical context for how each participant experienced apathy in HD and the results of the measures are discussed further in the next section. The measures also provide a useful way to think about the definitions and conceptualisations of apathy that each measure assumes; for instance, if a patient seemed apathetic when interviewed, but did not appear apathetic when analysed with the measures, this provides a useful talking point to evaluate both the measures and the researcher's own assumptions about what constitutes apathy that bolsters data from the systematic review in chapter two.

A brief description of the results of the measures is presented below and a further discussion of these results is included in chapter six.

Table 4: Results from the three measures

Participant	**AES-C**	**LARS-SF**	**DAS**			
			Exec	**Emo**	**B'our/Cog**	**Missing**
P1 – Mary	47	6	10	6	18	6
P2 – Daniel	37	2	6	11	15	3
P3 – Jane	28	-2	22	7	14	0
P4 – Jean	35	-12	6	10	15	0
P5 – Sylvia	52	-3	17	14	19	0
P6 – Philip	50	1	24	13	20	0
P7 – Emma						
P8 – Sophia	42	-8	13	11	10	0

5.2.1 Apathy Evaluation Scale-Clinician results:

When using the AES-C, there are a few important assumptions a user must make, as detailed by Robert Marin in the AES guidelines. First, that apathy is a "pathological construct" (p8, AES guidelines), meaning the measure views apathy as part of a medical disease, and second, leading from the first assumption, that the AES-C is a relative scale meaning the higher the score, the more a person is suffering from apathy, although there is also a cut-off, below which a person is deemed to be not suffering from apathy. Marin reports that mean scores of 26-28 (depending on the type of AES used) from healthy individuals is normal. This suggests a cut off score of 39-41, (+2 standard deviations). Marin suggests that, for a population over 60 years old, a cut-off of 42 may be appropriate, Throughout, however, Marin stresses that formal recommendations of cut-off scores are inappropriate. The relative performance of each participant will be discussed, particularly in relation to their scores on other measures, interview data, and perception of the researcher when interviewing each participant. The AES contains three subtypes of apathy, cognitive, behavioural, and emotional, although Marin explicitly describes apathy as a psychological dimension, singular, and therefore a uni-dimensional construct.

Sylvia and Philip scored the highest on the AES-C, while Jane scored the lowest. Bearing in mind the suggested, informal cut-offs Marin reports, Jane therefore appeared to be within a normal range, while all the other participants trended towards being apathetic. This matched closely with the researcher's interpretation of participants' presentation. Daniel and Jean appeared to be somewhere in the middle, although this was perhaps due to a lack of insight on Daniel's part and problems with the measure that seemed to inflate Jean's apathy because of her relative lack of behavioural interaction. The researcher disagreed with some of their assessments of their behaviour and emotional response, which several items of the AES-C consider, and this then led to a compromised score between the participants' answer and the researcher's assessment.

5.2.2 Lille Apathy Rating Scale-Short Form results:

The LARS-SF, similarly, uses seven dimensions (everyday productivity, interests, taking initiative, novelty seeking, motivation/voluntary actions, emotional responses, and social life) to describe apathy, but sums all of these to give an overall 'apathy score'. Using this interpretation, the LARS-SF implicitly makes apathy a uni-dimensional concept (although Dujardin does discuss multi-dimensional apathy in the development of the LARS). For the

LARS-SF, a total score of minus seven or above is the cut-off for diagnosing apathy (Dujardin et al., 2013).

Jean scored by far the lowest and therefore least apathetic on the LARS-SF and Sophia also scored low, below the cut-off. All other participants were above the threshold and therefore apathetic. The LARS-SF was particularly sensitive to how quickly participants responded, which explains why Mary, with her severe speech impediment, scored so highly. Although Mary also seemed apathetic (as well as potentially depressed), her high score was accentuated by her delayed responses, in large part due to her physical limitations. The LARS-SF was partially chosen because, although it was not validated in HD, its validation in PD was assumed to mean that it would be sensitive to physical limitations. This, however, did not seem to be the case; while the physical limitations in PD and HD are diverse, there are some similarities (mainly in terms of a lack of control over one's movements and a reduced ability to perform physical activity), but the LARS-SF does not appear to be well-suited to the needs of people with apathy in HD.

5.2.3 Dimensional Apathy Scale results:

The DAS does not suggest cut-off scores as it assumes that apathy is made up of several sub-types of apathy (executive, emotional, and behavioural/cognitive sub-types). Each sub-type is summed to give an overall score for each type, the results of which are presented above. It is true to say that the higher the score in each sub-type, the more a person suffers from that sub-type of apathy. The DAS seems to allow for a slightly more nuanced assessment of participants' apathy, perhaps unsurprising given the triadic, multi-dimensional assessment.

Mary and Daniel struggled to answer some of the DAS items, Mary unable to answer six of the 24 items and Daniel three. Sylvia, Philip, and Sophia seemed to struggle across all three sub-types of apathy, while Jane struggled with executive function and less with emotional and behavioural/cognitive. Jean seemed to be the opposite to Jane, struggling with behavioural/cognitive and less with emotional and executive.

5.2.4 Comparison of the measures:

Table 5, above, shows the participants' scores ranked in relative order of least to most apathetic (going from seventh to first, as Emma was unable to complete the measures). No participant comes out as most apathetic on all measures. The most consistently apathetic, according to the measures, were Sylvia and Philip, who both scored most apathetic on two of the five scales (when counting the DAS as three scales). Jean was perhaps the least apathetic across the scales, scoring towards the lower end on three of the five scales, although Jane also ranked low across several measures. Mary and Daniel were slightly difficult to judge as they were unable to answer some of the DAS items, which had the effect of lowering their scores as they were given a zero for those items. What is most striking, however, was the lack of nuance that these measures provide. The DAS was certainly the most sensitive, overall, largely because of its multidimensional set-up. The LARS-SF was difficult; it gave the best subjective feel when administering it as it allowed for consideration of how difficult the participants found it to answer the items as well as how they factually answered the items. It was, however, not sensitive enough to the physical limitations of symptomatic HD (with the important caveat that it was not designed to allow for HD). The AES-C seemed to rank the participants closest to how they presented in the interviews, although seemed to struggle to account for a lack of insight. Daniel seemed more apathetic during the interview (unable to talk much about his interests or what he did on a day-to-day basis) than the AES-C accounted for, and his rank of second on the LARS-SF was probably closer to his level of apathy relative to the other participants.

Table 5: Results ranked, least to most apathetic (7th-1st)

Participant	**AES-C**	**LARS-SF**	**DAS**			
			Exec	**Emo**	**B'our/Cog**	**Missing**
P1 – Mary	3	1	5	7	3	6
P2 – Daniel	5	2	=6	=3	=4	3
P3 – Jane	7	4	2	6	6	0
P4 – Jean	6	7	=6	5	=4	0
P5 – Sylvia	1	5	3	1	2	0
P6 – Philip	2	3	1	2	1	0
P7 – Emma						
P8 – Sophia	4	6	4	=3	7	0

6. Chapter Six – Data analysis and discussion

This chapter presents detailed analysis of the interviews with participants across three themes and demonstrates how the participants' achieved different scores on the measures of apathy. Finally, a novel interpretation of apathy is introduced where two different types of apathy are suggested.

6.1 Interview themes:

The themes presented below have been laid out to develop the story of how participants defined and experienced apathy, and how those experiences are reminiscent of the themes in the literature and theory surrounding apathy. It should be noted that there is often overlap between the themes, reflecting how apathy seemed to affect every aspect of participants' lifeworlds. A common theme running through all the participants' experiences was the feeling that apathy had caused a significant change in their life. For some, this was relatively simple; there used to be hobbies or activities they used to enjoy doing that they no longer did and they did not seem to be concerned by this. For others, the change was more fundamental and they struggled to reconcile their new, unwanted lack of desire with their previous identity. Those that struggled with this change were caught in a state of emotional turmoil where they were confused and, in some cases, embarrassed by their changed identity.

6.1.1 Theme one – Inauthenticity and the apathy-self:

Participants struggled to define apathy but all except Mary gave a short description of what they understood by the term, as shown in table 6 (page 137). None of the participants' defined apathy in the same way but did describe some similar experiences when it came to describing apathy. The impact apathy had on people's lifeworld seems to create a problem of identity characterised by a mismatch in people's ideas of themselves, what they wanted to do, and who they expected to be. This could be characterised as two separate beings; an apathetic-self and an authentic-self, similar to Heidegger's inauthenticity and authenticity (see chapter four). Heidegger suggests that many behaviours associated with a social existence are aligned to an external 'other', which he characterises as inauthentic and aligned with a they-self, something outside of our control. When looking at participants' experience of apathy, however, some seemed to experience a strong apathy-self that was different from their view of their authentic selves. Sometimes, the apathetic-self would win and the person would demonstrate behaviours that seemed representative of apathy, whereas at other times, their authentic-selves would shine through, temporarily banishing apathy. Their they-self, an external 'other', had become an internal part of themselves, but one that they did not recognise. This is not to say that participants were not influenced by inauthenticity, that the they-self does not exist for them, but rather that apathy's impact on their lives was similar in that it felt external to their true selves and was not a representation of who they wanted to be. By attributing their experience of a changed identity to apathy, participants internalised the they-self, morphing it into an apathy-self.

Jane was perhaps most emblematic of this struggle with the apathy-self and her personality change since being diagnosed with HD and subsequently experiencing apathy. Jane recognised that her behaviour had changed since she began experiencing apathy. She was no longer as emotionally reactive and found herself having to remember how she would have wanted to feel and attempting to feel this. Some of her emotions were no longer automatic but required Jane to try and actively trigger them:

"…it just becomes I don't know, I just- I'd probably say 50% more numb and that's how it feels. It's not there anymore, that isn't there anymore. So, it's like that feeling it's not as intense or it's not erm-."

Equally, however, Jane recognising her changed emotional state seemed to be a catch-22 situation. Every time she recognised a new way of feeling, emotionally, she attributed this to the disconnect she experienced with apathy, but also recognised that maybe it was a normal part of the experience. She describes becoming impatient with her young daughter but willing herself to be more patient:

"So, it's just like and I just have to keep reminding myself I've got to be more patient, I've got to be more patient, because I feel bad personally if [.] but you do as a parent anyway. You're- you're constantly just always feeling bad."

Jane attributes this impatience to her apathy-self. She seems to think that, if it was not for her apathy, she would be more patient automatically. Yet Jane is a single mother with a young child and describes situations where it would be reasonable to be impatient. Without the label of apathy, the impatience Jane describes would be reasonable. One could attribute the impatience to normal parental feelings, as Jane tries to halfway through this quotation. She seems to be trying to convince herself, more than her audience, that it's normal to be impatient with a young child, but still says that she constantly feels bad. She cannot seem to shake the feeling that it is her apathy causing her to be impatient, that she should not be feeling impatient, pathologising her experience of parenthood. She obviously wants to be a good mother, but feels that she is being held back by her struggle with her apathy-self. Where parents could normalise impatience as part of being a parent, Jane internalises this impatience and worries that is because of her apathy-self. Another possibility, however, is that attributing this impatience to her apathy-self rather than her authentic-self is a defence mechanism. Perhaps she would have been impatient even without the label of apathetic, but with the label, she can attribute her impatience to apathy, not her own self.

This possibility appears again later in the interview when Jane talks about her struggle when watching the charity television programme, Children in Need, with her sister. She knows that she used to get upset watching some of the intentionally emotionally moving parts of the programme, but struggles to connect with those feelings now:

"I give money to Save the Children every month cos I know, you know, that's what I want to do, that's the person I would have been and would have wanted to do but- but I don't necessarily all the time have the same feeling about things like that if you understand what I mean."

Jane expresses a wish to go back to how she used to be, possibly before she became symptomatic with HD and struggled with her apathy-self. Jane is willing to give charitably but instead of doing it from choice, she is doing so out of a sense of duty to her former authentic-self, not because she necessarily feels like doing it. This is not necessarily abnormal; many people perform actions that they feel are necessary to maintain their social façade rather than because they feel like doing it. As part of relatively normal behaviour, however, it is usually a passive process that does not require special effort or thought and is not then the subject of rumination, as it is for Jane. For her, however, this feeling, of trying to reclaim a former self and still appear emotionally involved, is an extremely active, conscious effort that she dwells upon. Her use of the perfect conditional tense in the middle of the sentence ("that's the person I would have been…") is only missing the "if" and continuation clause. Perhaps Jane thinks that she would be different and would perform those charitable behaviours automatically if it were not for the HD and apathy, but this is impossible to know. It is interesting that Jane uses the present tense when describing this at first but corrects herself and moves to the conditional. This perhaps demonstrates some of her struggle with apathy amid her changing personality. A lot of the things Jane says demonstrate this struggle of her changing, or at least changeable, personality. Jane, in her opinion, used to behave differently and she wishes to return to that state. This may simply be because she understandably wishes to return to a time when she did not have HD but seems deeper than that in that she also wants her personality to revert to her previous, charitable self. Jane's idea of her authentic-self and the reality of her apathy-self are incompatible. When Jane attempts to revert to her preferred, authentic-self, her apathy-self tends to create barriers, causing Jane emotional pain that she feels as apathy and anxiety. Although Jane would prefer to be emotionally engaged with things such as Children in Need or her daughter's activities, her apathy-self presents a barrier, pulling that drive away from her.

Sophia demonstrates a similar struggle between an authentic-self and an apathy-self, although represented more by physical limitations that may stem from cognitive problems. Again, however, similar to Jane's complicated relationship with apathy, things are not clear. The presence of apathy is certainly a factor in Sophia's life, but so are Sophia's physical limitations, problems with cognition, and logic dictating some parts of her life. The mitigating factors of Sophia's lifeworld contribute to Sophia believing that there are elements of her behaviour that are apathetic, and these elements concern her, but she feels in some ways justified in her apathy. Sophia struggles to do housework, both physically and mentally and when talking about her difficulty in doing housework, she stated:

"I do feel guilty sometimes that Luke is doing most of the- most of the other [house] work, but erm. [.] That's very much apathy, I should say… There are probably things I could do, that I don't do. But there are also things that I might do that Luke can do in a fraction of the time. So yeh [.]. But- remembering how to do things erm [.] yes, that's- coordinating all the- all that's involved in doing things makes you slower and more erm lethargic, yeh. I think erm yes."

Sophia encapsulates here the difficulty she has in identifying her authentic-self, her apathy-self, and what she tentatively seems to think of as a more logical approach to tackling things such as the housework. Sophia and Luke employ a cleaner to come every other week, and since Luke can do the housework quicker and more efficiently, it makes sense for him to do jobs in-between the cleaner's visits. Sophia's physical difficulty, in large part because of the impact of HD, means she struggles to do housework and this puts her off doing it, hence feeling justified in leaving everything to Luke. There is certainly some tension here, with Luke later stating:

"…it's because of all these things she doesn't do, she spends a lot of time in the house when she's home asleep. In the morning she gets up, has a wash, then goes to bed. But er, for a few hours. Now I'd like to do that [sharp intake and laugh] but I've got jobs to do."

Were it not for the presence of Sophia's physical limitations caused by HD, it seems plausible that the guilt she expresses in this part of the interview would encourage her to do at least some of the housework and it is likely that this is representative of her authentic-self since in a later section she describes having done housework previously when Luke was still working. Sophia's apathy-self, however, is different to Jane's. Where Jane actively struggled against her apathy-self, perhaps because of her desire to not show her apathy to her daughter, the presence of Luke and the cleaner seem to mean that Sophia does not need to fight against her apathy-self when it comes to housework. Perhaps Sophia and Luke would have come to this conclusion anyway, that Sophia's physical limitations mean that housework is out of the question. But what seems to have happened is that, gradually, Sophia's physical limitations have increased and this has resulted in an apathy that has seeped into both Sophia and Luke's lifeworld, in some ways unnoticed and unchallenged. For Jane, her apathy was challenging; for Sophia, her apathy is not challenging in the same way. This lack of challenge, when it comes to apathy, seems to occur in other elements of Sophia's life. Sophia talks about a struggle to do any sewing, a task she used to perform with relish as a seamstress. She describes struggling with the physical demands of sewing now:

"I have some problems with holding needles and things like that so, erm, gripping things and that, erm, now I don't know if that's the Huntington's or the arthritis or both, but erm, yes, to ge- to hold a needle and to sew and to concentrate is- is a difficult thing, yeh."

Again, Sophia feels guilty that she cannot do the same things she used to do. She used to help out family and friends with sewing tasks, but now feels unable and to a certain extent unwilling. Having moved away from the people she used to help (to be closer to her children and grandchildren), she describes her guilt as lessening because people in close proximity do not know that she used to be an accomplished seamstress:

"…it's not quite as bad having moved here because the people that know that I can sew don't exist, they're a hundred miles away. So erm, that erm guilt factor has gone down because I don't have to admit that I can't do it or that I don't want to do it."

Similar to the dilemma her and Luke faced with the housework, Sophia can hide behind people's ignorance of her sewing to ignore the complex reasons why she no longer sews and the guilt that caused.

These two physical examples of Sophia's limitations in life, the housework and the sewing, present her apathy as a 'chicken or egg' problem. In both cases, it is certainly true that physical limitations caused by her various comorbidities have made the tasks she used to perform more difficult. This difficulty has latterly caused her to stop performing these tasks, and in both cases, Sophia experiences guilt associated with this stoppage. Where does the apathy occur in this timeline? For Sophia, her guilt seems to be part of her authentic-self but, similar to Jane, it is often overcome by her apathy-self, which emphasises the (almost Stoic) logic that there is no point in doing these tasks if she cannot do them as well as she used to be able. Sophia does not experience the same level of emotional turmoil as Jane, however, although hiding behind that Stoical logic does seem to be a wistfulness or longing to be different that Sophia seems to cover with laughter. At one stage, Sophia laughs at her own suggestion:

"Yes, I mean I offered you a cup of tea but I would have probably looked to Luke to make it [laughs]… Well, not necessarily so but I mean- [.] yeh."

Sophia uses humour here to cover that wistfulness. The 'probably' and the 'necessarily' are both qualifiers that, without them, would change the sentences completely. Based on the rest of the conversation, Sophia would have asked Luke to make a cup of tea (he would have done it quicker and with much greater ease) but in Sophia's second sentence, she wishes this were not the case. It is an uncomfortable reality that Sophia and Luke have found themselves in, accidentally.

In a later section, Sophia describes a struggle with pernicious anaemia that seems to be resistant to high doses of medicine. There was a suggestion that it was the anaemia that was causing her to be apathetic and lethargic:

"Cos thyroid can make you tired and apathetic, so [.] it's- they don't know, it's just- it's just me [laughs]."

Sophia's apathy could be due to a thyroid problem, but high doses of medication, that normally would at least partially help, are not making a difference. At first glance, all of the mitigating factors contributing towards Sophia's experience of apathy suggest that there is perhaps a neurological or neurobiological cause of her apathy. With all of the factors taken into account, however, perhaps it is her apathy-self, her inauthentic-self, that is pernicious. It is impossible to draw the two apart, but given Sophia's experience of HD, both are plausible and could be co-occurring. Indeed, apathy does not seem like an unreasonable response.

Philip struggles with what he wants to do in a similar way to both Jane and Sophia, often describing scenarios where he avoids doing something in case his symptoms of HD cause him difficulty. His relationship with an apathy-self is different, however. Philip says he is often socially anxious because of the unpredictability of his symptoms of HD and his ability. There are things that he actively avoids doing, despite wanting to do them, because of this conflict:

"I'm doing less now than I used to, you know, it's a bit of a- it's a struggle for me to get out of the house and go shopping, you know… I'm sort of afraid of making a fool of myself getting out, because I can't work out money and stuff like that in shops…"

Philip experiences fear of multiple problems that result in him not leaving his house. His brother, who lives close by, helps Philip walk his dog and go out once a week and invites

Philip out to social events but these are often overwhelming. Philip has experienced panic attacks in the past and is concerned about triggering them again, and also struggles in busy social environments:

"…sometimes I can't face up to an evening where I've got to- you know, the noise and all those people and people, so I say to my brother I can't manage it tonight. It's quite often I do that actually."

For Philip, his assessment of his behaviour means that he often avoids any potentially difficult situations. He has found shopping difficult in the past, particularly counting money, so tends to avoid it. He has had panic attacks in the past, bought on by being overwhelmed in social situations, so often avoids these. He has trouble concentrating, particularly in conversations where there are multiple people talking, so takes steps to avoid busy environments. He manages risk in his lifeworld by reducing the possibility of harm. This is an understandable reaction given some of the harmful situations he has found himself in but one that is socially isolating. It means that instead of choosing what he would like to do, he predominantly decides not to do things in case they cause him harm or anxiety. This manner of behaving, or rather of not behaving, is perhaps at odds with how Philip describes apathy:

"…not caring about other people, know what I mean? Not caring about myself."

If Philip did not care about other people or himself, perhaps his social anxiety would not be so severe that it stopped him from enjoying shopping or socialising. Philip's inauthentic-self and apathy-self do not seem as inextricably linked as Jane and Sophia's. For them, they struggled with their inauthentic-self that was a representation of their apathy, but for Philip, his inauthentic-self and behaviour (mainly characterised by an avoidant coping strategy for fear of negative outcomes) is perhaps linked closer to his social anxiety. Philip does not want to worry about his ability to count change in a shop and finds it frustrating that he cannot follow conversations or television programmes anymore. The way he describes apathy, as a state of not caring about anything or anybody, including himself, sounds like a potentially desirable state for Philip.

For Philip, he is unable to be true to his authentic-self or even consider what that may entail because of the difficulty in not knowing how he or his body will react combined with the social anxiety he often experiences. Philip's apathy-self sits somewhere between his authentic- and inauthentic-self. He wishes he did not care about things (his definition of apathy) as a way to regain some of the confidence he associates with his former authentic-self, yet acknowledges that this lack of caring is undesirable. The way he now leads his life is almost entirely inauthentic in that he avoids potentially dangerous or problematic situations in order to reduce the potential harm he comes to. He has lost touch with his authentic-self in his struggle to cope with HD, and apathy has further muddied his capability, leaving him confused about who he is and what he wants. He is frustrating and confused by this state of affairs and seems to be trapped in a state of limbo, unable to move forward or backward and so staying still by using a coping strategy that relies only on avoidance of harm.

There are three versions of apathy-self that Jane, Sophia, and Philip demonstrate here, but all represent a disconnect between who the participants used to be, and who they are now. Jane struggles with apathy but is unsure if her feelings of apathy and her experience of emotions are normal, or if her apathy-self has dramatically changed who she is. She seems to wish that she could behave differently, how she used to behave, but recognises that her old way of being may not be who she is anymore. Sophia is unsure where the line is between her experience of apathy and her (and Luke's) rational assessment of her own ability. This confusion seems to further entrench her feelings of apathy; she seems resigned to her apathy-self, particularly in the face of worsening physical symptoms of HD that decrease her physical capability. Philip's experience of apathy is characterised by not caring about other people or himself, yet his experience of social anxiety suggests he does care about other people and wishes he was as capable as he used to be. This presents apathy as a partially desirable state for Philip, he seems to wish he could embody his apathy-self more but is held back in achieving this by his social anxiety. Yet, his love for his daughters, his brother, and his dog also hold him back from his apathy-self. This confusion leaves Philip in a state of near-constant limbo, unable to trust his body, wishing his level of caring was different. The apathy-self leaves these participants confused because they do not recognise the feelings of apathy as part of their authentic-self. They recognise that the apathy-self may be inauthentic, but worry that it is slowly becoming their new authentic-self.

6.1.2 Theme two – Apathy changed participants' emotional connection and response:

For all participants, the presence of apathy represented an identity-shift of varying degrees. The degree to which participants' identity shifted was affected by participants' varied experience, their differing personalities, and was perhaps related to their HD symptom burden. While theme one deals with participants' internal conflict between their authentic-self and apathy-self and their struggle to connect with their emotions, this theme looks at how participants' experience of emotions changed, further compounding their struggle to connect with their lifeworld.

Jane describes apathy in terms of the negative impact on her emotional capability, while Jean and Sophia describe how apathy reduces behaviour because of a lack of cognition in the form of interest or will, and Philip sees it as not caring about anyone, including one's self. Daniel explains that the way he felt when he was apathetic was due to the impact of being diagnosed with HD, immediately looking for a root cause of apathy and not seeking to define apathy itself. Sylvia describes apathy as akin to tiredness, while Emma attributes sadness and unhappiness as key experiential elements of apathy.

For Jane, apathy became wrapped up in anxiety, agitation, and frustration. She struggled to adjust to her prescribed medication and her desire to fulfil her role as a good mother for her young daughter clashed with her inability to connect with her emotions. When Jane begins to define apathy, she draws on her changing experience of emotion, thinking about how she used to emotionally care more for people, and how that seemed to have decreased over the years, both as a function of getting older and of having HD:

"J: …I don't get upset about it if you understand what I mean, it's just not- and that's one of the first things that goes. It not gone, it's just not as [.] obvious emotion you know. But the other side of that is I'm on anti-depressants and when you're on erm them because of my symptoms cos they can cause- erm- you can get erm mood swings and stuff with HD and anxiety and agitation and stuff and that's the main problems I have. And I think sometimes if you're on anti-depressants that numbs that- that side of you anyway, if you understand what I mean."

Jane defines apathy as an absence of the ability to show emotion. She is careful to correct herself and point out that, in her eyes, it is not a complete absence of emotion, but an absence of the ability to feel emotion appropriately, an important distinction for her. When watching an emotive programme that her sister was already crying at, Jane laughs at her sister. She does not get upset by the programme but does recognise that it is meant to be distressing and knows that the situation being portrayed is sad. She feels as if her emotions are being suppressed, possibly by anti-depressants, and this feels like apathy. Despite Jane's ability to think through this, however, it is still difficult for her to succinctly define. She seems to have difficulty describing apathy, her stuttering speech pattern seemingly reflective of her trying to think through her sentences but struggling. This internal conflict perhaps encapsulates the difficulty in describing an experience that is defined by absence. Apathy uses the Greek prefix 'a' denoting negation, meaning that apathy is the lack of something; Jane feels the presence of apathy because of what she feels she is lacking in her life, a difficult experience to describe. In one description, Jane's sister experienced sadness when watching the same TV programme, whereas Jane laughed:

"…things that should erm- my sister gets upset about on TV and some of the things like that like she'll cry at an advert on TV and I'll be laughing at her she'll- she's like, erm you know [.] laughing at me."

In isolation, this could just be a personality difference; one person cries at something on TV while the other laughs. For Jane, however, this exemplifies her lack of ability to feel emotion appropriately and is the main symptom of apathy. In this situation, she also seems to be stuck between wanting to be sad at the same thing her sister is sad at, but just not feeling that emotive response. As we will see later in this analysis, this situation dwells on Jane's mind; she often wants to feel more than she does, but struggles to connect with emotions that, she feels, she would have experienced before she was apathetic. This conflict, between two competing versions of herself, perhaps symbolises the conflict between Heidegger's inauthentic and authentic selves.

While Jane focussed on describing her emotional changes, Jean and Sophia focus on an inability to generate thoughts or be willing to do things. As can be seen in table 6 (below), Jean starts by defining apathy in terms of one's will or desire to do things. Apathy, for Jean, starts as a lack of will to do anything, which in turn results in inaction. This perhaps suggest that, for Jean, apathy is a cognitive deficit first, but one which is then demonstrated in a person's lack of behaviour. Jean's definition of apathy is perhaps the closest to how the literature defines apathy; representing a combination of a lack of motivation that is demonstrable by a person's sedentary behaviour. This similarity with the definitions present in the literature is particularly interesting given that Jean arguably demonstrates the least severe apathy of all the participants. Perhaps this reflects an experiential paradox; it is difficult to define apathy in a way that encapsulates people's experience of apathy. Further on in the interview, Jean added:

"It's one of those things you hear, and you know what it means but you can't put it in words. It's more a feeling than an attitude."

Jean struggled to define apathy and further expresses that difficulty here. In doing so, she reiterates her idea that apathy is predominantly an internal feeling or emotion rather than an externally visible behaviour. The externally visible part only comes after one's apathy has taken hold. Perhaps it is the former element of apathy, the personal, internal experiential part, that literature definitions miss. Later in the interview, Jean adds that apathy is:

"…just a lack of interest in things… Not wanting to go anywhere or do anything."

Again, Jean reinforces the idea that apathy is an internal struggle or lack of desire to do anything that comes from inside oneself. This becomes evident to others by a lack of activity, but as a symptom or syndrome, starts as a lack of desire or willingness to do anything.

6.1.2.2 Table 6 – People's core definition of apathy:

	Mary	**Daniel**	**Jane**	**Jean**	**Sylvia**	**Philip**	**Emma**	**Sophia**
Quotation	[None]	*Well, I don't know. Difficult to handle diagnosis. There's no cure. [.] But I'm alright now.*	*I don't get upset about [things] if you understand what I mean, it's just not- and that's one of the first things that goes. It not gone, it's just not as [.] obvious emotion you know.*	*Not being willing to do anything, so you just sit and vegetate. Not get up and go out and enjoy a nice day. But just let life roll by. Not taken an interest in life or anything that's going on around me.*	*Apathy is when you're tired all the time, isn't it? It's tiredness, mm.*	*Apathy, it means you don't care about doing things, isn't that what it means? I'd describe it as not erm caring about other people, know what I mean? Not caring about myself.*	*Like when you're feeling sad [.] and unhappy.*	*Disinterested in things, just can't be bothered to do anything.*
Comment	Did not define apathy	Description of his experience of apathy and why he thinks he felt apathetic	Has trouble engaging with emotions. Attributes some of this to anti-depressants and apathy	Defines apathy first as a cognitive deficit, then as a behavioural deficit. Lack of interest key	Unsure, but equates apathy with tiredness.	Apathy mainly a lack of caring for others and one's self.	Unsure of apathy but equates it to emotional struggles	Disinterest and lack of caring or concern

Sophia described apathy in a similar way, focussing on disinterest and a lack of interest in doing anything. Sophia described her disinterest as a normal part of her everyday life:

"Just erm- just disinterested in things, erm [.] yes. This morning I was supposed to have gone out erm [.] but I woke up and just dozed back to sleep again and thought about getting up but didn't, and that's quite normal. You just, yeh, but erm whether there was a measure of apathy interplaying in that that I just couldn't be bothered to make myself- make myself want to get up without [.]… Must be a certain amount of apathy."

This normality that Sophia defines is an interesting description and perhaps chimes with Jane's struggle to engage with her emotions. Normally, one might struggle with getting out of bed, roll over, and press the snooze button on any alarm. It might be too early, or one might not have slept well, both reasonable reasons why one would doze back to sleep without attributing this to a form of apathy. As with Jane's description of her struggle with emotional reactions, however, Sophia's description of apathy moves behind this potentially normal experience and becomes what she sees as a form of apathy. It is not that Sophia was necessarily still tired or that she slept badly, but that there is an extra step Sophia must perform in order to wake up and get out of bed. Sophia must force herself to want to get out of bed, not just force herself to get out of bed. In this sentence, Sophia knows that the normal, socially acceptable behaviour when one wakes up and has an appointment or a task to achieve is to get out of bed. But that thought, that desire, does not trigger for Sophia unless she can summon the willpower to manually trigger that feeling. As Jane struggled to manually trigger emotions that used to come naturally, Sophia struggles to trigger the will to do something. Apathy introduces an extra step in what used to be automatic behaviours. Things that Sophia used to get up for without a second thought, she now sleeps through and struggles to generate enough willpower to complete those tasks. For Sophia, that seems to define apathy.

While Jane, Jean, and Sophia struggled with feelings and behaviours that used to be automatic, Philip struggles to do anything for fear of the consequences. As described in the first theme, Philip has adopted an avoidant coping strategy to attempt to deal with his worsening symptoms of HD that have also worsened his social anxiety. In terms of Philip's emotional connection, he describes frustration at his symptoms and lack of ability to concentrate on everyday tasks, but rarely talks in overt emotional terms. This is perhaps representative of his overall emotional state; he seems both frustrated and confused by his emotional capacity. He obviously cares deeply for his family and his dog, but finds it difficult to engage with his feelings while he struggles with everyday tasks and anxiety. His everyday struggle seems to leave him feeling like he does not have enough emotional capacity to think about other people:

"Yeh I mean, yeh, I don't sort of- well- when I'm talking to other people, I can't sort of empathise with them, with their problems. I've got enough problems of my own, you know, so it's er it's difficult for me… Yeh it's getting worse. [.] Everything is getting worse very slowly."

It is not that Philip does not want to empathise with others but that he feels physically and emotionally incapable of empathising. It seems as if he is being held back, emotionally, when he wants to try and engage with the process of empathising. He finds concentrating difficult, is frustrated by his lack of physical capability, and struggles with everyday tasks. When it comes to engaging with other people emotionally, all of these issues seem to come together to understandably stop him being able to empathise.

The above examples of altered emotional reactions are emblematic of a shift in some participants' emotions that had a drastic impact on their lives, an impact that they recognised. Some participants, however, had a shift in their emotional reactions that was better characterised as a disconnect between their lifeworld and their emotionality. While it may still have had a large effect on their lives, these participants did not discuss or recognise this impact.

Daniel perhaps shows this emotional disconnect best. Throughout his interview, he is curt in his replies and rarely expands on relatively simple answers. When talking about apathy, Daniel stated that he used to be apathetic, but does not any more feel that way:

"D: Yeh, used to [feel apathetic]. My first diagnosis. [.]

K: If you remember back to that time, could you describe how it felt?

D: Well, I don't know. Difficult to handle diagnosis. There's no cure. [.] But I'm alright now…

K: So being diagnosed, that was what made you feel apathy?

D: Yeh.

K: What changed to stop you feeling like that?

D: You've just got to get on with life. [.]

K: How long did you experience apathy?

D: About a year maybe. [.]

K: Did it go away slowly, or just wake up one day-?

D: Probably, yeh. Just felt different. [.]"

Daniel does not go into detail about what apathy is or what his symptoms of it were but is sure that he got over it and is no longer apathetic after suffering with it for approximately a year. Daniel seems to follow this pattern, of stating that he used to feel something but no longer does, across other areas of his mental state. He also states that he used to worry about the progression of HD, but he no longer worries:

"Worried? I used to, with the job. With the job and my illness… Used to, don't really worry now. Got diagnosed, used to worry then, yeh."

Daniel seems to be saying that he used to worry about HD until he was diagnosed and started to experience symptoms, then stopped working and stopped worrying. While this makes a certain amount of sense in that the onset of symptoms is likely to have been a constant worry when one is working, it also seems unlikely that that worry would immediately stop. Almost immediately after stating that he does not worry, Daniel states that recently he has been worrying about his weight loss that may be due to his medication or worsening symptoms:

"Yeh, I worry about now my weight loss, cos I've lost weight now, […] part of the illness, I think. I think so. Supposed to see a dietitian but then they put me in touch with a doctor, nurse, a nurse, not a dietitian. All she did was weigh me, lost one stone in three months."

Interestingly, this exchange is one of the parts of the interview where Daniel talks the most. He continues, saying that he hopes a dietitian will put him on a high calorie diet in the future. In addition to talking about his weight and diet, Daniel also talked at reasonable length about his family history of HD, and about his involvement with a local support group. He was able and willing to talk about both topics in reasonable detail and length. When discussing his feelings to do with HD or apathy, however, Daniel talked very briefly.

Perhaps Daniel's brevity in answering any questions about his feelings may simply be a part of his personality. He may have never particularly enjoyed talking about his feelings and is more comfortable talking about external matters such as his family history and involvement in a support group or simple things like his potential weight loss. The problem with this interpretation, however, is that it implies that Daniel's brevity masks deeper feelings that he was actively withholding during the interview. Contradicting this, when Daniel talks about apathy, his answers are given extremely fast, with little pause for thought, and are all surface-level answers that, even when leading questions are used, Daniel does not elaborate. There does not appear to be any depth to his feelings about apathy or his recollection of the experience of apathy. Further on, apathy is revisited (including using a leading question about feeling "low" – although Daniel agrees with the statement, it is not appropriate to comment on this as he was led to agree):

"Researcher: You got diagnosed and you felt different for about a year. Would you say you felt low?

Daniel: Yeh, yeh. Then I thought, at the end of the day, obviously no cure, but at the end of the day you just have to get on with life.

R: But it took you a while to get there?

D: Yeh.

R: So what changes have there been since then?

D: I don't really know.

R: But better now?

D: Better now, yeh."

Daniel did not seem agitated at this point or aggravated with the questions, as one might expect if he did not want to talk about his experience of apathy and was trying to push the conversation in a different direction. Instead, Daniel simply did not seem to think about the questions and did not engage with contemplating his emotional state. Thinking back to Heidegger's ideas of the authentic and inauthentic selves, it is difficult to get a sense of either from Daniel. With Jane, Sophia, and Philip, there was a reasonably clear distinction between who they wanted to be, their authentic selves, and who they found themselves to be, their inauthentic or apathy selves. This was in part because of their struggle to engage with their emotions in the same way that they used to, and because this struggle was an active element of their lifeworld that they acknowledged caused them anguish. Daniel does not present in this way; he does not seem to have a relationship with either his authentic or inauthentic self, but instead seems to concentrate on relatively simple elements of his life such as his weight loss.

6.1.3 Theme three– Behavioural difficulty

The first theme concentrated on participants' identity struggle between an apathy-self and an authentic-self, while the second theme explored participants' difficulty to connect with their emotions. This last theme looks at participants' struggle with their behaviour. It is important to note that this is not looking at behavioural difficulty in the sense of people's behaviour presenting a problem to others (e.g. aggressive behaviour towards others), but rather looking at behaviours that participants talked about or presented during the interview that was striking. Some of these have been touched on in the two themes above but here are presented independently to unpack them.

Philip's confusion during the interview is striking, not because of how pronounced it is, but because of how specific it seems. He does not become overtly confused by many questions, and generally follows the conversation, although is constantly distracted by his (admittedly noisy) dog. Throughout the interview, however, he seemed on edge. He was nervously playing with the dog or clasping his hands together. Further on in the interview, Philip talked about being confused and anxious throughout most interactions and mentions that the interview itself was daunting for him (before the recording started). He states:

"I feel very confused all the time; you know. I have to concentrate on, you know, this conversation. I have to sort of physically, well- mentally concentrate on what- what we're saying."

This struggle to keep up with any conversation causes Philip anxiety and puts him off social engagements of any kind, be it going to a pub with his brother or buying something in a shop. This anxiety could seem like a form of apathy; Philip disengages from social occasions, has difficulty leaving the house, struggles to empathise with others, and his difficulty following conversations could be easily interpreted as a lack of desire to have a conversation. Later, Philip describes another problem he has while watching television:

"…I've got a very low level of concentration [.]. When I'm watching TV, I can erm often erm have missed a whole programme [laughs], and er I can't remember what it's been about…It's as if I've been watching it but not recording it, not taking it in, you know, it's annoying. It reminds me of this, of the problems and that I'm not all that well."

Some of Philip's language suggests he may be upset and angry with his situation; even when trying to merely watch television, he cannot sit there and enjoy the show, but ends up being reminded of his illness. This, combined with his inability to empathise with other people or himself, to a degree, suggest that as well as being anxious, Philip does also get annoyed and angry at the situation he finds himself in. Apathy, in terms of it being a state of being without emotion or passion, seems relatively far from Philip's experience. He may come across as apathetic in a brief interaction as he is unable to concentrate on a conversation and may seem detached, but the emotion bubbling underneath, the anxiety, confusion, anger, and annoyance, reinforce the idea that Philip's apathy is at least in-part an avoidant coping strategy. The perhaps overwhelming nature of Philip's emotional confusion lends itself to avoiding situations that could cause an emotional flare-up (such as struggling to follow conversations in a crowded environment).

For Jean, a lot of her thoughts about apathy revolve around her mother who had HD and became very apathetic. While researching HD, Jean and her son found out that people can follow a similar pattern to their relatives who also had HD; this led Jean to try and find out as much as possible about her relatives, her mother in particular. This seemed to be a preoccupation for Jean, who explained how she watched her mother become increasingly apathetic after she was moved to a care home:

"My mother became apathetic later on when she was in the home. They would sit her in a chair in front of the television and regardless what was on, and er they would come and feed her when she needed feeding and leave her the rest of the time, so it wasn't very satisfactory… It was- it was all that was available when she became incapable. She did become very apathetic. Just shutdown. So er – I'm not like that [laughs]."

Jean is quick to point out that she is not like her mother. Previously she mentioned that she was "roughly following the same pattern" as her mother but is keen to point out where she is not. She went on to explain her mother's behaviour in more detail:

"…she was quite a nosey person, she liked to know what was happening, what was going on, but when she got the Huntington's, she just withdrew, and she would just sit for hours. She wouldn't look at a book, she wouldn't look at a magazine and she had no interest in TV although it was on. She just withdrew completely."

At the start of the interview, Jean mentioned that she reads books, mostly fiction, frequently. Perhaps, again, Jean is offering points of difference between herself and her mother. Jean seems to have spent significant time researching her mother's medical history to try and deduce how her own symptoms will progress. Although she does not report being overly affected by HD (at the time of interview), she seems very persistent in deducing her mother's symptom progression and thereby possibly learning about her own future. Around her house, there were pictures of her mother from various family occasions or portraits. Jean mentions these in passing later on when talking about following the same pattern as her mother. This paints these pictures in a slightly different light; comforting in having familial pictures around the house but also a portent of what may be her future. Like Jane, Jean started talking as soon as the Dictaphone was switched on:

"Well my mother had Huntington's; she was far worse than me. My tremors are only in my jaw and her arms and legs were flailing and she was incapacitated with it and er it hasn't happened to me and we want to know why [laughs]."

Jean immediately compares herself to her mother's symptom progression, and notes that she is not experiencing the same severity of symptoms as her mother. Jean's use of words here is intriguing. There are very few emotive words and the point of the anecdote is to highlight Jean's curiosity. She is aware that the most likely disease progression will follow a similar pattern to her relatives' and knows that she is not following that pattern. Her mother seems to have suffered severely with chorea and apathy, whereas Jean states that she does not. Throughout the interview, however, Jean remains relatively impassive. She does not express relief for her relatively minor symptoms, when compared to her mother's, but is instead constantly intrigued by the possibility that she will begin to follow the same pattern at some point. Even in that intrigue, she does not appear concerned, but more perplexed as to why she is not following that pattern. Jean's perplexed preoccupation with her mother's apathy and HD, while understandable to a degree, could suggest that Jean does feel some inexorable, self-fulfilling prophecy-type pull towards apathy that she has not revealed to her family and certainly not during the interview.

Sylvia's impassivity during some potentially upsetting moments in the interview perhaps show an emotional and behavioural difficulty. When describing her diagnosis with HD, Sylvia and William seem to come at the issue from very opposed emotional standpoints that hints at Sylvia being apathetic pre-diagnosis. William describes his wishful thinking, hoping that Sylvia has restless leg syndrome rather than HD, while Sylvia describes a sort of resignation, of being certain that she has HD from the outset, passed down from her mother:

"Sylvia: Mm. My mum died of Huntington's, and my uncle as well. […] [Turns to Will] You were convinced I didn't have it, you thought I had rest- rest syndrome-

William: Restless leg syndrome.

S: Yeh, wasn't you?

W: I was hoping you did.

S: Yeh, but I knew, I knew that I had it before I had the test.

Researcher: So did you have the test to confirm after you'd started to show symptoms?

S: Yeh.

W: She was absolutely 100%, no, I've got it, I've got it. And when- it was [your Doctor] who told you, wasn't it?

S: Yeh.

W: And [Sylvia] said, yeh, OK, and that was it.

S: She burst into tears, bless her [the Doctor].

W: Yeh.

S: I was fine, I was. I knew I had it so- I had a day off and went back to work the day afterwards.

W: Yeh, one day.

R: So is that because you were already prepared?

S: Yeh, I just got my head around it for one day, and then I was alright [laughs]. […]"

Sylvia remained impassive throughout this exchange, imparting this story in a matter-of-fact manner, while William was visibly shaken by the recollection. Sylvia seems to have come to terms with the idea of having HD before the diagnosis and does not seem to react when the confirmatory diagnosis comes through. Although she states that she took a day to come to terms with the diagnosis, this seems like a monumental thing to achieve. Sylvia was already exhibiting some symptoms of HD and describing her reaction here as apathetic seems appropriate. Perhaps, in these early stages of the disease, Sylvia was already apathetic, and this emotionless reaction has now become relatively normal for Sylvia.

For Sylvia and William, apathy is characterised by tiredness, and Sylvia's tiredness was apparently cured by the B12 injections, therefore Sylvia is no longer apathetic. Given these data from the interview, however, it seems more likely that Sylvia was at least partially apathetic when she was diagnosed with HD and remains so now. Her emotional reactions throughout the interview were at least blunted, and in places absent when it would have been appropriate and reasonable to become upset by some of the stories she was recounting. In the diagnosis story, Sylvia could have been adopting a stoical coping strategy, remaining strong (perhaps for William) in the face of great adversity. It is a well-known early coping strategy in life-changing diagnoses. If this were the case, one would expect further stages of coping, similar to how people process grief. For Sylvia, however, she only seems to have this one form of stoic reaction that presents as apathy. Another possibility is that perhaps this initial apathy at the time of diagnosis was a partially conscious protective mechanism to cope with the horrible, terrifying prospect of HD. Again, however, Sylvia's emotional disconnect throughout the interview suggests that it is not just a reaction to the diagnosis, but a part of who Sylvia is now. Sylvia's authentic self is now apathetic.

Emma presented another interesting behavioural difficulty, and perhaps one that could be called more 'classically' or stereotypically apathetic. Emma frequently responded to questions using as few words as possible and would quite often choose one-word, affirmative answers or look to her mum to answer. On a few occasions, Emma did visibly brighten up and combined her monosyllabic responses with animated facial movements. Here, Liz (Emma's mum) showed the interviewer a piece of pottery that Emma had painted butterflies and coloured patterns on:

"Researcher: So with that did you choose all the different colours and stuff and then do it all yourself?

Emma: Yeh

R: Oh wow

Liz: Pretty

R: Yeh. How long did it take?

E: Ages [laughs]"

Although it was difficult to engage Emma in an extended conversation, this was perhaps the most animated she became during the interview, her facial expression livening up while exclaiming 'ages'. Unprompted, Emma did not say anything and seemed content not saying or doing anything. When prompted, Emma did seem keen to try new things wherever possible, such as riding a bicycle in the park or watching any football match involving Manchester United. These activities, however, seem to be suggested by others rather than by Emma herself. There seems to be very little that Emma does when she is left on her own. Liz explains this:

"Just about the fact that she's- just needs really to sort of do anything cos otherwise if you don't, she'll come down and just sit and watch TV all day. Unless you told her- I mean sometimes as much as saying, you know, get up, don't you need the toilet, you know, something like that cos she just sometimes focusses on- focusses on the TV and that seems to be a lot of what it's about. You need some other motivation cos otherwise it's just a case of just sitting and watching TV."

Emma seems to lack motivation and finds it difficult to think of things to do unless prompted by Liz. More than this, it does not seem to be the case that Emma struggles to think of anything to do, as this implies that she initiates that thought process in the first place. It seems to be the case, from what Liz says, that there is no initial initiation of thought processes to begin to find motivation. This is perhaps similar to Sophia who struggles to find the motivation to get out of bed but a step worsened. Where Sophia struggles to initiate the will to get out of bed and recognises this lacking, Emma does not seem to recognise this lacking. During the interview, Liz bought up the issue of insight into apathy:

"I think it's difficult- I don't want to answer for you, but I think it's probably difficult for you because- she doesn't realise she's like that. Do you s- until somebody points it out, you don't realise, because that's just as normal as you going to work and me doing what I do during a day, because it's just what's normal for Emma, because that is- it's only other people that look in and see that really so I think that's hard for her to describe because she doesn't realise that's what's happening."

Emma agreed with Liz about this (although Emma agreed with Liz about almost everything and this was her default response). Emma did seem to lack insight into her apathy and did not seem to see a problem in sitting on the sofa watching television all day or not motivating herself to do something. Towards the end of the interview, the issue of whether apathy was a problem was discussed between all three:

"L: And is that what you've found yourself, that everybody is different?

K: Yes, everyone is a case unto themselves. Originally the plan was to have a great unified theory of apathy, but I don't think that's going to happen because everyone is so-

E: Different.

K: Yeh, yeh, everyone's experience is different. You know, you seem to be great at coping, Emma.

E: Yeh.

L: That's it, isn't it? I suppose it's whether or not [.] that word [apathy] makes a difference to you whether or not it affects the way you are, and yes it does affect you because obviously you may be different to the way you were before, but if it's not bothering you-. I suppose that's a silly way of putting it, but do you know what I mean?

K: As long as you're coping.

L: Yeh, if you can cope with that and carry on and still be OK and be fine about it then that's OK. If you want to do things and you're not able to do things then that's different, isn't it? So erm [.]"

This is perhaps one of the dilemmas of both identifying and treating apathy. Perhaps, for Emma and others, this form of apathy is a defensive coping strategy to avoid the struggles of coping with the HD symptom burden. Philip seemed to have developed an avoidant coping strategy where he actively avoided situations that may reveal his physical or mental struggles and this presented as a form of apathy. For Emma, perhaps this lack of thought generation or process is an extreme coping strategy. If one does not think of things to do, there is unlikely to be a problem. When others suggest things to do, Emma enjoys them, but the responsibility and mental load of thinking about those things is not required. This perhaps makes sense when again thinking about Heidegger's authentic and inauthentic selves. Everything that Emma does is down to others, down to an inauthentic self, but this does not seem to be a problem for Emma. It has, perhaps, become her authentic self who acquiesces to others, to the inauthentic self, as a means of subconsciously coping with the general burden of HD.

This theme highlights different behavioural problems that are either impacted by apathy or are a fundamental part of people's experience of apathy and emphasises the variability in how people experience apathy. Apathy, for these participants, is not a uniform experience but some of the impact of the experience of apathy is the same; all participants' lives have altered, including how they think of themselves and how they feel able to behave.

6.2 Empty and bewildered apathy – combining the interview and measures' data:

Analysing these data from the semi-structured interviews and measures of apathy, participants' experience of apathy seemed to fall into two distinct types. Firstly, Jane, Philip, and Sophia seemed to consciously struggle with apathy, experiencing anxiety, confused emotions, and had difficulty doing things they used to be able to do easily. They were able to discuss their problems, their feelings about apathy, and how apathy seemed to be affecting their lives. These three participants will be referred to as experiencing 'bewildered apathy'.

Secondly, Daniel, Sylvia, and Emma also experienced apathy and seemed to have impaired emotional responses, had difficulty maintaining thought processes, and seemed less aware of their relative lack of activity. These three participants will be referred to as experiencing 'empty apathy', as they could not explain their experience of apathy in the same detail as the first group.

The data from the three measures of apathy were particularly useful in exploring discrepancies and potential misunderstandings in the interview data that helped to differentiate these two types of apathy. Perhaps most emblematic of the utility of the measures' data was the comparison between Philip and Sylvia. Philip seemed to experience bewildered apathy, while Sylvia experienced empty apathy, yet they scored relatively similarly on both the AES-C and DAS. Their scores varied more on the LARS-SF but, when looking at their raw data from the LARS-SF, this can be explained by their different responses to only two parts of the measure; emotional responses and taking the initiative. Their responses on these two parts accounts for their scores differing by four points.

As for differentiation, their responses to the AES-C show this nicely. Taking their scores on the AES-C, Sylvia scored 52, while Philip scored 50, but the way they amassed points was subtly different, despite them presenting as completely different across the interviews. They both presented as apathetic, but their emotional capacity and cognitive ability levels appeared to be very different. The measure itself failed to differentiate between them and did so generally by level of intensity. On item 18, related to motivation for example, Sylvia scored a three, while Philip scored a two. On item six, related to how much effort they put into things, Sylvia scored a three, while Philip scored a one. Based on the interviews, Philip seemed to put more effort into things than Sylvia did, but his self-belief had been eroded by his anxiety, as demonstrated in the interview. Philip scored low on multiple items that, relative to Sylvia, he demonstrated more of during the interviews. Yet, because his subjective interpretation was that his ability had significantly dropped, the AES-C forced a low score. Philip over-thought his answer to each item on the AES-C, which contrived to give him a high score, while Sylvia's lack of ability to consider the items contrived for her to score highly. One interpretation here is that both Philip and Sylvia were apathetic, the AES-C recognised their apathy, and both were therefore accurately described as apathetic according to the measure. This ignores the complexity of each participants' individual experience, however. It remains that Philip and Sylvia experienced apathy in very different ways. It was only when considering their presentation at interview and then delving into each individual item of the measures that it was possible to begin to analyse this difference and look beyond the global sum-score that the AES-C generates.

The different way in which Sylvia and Philip were diagnosable with apathy given the measures of apathy demonstrates the difficulty in both constructing a clinically useful measure of apathy and of diagnosing a single type of apathy. From the interview data, Sylvia and Philip's presentation was entirely different, yet from the AES-C in particular, their score was relatively similar. The different dimensions in which they accumulated that score, however, show that apathy is more likely to be multi-dimensional than uni-dimensional. It is true that the AES-C loads the questions on to three dimensions of apathy, but then still sums these scores into a global apathy score, thus eroding the sensitivity of the scale. The DAS does do better by maintaining separation of the three dimensions of apathy that it purports to measure, but Sylvia and Philip still scored relatively similarly on the DAS dimensions despite presenting entirely differently and being split across the two types of apathy described here.

Mary and Jean do not fall into either of these types. A case can be made for Mary to fall into the second, empty apathy category (she had difficulty thinking, could not explain apathy, and made some choices about her behaviour that seemed demonstrably apathetic), but further interviews with Mary would have been necessary to draw this conclusion. The unfortunate timing of her bereavement prior to the interview badly impacted her emotional state and made it difficult to judge which behaviours were related to what area of her life. Jean could be placed into the bewildered apathy type, but throughout her interview, she did not seem particularly apathetic. She demonstrated some behaviours that could have been due to a level of apathy, such as her preoccupation with her mother's apathetic behaviour, but not consistently. Jean's emotional responses were slightly blunted, but this seemed more like a general personality trait over a symptom of apathy. On balance, she has not been included in the discussion of the bewildered apathy type.

These two types of apathy, bewildered and empty apathy, will be discussed further in chapter seven, the discussion. This brief description introduces the types and shows how the interview and measure data contributed to differentiate the two.

6.3 Summary of analysis:

Apathy is a complex phenomenon, with participants experiencing different impacts on their lifeworld. Theme one highlights the disconnect some participants experienced between who they thought of as their authentic self and the self they became when experiencing apathy. Participants who experienced this disconnect were often confused by their inability to overcome these feelings and wished to return to their previous self. Theme two is necessarily linked to the first theme but emphasises that participants' emotional responses have changed and that some participants felt they could not predict or alter their emotions, while others struggled to engage in any form of emotionality at all. The third theme then emphasises how participants' behaviour altered when faced with apathy in HD. Running through all themes was a sense that participants' internal identity and understanding of their self was fundamentally changed by both apathy and HD.

Contrary to the majority of the literature, including Simpson et al.'s (2015) qualitative inquiry of apathy in Parkinson's disease, the majority of participants did not directly refer to a lack of motivation as a component of apathy. Only Liz, Emma's mother, talked directly about a lack of motivation and equated it to apathy. Some of the participants found it hard to talk about apathy due to a combination of not being sure what apathy was, not being able to define it, or not having the vocabulary to describe their feelings and experience. For some participants, their symptoms of HD, predominantly those that affected their cognitive abilities, seemed to increase the difficulty they had in talking about apathy and reduced their ability to think about their answers to the measures of apathy. Talking at any length or in detail was difficult for those with cognitive deficits, while clarity of thought was often a problem for participants whose cognitive processes were better. The measures of apathy were useful when analysed at the level of individual items, but struggled to offer enough nuance to be able to rely on the sum-scores.

7. Chapter Seven – Discussion

This thesis has explored the concept of apathy, including an exploration of the concept's etymology, a systematic review of relevant literature, and two phases of empirical data collection with people with apathy in Huntington's disease (HD). This chapter will bring together these investigations of apathy in HD and relate it to relevant literature, looking at how they are interrelated and will begin to unravel the concept of apathy. The following and final chapter will then relate these findings to the research question posed at the outset of this work as well as explore important avenues for further research.

This chapter will explore:

- The two types of apathy outlined in chapter six;
- Apathy's status as uni- and multi-dimensional;
- The overlap between depression, apathy, and different types of apathy;
- Apathy as a protective factor in HD;
- The use of IPA to explore apathy; and
- The limitations of this body of work.

7.1 Empty and bewildered apathy:

The interviews with participants show that apathy in HD is a complex phenomenon that altered all participants' identity. All eight participants demonstrated a form of apathy during their interviews but also showed that apathy, similar to depression, can be a problem that changes over time with ebbs and flows related to other elements of a person's life, or *Dasein*, and is experienced in different ways by different people. As mentioned in the analysis, participants seemed to show a different type or stage of apathy across the interviews that will be further explained here and related to relevant literature.

Apathy is bound to encompass a range of experience, akin to other psychiatric conditions such as depression where people often experience a broad spectrum of related symptoms. In depression, however, people are often diagnosed with more specific conditions such as major depression disorder and the type and severity of depression will be specified based on a diagnostic manual. With apathy, the current approach allows for no specificity other than stating that people have apathy. Given these participants' experience of apathy in HD, this is insufficient.

The way Jane has difficulty in precisely defining apathy and the struggles she talks about in differentiating between anxiety, depression, and apathy is very different to Daniel's relatively blasé attitude to apathy where he brushes it off but is perhaps unaware of its impact on his lifeworld. Jean and Sylvia define apathy in a similar way to each other, referring mainly to a lethargy or tiredness that reduces one's ability to do anything, but present this definition in completely different ways. During the interview, Jean evidently spends time thinking about the conundrum of defining apathy and what exactly it is, while Sylvia does not pause to think about her answer, stating instead that apathy is like a form of tiredness and sees no need to elaborate or differentiate further. Philip, meanwhile, states that a component of apathy is not caring about other people and thinks that he does suffer from apathy but is also terribly aware of how slow he is and feels social anxiety whenever he steps outside his front door. Daniel states that it is difficult to cope with the diagnosis of HD and that he probably suffered from apathy for a while but then, according to him, got over this problem as he realised that he needed to get on with his life. According to Daniel's account, this was a relatively simple revelation that required little cognitive effort and seemingly no mulling-over on his part.

The point here is that all the participants' experiences were different. This may seem slightly obvious given the data but is a fundamental point that is not made in the literature, where many papers keenly point out differences between depression and apathy (e.g. Ishii, Weintraub, and Mervis, 2009), but do not discuss differences inherent in people's experience of apathy. For the participants in this study, apathy represented a heterogenous experience with overarching similarities. Similar to Simpson et al.'s (2015) experiential study of apathy in PD, participants' struggle with activities of daily living was often related to symptoms of HD; their ability had changed because of their symptom burden, and this often seemed to reduce their ability to do many things. Participants experienced a disconnect between their authentic self and their apathy self, as presented in the previous chapter, and this led to a state of emotional turmoil in those experiencing bewildered apathy, and a state of emotional disconnect (rather than emotional blunting, an overly crude term, discussed below) in those experiencing empty apathy.

It is important to note, particularly given the heterogeneity described above, that these two types of apathy are not necessarily exhaustive but serve to show that apathy is a complex, diverse phenomenon that warrants the specificity that other psychiatric conditions are given. Bewildered and empty apathy are two possible types of apathy that the participants in this study demonstrate, but further research is needed to confirm that the distinctions identified here exist across different groups of people and in different disorders.

7.1.1 Empty apathy:

Daniel, Sylvia, and Emma seem to display 'empty apathy', characterised by:

- An inability to explain their relative lack of activity;
- Lack of analytical and cognitive processes;
- Lack of generation or initiation of thought;
- Lack of awareness of their emotional state;
- Struggle to engage socially for any extended time, seeming 'empty' in social situations.

These participants' experience of apathy seems to be similar to the most common definitions of apathy in the literature, particularly those that centre around a loss of motivation (Marin, 1991; Starkstein, 2012), and is possibly easier to identify than the second type of apathy that Jane, Philip, and Sophia demonstrate. There are, however, important elements of these participants' experience of apathy that are more subtle than merely a loss of motivation. For Daniel, Sylvia, and Emma, they seem to lack motivation but this is not their main problem. Instead, empty apathy seems to be a state where people struggle to initiate thoughts or experience emotions, which may present as a lack of motivation but is a more fundamental problem that may strip people of their identity.

Sylvia, throughout the interview, was dismayed when William pointed out changes in her behaviour that she herself had not recognised and consistently struggled, without turning to William for help, to identify activities she likes to do. When asked to elaborate on things she enjoys doing, she was often perplexed by the question and seemed to find it difficult to understand why she was being asked to explain further. William would often elaborate for Sylvia. In terms of Sylvia's emotional response, she seemed to find it hard to fully engage with her feelings. On one occasion, she became angry when discussing losing contact with ex-colleagues, but again found it impossible to explain why, without William bearing the brunt of this explanation. Sylvia found it difficult to understand or explain her own emotional response, could not explain why she did less now than she used to do, and seemed disinterested in thinking about these issues as problems.

Emma's experience differs to Sylvia's but is perhaps emblematic of similar issues. Liz explains that Emma will sit in front of the television watching whatever happens to be on until an external factor forces her to change her behaviour. In this instance, Emma seems to have lost all motivation; it is not that she wishes to do something and cannot find the motivation but that there is no thought process instigating a potential new behaviour unless Liz jolts Emma into doing something. It is perhaps more accurate to describe Emma as being unaware of her own interests unless prompted. Emma has very little ability to determine what it is she likes doing unless she is reminded of those things and this makes her seem unaware of her own interests rather than merely suffering from a lack of motivation to do those things. This interpretation of Emma's lack of behaviour and lack of cognition does perhaps support Levy and Dubois' (2006) criticism of describing apathy as a loss of motivation as being overly reliant on a psychological state. Robert et al. (2018) discuss referring to apathy as a lack of goal-directed activity, which is perhaps more inclusive and closer to describing Emma's state. Emma's apathy begins before motivation has become an issue; there seems to be no psychological 'problem' with Emma's behaviour, but she seems unable to initiate thoughts or feelings without external aid

Daniel's experience of apathy is different to Emma and Sylvia's but shares similarities. Daniel does identify things he likes to do without prompt, unlike Emma, but struggles to appreciate that this is evidently fewer activities than he used to do, even when this is explicitly questioned. This lack of insight and understanding that Daniel, Sylvia, and Emma demonstrate is partially explained by the cognitive decline inherent in HD, but the loss of motivation is evident in all three participants, despite their range of different cognitive levels. It therefore seems to be correlated with the loss of cognitive ability but not exclusively caused by it. It is not possible to rule out that cognitive impairment is the cause of Daniel, Sylvia, and Emma's apathy, but it is also improbable that cognitive decline is the sole cause of their apathy. Additionally, even if the cognitive impact of HD was the cause of their empty apathy, this would be an important finding as their apathy presents so differently to the apathy demonstrated by Jane, Philip, and Sophia.

In Simpson et al.'s (2015) study of apathy in Parkinson's disease, their first theme in the analysis of their interview data shows that their participants experienced a reduction in motivation due to the symptom burden of PD. This implies, as they state, that apathy was directly related to their reduced mobility and ability, and reinforces some previous studies of the impact of PD on quality of life (Starkstein et al., 2009; Leroi, Ahearn, Andrews, McDonald, Byrne, and Burns, 2011). This also implies a heavy cognitive load when considering apathy. The participants in Simpson et al.'s study recognised the change in their lifeworld, evaluated their options, and ended up in a state of reduced motivation. This is a complex process of cognitive evaluation that, in Daniel's, Sylvia's, and Emma's description of their experience, they were incapable of conducting. Empty apathy seems to be a state where those complex cognitive and evaluative tasks are no longer possible and is, therefore, related to cognitive decline. It is an 'empty' state because the participants in this study demonstrated an emptiness when it comes to creation of thought and recognition of their relative lack of activity or cognition.

7.1.2 Bewildered apathy:

This leads to, secondly, Jane, Philip, and Sophia, who seem to experience 'bewildered apathy', characterised by:

- An awareness of their relative reduction in activity;
- Ability to hypothesise or judge why they may have reduced their activity;
- A struggle to identify their emotions, but retained emotionality;
- Experience of distress in response to their problems;
- Bewildered or confused by their emotional state;
- Struggle between their authentic-self and inauthentic/apathy-self.

Jane, Philip, and Sophia explained that they thought they did suffer from apathy but, unlike Daniel Sylvia, and Emma, could explain or at least discuss their symptoms of apathy, could rationalise some of their reduced level of activity, and were preoccupied by their symptoms related to both apathy and HD. Similar to Simpson et al.'s (2015) participants, they struggled with the impact of HD and apathy seemed to be, at least in part, a response to the symptom burden of HD, contrary to Pluck and Brown's (2002) suggestion that apathy in PD is a consequence of physiological rather than psychological reaction. They retained a good degree of insight into their apathy, demonstrated by their ability to talk about their experience and feelings about apathy and seemed to retain an ability to recognise at least a desire to do things, but often struggled with executing those things.

Jane wanted to donate to a charity because she knew that when she was younger, she would have wanted to donate; she retained the goal of donating but felt disconnected from either the positive emotion of donating or the negative emotion (predominantly guilt or a sense of responsibility) that may have pushed her to donate. Philip explained that he rarely went shopping because he was worried about counting money and would frequently cancel social engagements if he perceived them to be potentially difficult. In one instance, he described cancelling going to see a local band in a pub with his brother. He was concerned that he would not be able to find a seat, that if he had to stand, he would not be able to for very long, and that he would not be able to hear anything his brother said. He seemed to spend all day worrying about the potential pitfalls of leaving the house, many of the problems related to his symptom burden, and so decided to stay at home for fear of disappointing his brother whilst at the pub. Prior to his increased symptom burden, it seems likely that Philip would have gone to the gig. It must be noted here that most models of apathy stipulate that apathy must not be predominantly attributable to things associated with disease burden (e.g. Marin, 1991). Here, Philip's decision to stay at home is not directly related to his symptoms of HD. It is related to his social anxiety and complex emotional state; his interpretation of what may happen partially because of his symptom burden, exacerbated by his apathy, is what means he stayed at home that day. He is able to fully explain this decision but felt that he could not do anything else. It was not the decision he wanted to make, it was the decision he felt forced into because of his symptom burden and apathy.

It is the nuance of bewildered apathy that is novel here. Those who displayed empty apathy follow the definition of apathy as identified by Marin and others in terms of a lack of motivation, emotional blunting, and relative indifference (e.g. Marin, 1991). The participants with bewildered apathy do not fit with what may be termed this classical understanding of apathy; they retain insight, are not emotionally blunted but experience difficulty interpreting and analysing their emotions, and their activity level is impaired rather than entirely lost. Yet, they identify as suffering from apathy and certainly did not do as much as they used to do. Their experience of a struggle with apathy is seemingly more overt than the participants with empty apathy. They are aware of their struggle and struggle against apathy. Jane, in particular, is keen for her young daughter not to recognise that Jane is apathetic, but still struggles not to be apathetic. This emotionally charged dichotomy is unrecognised in the literature where a more classical, extreme type of apathy is recognised that prioritises a lack of motivation and emotional blunting and aligns closer with the empty apathy identified here.

The notion of the apathy-self, introduced in chapter six, the analysis, was particularly important for those participants experiencing bewildered apathy. In some illnesses, particularly neurodegenerative disorders, it is common for people to experience physical inability even though they think they still can perform those physical actions. In Parkinson's disease (PD), for instance, a person may think they can drink water steadily from a glass but motor symptoms such as tremors (Jin, Wang, Liu, Zhu, Loprinzi, and Fan, 2019) means they cannot drink from that glass without spilling. This lack of control disrupts people's independence, autonomy, and identity, with some describing their body as alien to their mind, a form of mind-body split (Bramley and Eatough, 2005). For the participants experiencing bewildered apathy, there seems to be a struggle with a mind-emotion split, as well as a mind-body split. Instead of purely a rift between a person's physical capability and their mind, as described by Bramley and Eatough (2005) when looking at symptoms of PD, apathy could mean that they experience a rift or disconnect between their emotional reaction and their identity. People's emotional reactions are often directly tied to their own identity; they see themselves as someone who cries at certain films or gets angry at a particular behaviour or scenario. Here, the apathy-self seems to take away that part of their identity, severing a crucial link in their narrative of who they are and reducing their experience of Heidegger's authentic-self. Participants described fighting against apathy, of trying to regain their old emotional reactions but struggled to do this. The apathy-self seemed to be becoming their new norm, their new authentic-self, despite their best efforts to stop this process, and this caused a large part of their bewilderment. This finding is again similar to Simpson et al.'s (2015) finding that people with apathy in PD often experienced or chose disengagement as a way to manage the emotional impact of PD. There are also similarities with Bramley and Eatough's (2005) analysis of symptoms of PD, where people described a pre-diagnosis and post-diagnosis self, the pre-diagnosis self being people's desired but impossible state. The apathy-self took away people's ability to express the emotions they used to feel, replacing them with a form of emotional turmoil and bewilderment.

7.1.3 Two types of apathy or apathy as a continuum:

The way patients' apathy presented was split into two types. Rather than apathy being a continuum that gets better or worse depending on either disease progression, management, or other factors, the semi-structured interview data suggests that apathy consists of different types. The data presented suggest empty and bewildered apathy are two distinct types of apathy that may be unrelated. People with apathy in HD could experience either empty or bewildered apathy, could experience both types at different times, and could experience an entirely different form of apathy at different stages of their life. People's experience of apathy is also likely to decrease or increase at different times. It is also possible that empty and bewildered apathy are interrelated and exist on a continuum. People may move from one type to another, possibly related to disease progression or their current level of insight into their condition. As the data were not longitudinal, no conclusion can be drawn about these possibilities until further research is carried out.

At this early stage of investigation, it is unclear if this division of bewildered and empty apathy represent two types of apathy that are unrelated or if they are part of a progression of apathy within HD. Without any longitudinal data, again, it is important to stress that no clear answer can yet be given to this. Given the progressive nature of HD, however, it seems plausible that patients could progress from bewildered to empty apathy via a loss of insight that is common in HD (Sitek, Thompson, Craufurd, and Snowden, 2014). Perhaps, if these two types of apathy are a continuum, patients lose much of their insight into their condition and behaviour due to increased cognitive impairment related to HD and this causes a shift from bewildered to empty apathy. Krishnamoorthy and Craufurd (2011) suggest that apathy is an inevitable part of HD that worsens the longer a person is symptomatic, potentially supporting the idea that people move from bewildered to empty apathy with disease progression. While this is possible, further research is needed in this area.

7.2 Emotional blunting and the passions in apathy:

It is important to note a key feature of both empty and bewildered apathy related to the somewhat generic term emotional blunting. Overall, participants did not seem to experience typical emotional blunting, but rather experienced difficulty in narrowing down and comprehending their own emotional response. The participants with empty apathy perhaps experienced an emotional response most similar to emotional blunting; Emma and Daniel displayed very few emotive responses, while Sylvia seemed mostly confused by her feelings. The participants with bewildered apathy experienced a more complex relationship with their emotions than the term emotional blunting allows for. Some of Sophia's responses to things that would otherwise likely make someone sad did not have that effect on her. She no longer did anything associated with her career as a seamstress but did not seem to react emotionally to this. At a cursory glance, this could have been interpreted as emotional blunting. Yet, when digging deeper, this did impact her life and she did have an emotional reaction to her lack of ability; it was only when recounting this story that she seemed emotionally blunted. Perhaps this was because she had already experienced the emotional response and it was the retelling of the story that did not induce as much of an emotional response, not the event itself.

This is a subtle but important difference. Emotional blunting implies that someone does not have appropriate emotional reactions to events. In Sophia's case here, we would therefore describe her as having emotional blunting if she experienced no emotion related to losing her ability as a seamstress. As Sophia clearly states, however, at the time she was distressed by losing this skill. When retelling the story, perhaps an impact of the apathy that Sophia was experiencing led to her losing the connection to the emotions she experienced. This loss of connection to her emotional reaction in turn led to her being unable to feel the same emotion that she once felt towards her lack of ability, and instead led to her presenting as emotionally blunted. Underneath the surface of this apparent emotional blunting, Sophia's relationship to those emotions was much more complex. She had experienced loss and, when recounting the story, seemed to be rationalising this loss. She discussed the problem she had with her hands due to some symptoms related to HD and arthritis and noted that this increased the difficulty in doing anything related to dressmaking. Her apathy was worsened by her perception of physical difficulties and this produced a very complex emotional reaction. Describing this as emotional blunting detracts from the power and complexity of Sophia's relationship with her hands, her mind, and her disease burden.

Jane and Philip present similarly, seeming to be emotionally blunted, but instead seemed to be experiencing a form of emotional disconnection. Jane ruminates about all of her feelings a great deal. Of particular note, she worried about not feeling emotionally connected to emotive television shows. While Jane described laughing at her sister who cried at an advert, Jane recognised that she would have cried before. Philip described struggling to concentrate and feeling a lack of empathy, but also reported being socially anxious and often unable to leave his house due to his anxiety. This seems to be representative of emotional confusion, both feeling that they lack empathy but also very worried about other people's responses to their lack of empathy. For both Jane and Philip, this appeared to be an almost endless cycle of co-occurring non-existent and existent empathy. Describing this confusing experience as emotional blunting seems to minimise their suffering and, further, given how distressed they both seemed at times during the interview, inappropriate. Rather, Jane and Philip seemed to experience emotional turmoil where they had a problem connecting with and understanding their emotions.

Returning to chapter one's exploration of the simplistic translation of the etymology of apathy may help to explain this disconnect. Apathy is broadly translated as a-pathos, or 'without passion'. This is again interpreted as 'without emotion' as the term passions was replaced by the all-encompassing emotions. In that translation and interpretation, much linguistic nuance was lost. When we now talk about emotion, it can mean anything from love and hate to sadness, depression, or jubilation. Emotion refers to everything that one may experience as a reaction to an event that is not (necessarily) behavioural, but in doing so, is reduced to meaning nothing. Therefore, when referring to someone as displaying emotional blunting, it is assumed that this means all (or at least a large proportion) of a person's emotional reactions (love, hate, etc.) are now less than whatever they used to be for that person. The implication in this is that the emotion itself has remained the same but the amount of that emotion has reduced. The complaint lodged against the term emotion is generally that it has lost all sensitivity and is not as specific or meaningful as passions, sentiments, or affections of the soul. Here we can see the impact of this lack of specificity. The idea that an emotion can stay the same but merely reduce is an extremely rigid, insensitive interpretation of a complex psychological process. If one loved someone but then found out that they disagreed with that person about a fundamental belief, it would be inaccurate to say that this merely reduced one's love for that person. The relationship would necessarily change, and the emotions and feelings attached to that relationship would change. Similarly, participants experience of apathy in HD is not a simple experience. Describing Sophia, Jane, and Philip as emotionally blunted does not accurately describe their feelings. These participants were not experiencing merely a reduction or blunting of their emotional response, but rather they experienced a change in how they reacted to things. The change in emotion they experienced was potentially different to how they would have experienced it prior to being apathetic and this distinction, between the overly simplistic emotional blunting and recognition of the complexity of emotional change in participants' lives, is key in understanding apathy in HD.

Much of the research into measures of apathy focuses on emotional blunting as a key driver of difference when looking at apathy and other conditions such as depression or anhedonia (the inability to feel pleasure). Ang, Lockwood, Apps, et al. (2017) in their development of the Apathy Motivation Index, point out that apathy is considered to be characterised by emotional blunting while depression is viewed as a state of extreme emotional fluctuation. We would therefore look for emotional blunting when considering apathy in a person and emotional fluctuation would preclude apathy. Some of the participants in this study, however, demonstrated that emotional blunting is an overly simplistic description of a person's emotional state, and that a state of emotional change or emotional disruption (for these participants) is a better description of their state. They still felt strong emotional reactions but could not always convey or demonstrate these responses clearly. While blunting implies a reduction or complete loss of response, disruption implies a state where emotions can become confused and potentially overwhelm a person's normal response to the extent that they may present as emotionally blunted at a cursory glance. Deeper exploration should reveal the disruption of a person's emotional reaction certainly in those experiencing bewildered apathy. In empty apathy, people's experience may be closer to a form of emotional blunting, but it is important to note that experience of complex emotional change in apathy seems more likely than emotional blunting.

7.3 Is apathy uni-dimensional or multi-dimensional?

Recent literature that conceptually explores apathy seems to consider it a multi-dimensional construct generally triadic in structure (e.g. Radakovic and Abrahams, 2018; Caravaggio et al., 2018), other studies that explore the prevalence of apathy or other aspects tend to refer to apathy as a loss of motivation. As such, there is still confusion about the number of dimensions that apathy may consist. The problem does not seem to be that authors disagree about the multi-dimensional status of apathy, but rather that it is easier to define apathy as a loss of motivation and continue reporting the more interesting elements of a study's exploration of prevalence or some other aspect. Bogart (2011) was perhaps the last author to publicly doubt that apathy was anything more than a symptom, and much research since then has demonstrated the multi-dimensional nature of apathy (e.g. Arnould et al., 2013). The debate about apathy being uni- or multi-dimensional is essentially closed but defining apathy as a loss of motivation continues. This is a problem as it oversimplifies what is an evidently complex phenomenon into an inappropriately simple statement. Much as referring to 'pathe' as 'emotions' was shown to be overly simplistic in chapter one, referring to apathy as a loss of motivation is only describing a small element of the impact that the participants' in this study described and experienced. As the triadic structure of apathy is developed further and literature recognises the difficulties people endure while living with apathy, this should become less of a problem.

This is not to say that a loss of motivation is not rightly part of a description of apathy or a large part of a dimension of apathy, but to point out that this it is most likely to be one of many symptoms of an apathy syndrome. It must also be pointed out that recent definitions and diagnostic criteria have moved away from talking about apathy as a loss or lack of motivation, instead preferring to talk about apathy as a loss of goal-directed activity (Robert et al., 2018). This difference is potentially important in diagnostic criteria to avoid subjective interpretations of how to recognise a lack of motivation (as goal-directed activity is better defined) but is less important when discussing people's experience of apathy. In the semi-structured interviews reported in this thesis, it was occasionally necessary (as in the interview with Mary) to briefly define apathy to help a participant. Defining apathy as a loss of motivation was preferable to describing a quantitative reduction in goal-directed activity as it was simpler and easier to understand. Describing apathy as a loss of motivation and therefore potentially uni-dimensional is perhaps, therefore, a good lay description of apathy, but if one is thinking about apathy as a clinical problem, a more appropriate definition would emphasise the triadic structure and complex nature of the phenomenon prior to discussing a loss of motivation or reduction in goal-directed activity.

Perhaps part of the reluctance to acknowledge the complexity and multi-dimensional structure of apathy is related to the problem that Fried (2016) identified with uni- and multi-dimensional depression. With the relative proliferation of measures of apathy, there is a necessary link with apathy being uni-dimensional for the purposes of measure-consistency. To show reliability and to be able to provide a global sum-score of apathy, any measure must show that apathy is uni-dimensional and that the items in the measure represent a proxy for measuring one factor. This forces apathy into a relatively narrow construct, such as defining it as a loss of motivation, for the sake of ensuring the various measures of apathy are consistent and all load primarily onto one single factor (apathy). Radakovic and Abraham's (2014) Dimensional Apathy Scale avoids summing scores to give an overall apathy score, but is in the minority.

7.4 Apathy, depression, and differential diagnosis:

The similarities between apathy and depression present diagnostic and treatment difficulties, particularly when using diagnostic measures, as discussed in the systematic review. Mary and Emma offer the most direct, useful comparison here that perhaps shows the diagnostic challenge in differentiating between apathy and depression and also between different types of apathy. This difficulty is particularly salient as recent models of apathy look to describe subtypes of apathy, a welcome step, although with the caveat that this is generally based on psychometric data alone and does not include experiential data (Ang, Lockwood, Apps, et al., 2017). Relying on psychometric data to describe types or subtypes of apathy potentially repeats the problems identified in the systematic review, namely missing the importance of understanding people's experience rather than merely symptoms.

The misdiagnosis of depression when a type of apathy may be more appropriate is plausible given Mary's presentation, in particular. Mary established in her mind that she had nothing to get up for and that decision made her sad, potentially verging on depressed, when combined with everything else that was happening to her (her separation from her husband and close friend recently dying). Although Mary did suffer from many physical limitations due to her illness, there were activities that Mary could get involved in with minimal help. She decided that she did not want to do these activities and therefore stayed in bed, partly to avoid these activities (e.g. going into the common room in the care home to watch television with the other residents). From this perspective, Mary made an active decision to isolate herself by staying in bed, which could be reasonably said to be a symptom of a depressive disorder. When one considers, however, that one of the aspects of the operationalisation of apathy is a lack of motivation, perhaps choosing to not get out of bed could be interpreted as a symptom of apathy instead. Mary is apathetic because she is upset by her situation, which has caused her to make the active decision not to be involved in various activities. It is perhaps the active element of this decision, however, that could make a difference. If Mary could not explain why she stayed in bed, if she stayed in bed because no-one suggested she should do anything else, then apathy may be a better descriptor for her state. Mary stated, however, that she stayed in bed because she had no reason to get out of bed. Perhaps, then, Mary is suffering from co-occurring bewildered apathy and depression. Simpson et al.'s (2015) experiential study of apathy seemed to demonstrate people in similar confusing states. In that study, participants described making an active choice to not wash their car (for example) partly due to their worsening symptoms related to PD. This was interpreted as a form of apathy, but one that was heavily influenced by disease progression. This leads on to the section below, which discusses the possibility that apathy is a reasonable response to neurodegenerative disorders.

Returning to the diagnostic difficulty between depression, apathy, and types of apathy, Emma presents perhaps the antonym to Mary's confusing diagnostic state. When asked what she does, Liz, Emma's mother, says that Emma tends to watch television when at home. The implication in what Liz says is that Emma only watches television because the television is there; it is not an active choice that Emma makes, but a passive one. If the television was not there, Emma may merely sit there and do nothing. Whereas Mary seems to make the active choice to stay in bed, Emma passively watches television. This difference, between an active and a passive decision, is perhaps a key difference between a symptom of depression and a symptom of an apathy type. If a person tends more towards actively making a choice, even if that choice is to not do something, they may be doing so because they know that activity will make them upset and may therefore be suffering from depression or bewildered apathy (or potentially both). In this case, Mary chose not to get up because the options she had available to her were no better than staying in bed. She could explain her reason behind staying in bed but was also upset at being put in the position to make that choice, perhaps demonstrating the co-occurrence of depression and bewildered apathy. Sophia, meanwhile, when she reported not being able to do any housework, is not upset at the prospect but instead struggles to connect emotionally with that decision. In this way, perhaps Sophia is therefore suffering from bewildered apathy without the presence of depression (which would be likely to induce sadness in that decision). If, as in Emma's case, a person tends more towards passively doing something and makes no effort to do something better (in terms of what they personally prefer doing), it is more likely that they are suffering from empty apathy, and further, may have already progressed through bewildered apathy, lost further insight, and now are incapable of describing their apathy.

7.5 Is apathy a protective factor in HD?

Depression has many causes, some of which can be resolved by pharmacological intervention. Alongside this, however, some suggest that a state of depression is an appropriate and understandable reaction to the events occurring in one's life. The ICD-10 (WHO, 1992; before being replaced by the ICD-11) differentiated between episodes of depression that meet the criteria for severity but not for duration, allowing for a depressive episode (F32, ICD-10) to first be diagnosed before a recurrent depressive episode (F33 in ICD-10). The ICD does not mention mitigating circumstances in its stipulations for diagnosing either a single depressive episode or recurrent depressive episode. It is plausible, for instance, for a person to experience a depressive episode that is instigated by the death of a close family member or other significant event in their life. Gross, Smith, and Stern (2007) discussed the potential for depression to be an appropriate response to a diagnosis of cancer. In a similar way, it seems plausible for apathy to be an appropriate response to the increasing symptom burden of HD.

As well as considering the possibility that apathy is a biological or neurological symptom of HD, it is also possible that apathy is a partially unconscious psychological reaction to HD that provides a level of protection against the reduction in quality of life that disease burden induces. Given the impact of the physical, neurological, and psychiatric impact of HD, struggling to adjust to this new life and struggling to complete tasks seem like reasonable psychological and behavioural responses. If things are becoming more difficult to do and trying to do them constantly reminds someone of their infirmity, stopping doing them seems reasonable. Simpson et al. (2015) point out that some of their participants' apathetic behaviour was likely to have a secondary effect of protecting them against the recognition of increasing physical infirmity. By not performing activities they used to do, they did not have the opportunity to 'test' themselves or recognise that they could no longer perform a specific task. In this way, their apathy was protecting them against potential emotional distress. No participants discussed this element specifically in either Simpson et al.'s (2015) study (it was an interpretation proffered by the researchers, not the participants) or this study, but this potential protective element of apathy could go some way to explaining some of the literature's lack of acknowledgement of the severity of apathy as a clinical problem. Although all participants' activity level had dropped, partially due to being apathetic, this could have had the impact of not reducing their quality of life as much as if they tried to maintain the same level of activity but consistently failed because of their disease burden. It also, however, had the potential impact of restricting their quality of life as they were not doing as much as they used to do. Future research is needed to further explore this hypothesis and determine if this secondary impact of apathy as a protective factor is artificially increasing (or at least not decreasing as much) people's quality of life.

7.6 Using IPA to study apathy:

The use of interpretative phenomenological analysis could be viewed as unorthodox or inappropriate given the relative difficulty participants had in talking about apathy. Typically, IPA is used in populations where participants will have a high degree of understanding about the phenomenon under investigation and are able to cogently and articulately discuss their experience of the phenomenon. This was a known risk in the thesis and is discussed further in chapter three, the methodology. As expected, participants struggled to define apathy; Mary in-particular was unsure about apathy and could not provide a definition without multiple prompts. As has been pointed out in chapter two, however, the definition and conceptualisation of apathy is in its conceptual infancy. A different qualitative method (e.g. discourse or thematic analysis) would have struggled in a similar way to IPA but would not have allowed the flexibility of interpretative analysis to discuss the detail and nuance that chapter six, the analysis of the semi-structured interviews, goes into.

Alternatively, further quantitative elements could have been incorporated into the study (and these were considered in the design of the empirical work), perhaps asking participants to respond to a series of novel questionnaires to elicit the words they associate with apathy. A technique along these lines could have gathered data from more participants but would not have allowed the analysis to be as detailed or as idiographic as is possible here with the semi-structured interview data using IPA. Given there is conflict among a number of academic and medical professionals about the conceptualisation of apathy (as shown in the systematic review), this study set out to take a novel approach to the study of apathy and focus on individual's experience of apathy rather than concentrate on identifying neurological correlates or models of apathy that did not contain experiential data. A quantitative methodology would not have allowed the conceptual space to explore people's experience and use this knowledge to try and understand how apathy has impacted their lives. This is not to say that these potential methods are inappropriate, but that they would have drawn the focus of this thesis in a different direction.

The interviews and subsequent analysis concentrate on identifying the individual (employing an idiographic style) and is entirely in keeping with phenomenology. This was a deliberate choice; the definition of apathy and understanding of the concept should include individual's experience of apathy and this thesis prioritises individual experience in defining and understanding apathy.

7.6.1 Reflexive process:

As part of the process of using IPA to study apathy in HD, a short biography of the researcher's position is reported in chapter four (section 4.5) and a reflexive diary was kept throughout the research process. In particular, notes were taken before and after each interview to note the researcher's anticipation and reaction to the interview.

This work has tried to "reflexively uncover" (Finlay, 2003) people's experience of apathy and part of that process is recognising one's own emotional reactions, state, and how they have affected the research. Most salient to note here is that the emotional impact of every interview was different, but also that the researcher's emotional reaction was strong following each interview. These emotional reactions are bound to have shaped the subsequent research direction. The strength of emotional reaction, in particular, led the analysis to look in-depth at participants' emotional states throughout the interviews and, in places, considering the appropriateness of this emotion (in the researcher's opinion). The identification of a struggle with identity, a key theme in the analysis, in part emerged from the researcher's emotional reaction to some of the difficulties the participants described.

Acknowledging and recognising the impact of emotion and of human interaction is an integral part of conducting any qualitative work, particularly that which relies on phenomenological roots, as this work does. Following these strong emotional reactions and after considering the direction that each interview took, further reading around bracketing (Tufford and Newman, 2010), reflexive processes and emotional intelligence in research (Collins and Cooper, 2014), and the importance of debriefing in qualitative work (Rager, 2005) was undertaken.

7.7 Limitations:

Most of the limitations of this thesis are related to the difficulty of working with an ill-defined concept. Apathy, as the systematic review in chapter two showed, is a broad concept that most authors define differently. On an individual level, it is also a symptom or syndrome that people experience differently and therefore talk about very differently, as the results of the semi-structured interviews demonstrate. It is likely that most people, in both a medical and social context, are talking about a slightly different construct of apathy in any discussion.

This makes studying apathy particularly difficult. This also demonstrates the importance of carrying out this research in order to move towards a better understanding of what it is to experience apathy and how an improved understanding would aid recognition, identification, and management of apathy. It is unlikely that meaningful treatment or management techniques will be effective without this understanding.

7.7.1 The complexity of apathy:

Chapter one explored the concept of apathy, delving into the etymology of apatheia and looking at how apathy is defined in medical literature. This has been a useful investigation that has informed the entire thesis and the exploration of the passions and emotions, in particular, was useful when thinking about the complexity of people's emotional capacity in apathy.

The problem with this approach, however, is that there remain several interpretations or definitions of apathy that have not been explored. The predominant ones have been presented across this thesis, but there is a large swathe of types of apathy missing, particularly those that track the journey between the Stoic philosopher's ideal of apatheia to the modern derivative apathy. Some elements have been traced to Christian writings that referred to apathy as a form of disbelief in a higher being, but this shift from apatheia to apathy is more complex. Dixon's (2003) work, in particular, points out further distinctions between the passions and emotions that there was not space to discuss here, but which may help to illuminate future research into the construct of apathy.

7.7.2 Defining apathy is problematic:

Participants' struggle to define apathy (shown in table 6 in chapter six) is emblematic of the struggle the participants had in not only defining apathy but in expressing the impact apathy had on their lives. The Greek prefix 'a' denotes negation (as chapter one describes in more detail), meaning that apathy is a word that describes lacking something. Further, defining what is lacking is difficult, something that is reflected in participants' definitions of apathy and descriptions of their experience. Often, their definitions of apathy did not necessarily describe their experience of apathy. People's experience of apathy was never a standalone experience, as their definitions were, but an experience caught up in the totality of their *Dasein* and therefore one that did not always tally with their static definition of apathy. This is particularly important as it suggests that static definitions of apathy are not necessarily representative of people's experience. The lack of alignment between participants' definitions of apathy and their experience of apathy is important as an experiential lesson when considering clinical matters related to treatment and management.

It must also be pointed out that asking someone to define a complex phenomenon that they are potentially experiencing is problematic. This is certainly a possibility that must be taken into account. In this case, however, the problem is the static nature of defining apathy in the first place and not a problem created by the variable nature of people's experience. Take Sophia's definition of apathy, for example. She describes apathy as being a disinterest in things that includes being disinterested in doing anything. This is a reasonable, understandable definition of apathy that much of Sophia's behaviour reinforces, but is an extreme form of the behaviour and experience that Sophia describes.

The point here is that in trying to define apathy in a succinct way (at the behest of the interviewer, it must be noted), participants often end up contradicting themselves in what they attribute as apathetic behaviour. Defining apathy statically, as the literature also attempts to do, predominantly in order to allow it to be quantifiable, ignores the totality of participants' own *Dasein*. Jane's experience of apathy naturally bleeds into her experience of frustration and of fear. Philip's experience of apathy blends into his experience of social anxiety and worsens his stammer. Sophia's experience of apathy compounds her disinterest in things which then heightens her guilt at her husband being forced to do more household chores. Here, apathy cannot be disentangled from each participant's experience of *Dasein*. All of their definitions of apathy are, of course, genuine and accurate, but because they were asked to provide a static definition, as the literature tends to do, this definition does not represent an accurate picture of the totality of what apathy constitutes.

It is important to note that this is not criticism of the participants' definitions of apathy. The participants defined apathy in their own words, informed by their experience, and this is indisputably useful. What it shows, when examined closely here with the other experiences of apathy that participants describe, is that these standalone definitions do not survive *Dasein*. Sylvia defines apathy as "when you're tired all the time" but the experiences she and William describes, those of struggling to cross the street or having difficulty following conversations are not necessarily representative of merely being tired. Tiredness may be a part of it, but further on in the interview, Sylvia says that since her regular B12 injections, she does not experience tiredness anymore, but William reports that she still seems to daydream when walking down the street and he has to be careful that she does not wander into the road. She still has difficulty following and maintaining conversations and particularly struggles sustaining or elaborating on any of her answers.

7.7.3 Recruiting participants with apathy:

As chapter two, the systematic review, points out, there are no accepted diagnostic criteria for apathy and there are multiple measures of apathy, none of which have been adopted as a diagnostic tool. This meant that when recruiting participants to the study, the research relied on clinical experience and judgement to assess whether a participant had experienced or was experiencing apathy prior to interview. Participants were only recruited when consensus was reached within a specialist, multi-disciplinary team (including several senior clinicians) that those recruited had experience of apathy. The lack of accepted diagnostic criteria that could be appropriately used to recruit participants to the study raises the possibility that participants were experiencing different phenomena that, perhaps, clinical staff interpreted as apathy.

7.7.4 Defining apathy in interviews:

Another limitation, related to the above methodological problems, is that most participants struggled to define apathy. This was a known problem with using a qualitative form of data collection to explore apathy within a population likely to have cognitive capacity issues. Interviewing people with apathy and some of their caregivers certainly helped to gain multiple perspectives, and the identification of trends and similarities in participants' experience was reassuring in ensuring that the data were of as high a quality as possible. There were only two interviews in which it was necessary to explicitly define apathy for participants before they had offered a definition, and on both of these occasions, a script laid out in the semi-structured interview schedule was followed.

7.7.5 Clinical view of apathy:

Healthcare professionals' (HCPs) views of the concept of apathy are undoubtedly important and are currently unrepresented in the literature. This thesis has focussed on identifying the experience of people who are living with HD and apathy, and as such, did not investigate how HCPs such as specialist nurses, consultants, or junior doctors identified or understood apathy. As so little is currently understood of people's experience of apathy in HD, interviewing HCPs was decided to be overreaching this thesis but is certainly an area of important growth. Now that more is understood about people's experience of apathy in HD, future work can focus on understanding HCPs' views and looking for alignment and divergence on the path to managing and treating apathy in neurodegenerative conditions.

7.7.6 Neural correlates of apathy:

Much of the literature, particularly in the field of neuropsychology, focusses on identifying neural correlates and underlying neurological impairment as a means to identifying a certain form of apathy. There are some authors, particularly Radakovic and Abrahams (2018), who are starting to combine the exploration of behavioural correlates of apathy with these neurological models. This thesis, however, is focussed on exploring the construct of apathy from an experiential perspective. Future work should indeed endeavour to combine these investigations to form a fuller exploration of apathy, but this is an incremental process. This exploratory work into the experiential understanding of apathy is a relatively new field with few studies and justifies more investigation before combining with neural models of apathy.

An area of potential overlap between the neuropsychological and experiential fields in this respect is the work of Leander, Shah, and Sanders' (2014), looking at 'goal-directed behaviour' (GDB). They suggest that any observed lack of GDB is not a novel preference but an exploitation (by an external factor) of pre-existing (potentially latent) preference. There were times, Leander, Shah, and Sanders suggest, that people's apathetic behaviour was influenced by a preference that they would not normally yield to, but that they did because of their apathy. This pattern of behaviour is potentially recognisable in some of the participants in this work, while others explicitly contradict this narrative. These similarities and differences may yield useful areas to begin to unravel different types of apathy and different neural mechanisms underlying apathy.

7.7.7 Interviewing participants as a dyad:

This issue was addressed in chapter four but is important to emphasise here. The original design of the study allowed for interviews with people with apathy and HD and their carers, either together, as a dyad, or individually. All interviews where a carer was involved were conducted as a dyad, following the participants' wishes. It would have been methodologically useful to have a mix of people with apathy and HD on their own, with their carers, and carers on their own. There may have been differences in how each participant responded based on the type of interview. This was not possible in this study so should be integrated as part of a larger study into apathy in HD. Although not possible here, participants were frank and open in interviews, with some polite disagreements occurring, meaning it was likely that little data were lost. Future work is needed to verify this, however.

7.7.8 Cognitive impairment in people with HD and apathy:

Considering the participants' potential cognitive impairment when thinking about their experience of apathy is one of the challenges of this thesis; the interviews demonstrate the difficulty of asking for experiential data from people who are potentially struggling to recall their experience. It is, however, imperative that participants' voices are heard and factored into the discussion of what apathy is and how it should be defined. There are no previous studies that report people's experience of apathy in HD; most of the existing literature focusses on other elements of the discussion of apathy, with the exception of Simpson et al.'s (2014) experientially-driven study. Mele et al. (2019) explore apathy using a qualitative method but do not focus on people's experience of apathy but rather their understanding of the barriers and facilitators to managing apathy. Despite some of the problems with cognitive impairment that were evident in the interviews, this work has demonstrated that complexity exists and that hearing from people who are experiencing this complexity is vital.

When recruiting for the interviews, a relatively broad set of inclusion criteria were used when assessing potential participants' apathy status. This could be seen as insufficiently rigorous. Instead of using a measure of apathy as inclusion criteria, this study used the clinical team's judgement, bolstered by the opinion of the participant's family, where possible. Participants were only interviewed if they were believed to be apathetic by all of their clinical team, thereby reducing any one individual clinician's bias towards a certain definition of apathy. Mele et al. (2019) used the Starkstein Apathy Scale (Starkstein et al., 1992) to screen participants but subsequently noted, however, that healthcare professionals recognised the lack of standardisation in assessment of apathy as a barrier to discussion of apathy. Given this lack of standardisation and, as chapter two's systematic review identifies in this thesis, that different measures of apathy focus on slightly different conceptualisations of apathy, using a measure of apathy to screen participants may have introduced a bias towards a certain type of apathy. Using a measure of apathy as a screening tool may reduce bias introduced by the study team but introduces bias from the creator of the measure, while using the clinical team's judgement reduces bias from outside the research team but introduces potential internal bias. Given the conceptually unstandardised position of apathy, however, and given that this thesis is exploring the concept of apathy, not using a measure and relying on clinical judgement was the best fit. Additionally, the use of measures of apathy alongside the semi-structured interviews showed that most participants were apathetic according to multiple measures.

8. Chapter Eight – Conclusion

This research sought to explore the concept of apathy, looking at a number of aspects including an etymological exploration, a systematic review of the concept, and experiential data. This body of work shows that the etymology of apathy is complex and its meaning highly influenced by where, how, and by whom it is used. This work has also shown that apathy is ill-defined across different fields of study and that experiential data, in particular is lacking. Novel to this work, alongside the exploration of people's experience of apathy, is the development of distinct types of apathy that influence people's experience and presentation. These distinct types of apathy provide avenues for future research to look at tailored treatment based on apathy type. Further etymological work into the journey from apatheia to apathy may also bolster treatment methods, utilising the positive aspects of Stoic apatheia.

As societies change, so do meanings of words and concepts. The switch in meaning that chapter one's etymological work uncovered, from apatheia being a positive state to apathy being an undesirable state, demonstrates the complexity of language and of meaning-making in a changing world. Some of this change was charted in chapter one, but further exploration of this shift is certainly warranted. We can see that the positive ideal of apatheia was part of a society that valued philosophical oration from the 'Stoa', and the ideal itself was seen as a way to further knowledge through controlling one's state of being. Conversely, apathy as an undesirable state now exists in a society that prioritises different aspects of living, often concentrated on individuals being engaged in meaningful activity. In some ways, this is similar to the ideal of the 'Stoa', but with a different focus. Apathy is therefore seen as undesirable and potentially dangerous to both the individual and to society if people disengage from meaningful activity. It is the negative meanings that seem to have spilled over into the medicalisation of apathy. The reliance on describing apathy as a lack of motivation, as much literature continues to do, is perhaps symbolic of how our society treats disengagement as a personal problem that must be solved. Future research would therefore do well to remember the origins of apathy and explore why some people experience apathy, or a disengagement with the world. Perhaps embracing the positive aspects of apatheia may help people to reunify their identity in the face of apathy and be able to choose how they wish to live their lives.

8.1 Clinical apathy and societal apathy:

This research explicitly contradicts the narrative, present in much of the literature, that apathy is a problem primarily of motivation or disinterest and suggests ways in which the clinical problem of apathy is distinct from social ideas about the meaning and experience of apathy.

Apathy is a distressing clinical problem for people with Huntington's disease (and other neurodegenerative conditions) that adversely affects their quality of life and that of those around them. Although most existing literature reviewed in chapters one and two identifies that apathy is a clinical problem in need of further study (see e.g. Levy et al., 1998), the role of motivational impairment or disinterest is generally emphasised over people's experience of distress. Apathy is seen as a problem of motivation or a generalised lack of caring that causes emotional blunting, and therefore is not necessarily distressing to the individual experiencing apathy. Many of the papers look at impairment in quality of life as a structural problem that suggests neurological impairment, potentially ignoring the personal, social, and experiential aspects of apathy. All participants interviewed in this thesis were adversely impacted by their experience of apathy and experienced varying levels of social disengagement as they struggled with a shift in their identity. There were some aspects of apathy that may have served as a protective factor to shield them partially from the impact of their HD symptom burden, but the predominant impact of apathy was characterised by emotional turmoil.

It is perhaps our reliance on lay or historical understandings of the term that has led to research concentrating on apathy following the narrative of a lack of motivation. Apathy, when used generally in a social setting, is often used as a synonym for a lack of caring and this has continued relatively unchallenged in the literature. This narrative has perhaps even been reinforced in recent years as the societal idea of apathy in younger people is held up as a danger to society because of a supposed lack of caring, and apathy, in this context, can be understood as a synonym for boredom, resignation, lethargy, or indifference. When thinking about apathy in general terms, it is perhaps best at this point to accept that these potential synonyms carry societal similarities in terms of their definition, but when thinking about apathy as a distressing clinical problem, as this thesis does, there are fundamental differences between apathy and its societal synonyms.

It is also important to note here that considering the parallels between clinical apathy and societal apathy moves the focus away from the experience of apathy as a clinical problem, as this thesis has focussed on, and moves towards general concerns about apathy in society. General concerns about apathy are, of course, valid, but are a different problem to the problem of clinical apathy in the context of neurodegenerative conditions that warrant their own investigation. The previously discussed parallels between apathy and depression, a topic the concept of apathy is never far from in the literature, further delineate this. Depression exists as both a clinical problem and a way of describing one's mood or general feeling. As a clinical problem, it is a different experience from being sad, but as a mood, it would not be unreasonable to describe someone as sad if they stated that they are feeling depressed. In a similar way, the clinical problem of apathy denotes, in the case of the participants interviewed in this study, an experience of either bewildered or empty apathy that involves emotional turmoil, identity confusion, and behavioural difficulty. A general feeling or mood of apathy could include some of these problems but is more likely to be a description of boredom or indifference. Without the clinical aspect or a significant reduction in quality of life, it is a different problem with different assumptions and goals behind its identification. Apathy, as a clinical problem, is subjectively and structurally different from the feeling of apathy as evoked by the societal usage of the term.

The data in this thesis serve to reinforce this point. The clinical problem of apathy has a drastic, severe impact on people's quality of life. While some of the literature notes this impact on quality of life, it usually does so from a detached perspective. The adverse impact of apathy, particularly in terms of people's struggle to identify their authentic-self and therefore to choose how to live their life, is an important finding of this work. In addition, this work was able to highlight that participants' experience of distress was greatly impacted by the type of apathy they experienced. This carries implications for the development of future management plans (see below).

8.2 Types of apathy:

This thesis challenges the generally held conception of apathy as a singular symptom, syndrome or experience. Instead, the work undertaken here shows that different types of apathy can be identified in a clinical population. In this case, participants with HD were grouped into two types of apathy, termed here as 'bewildered' and 'empty'. This is an important finding as it emphasises the variability of people's experience of apathy, and also serves to point out that apathy is a more complex structure than previous research suggested. Whereas earlier research tended to focus on a lack of motivation or lack of caring as being central to the identification of apathy, the research presented here, by exploring in depth the experiential aspects of apathy, was able to reveal more about the phenomenon of apathy. In doing so, it becomes perhaps more accurate to talk about the multiple phenomena of apathy.

It is also important to note that while two types of apathy have been identified here, it seems both plausible and likely that more types of apathy exist and that these types may vary depending on underlying condition, socio-cultural factors, personality, and symptom burden. Apathy is naturally impacted by the totality of a person's *Dasein*, and this should be an important element of future research into different types of apathy.

In terms of the experience of these types of apathy, all participants tended to experience distress, demonstrated by emotional turmoil, problems, or generalised confusion. Those with bewildered apathy demonstrated worse, or at the very least more evident and generalised distress, while those with empty apathy tended to experience emotional confusion. This emotional confusion could present as emotional blunting, an aspect of apathy that the existing literature tends to focus on. This work shows, however, that the term emotional blunting, similar to motivation, is an overly simplistic and reductive explanation of apathy's impact on people's emotional state.

The complexity of apathy and the potential for different types of apathy is well represented by participants' experience of identity crisis. People with bewildered apathy tended to experience a deep identity crisis that led to them questioning much of their everyday behaviour. The misalignment between participants' authentic-self and their inauthentic- or apathy-self was of paramount importance to their experience of distress. Apathy seemed to make participants doubt who they were, in turn causing them to question their every decision. The impact of this was, often, to paralyse their ability to choose. It is tempting here to talk about this state of being as representing a lack of motivation, but to do so is to miss the point about the complexity of this experience. It is not motivation that these participants are missing, but identity, a much more fundamental problem than simply a lack of motivation. To a lesser or greater extent, these participants lost their place in society because of the identity rift caused by apathy. Further, they actively experienced this loss of place in society as they recognised their loss of identity. This is a complex emotional state that perhaps shares more with a lack of confidence than a lack of motivation.

Participants with empty apathy also experienced a form of identity crisis, but often in a passive manner. Where participants with bewildered apathy recognised the conflict between their authentic- and apathy-self and actively railed against it, participants with empty apathy demonstrated a changed identity, but either did not see this as a problem or did not recognise it. This lack of recognition of their changed identity had important ramifications for the experience of distress. Those with bewildered apathy were extremely distressed by their changed identity, while people with empty apathy were more likely to be distressed by the social impact of their changed identity, such as a changed relationship with family and friends.

The complexity and variability of participants' experience of apathy reinforces the identification of different types of apathy in order to better understand the clinical problem of apathy. If this work can serve as a way to begin to differentiate between different types of apathy, future work can build on this and begin to tailor management and treatment to the different types of apathy.

8.3 Future research directions:

This thesis has raised some important directions for future research, with the ultimate aim of better understanding apathy and developing effective management and treatment for the different types of apathy.

The etymological work carried out in this thesis was particularly useful in tracing the history of apathy and giving context to the concept. Further work in this area could continue this investigation, in particular tracing the switch in meaning from apatheia's positive state of being to apathy's undesirable trait. The societal ramifications of this switch seem particularly salient; what changes in society across time made this switch more likely or hastened the process? Is it emblematic of a more general societal change? This early work suggests that changes in philosophical thinking played an important role in this switch, but further work is needed. This exploration would also help to further describe the positive elements of apatheia that have been lost over time and begin to use these as potential avenues for developing treatment or coping strategies, along with the other research suggested here.

The identification of the two types of apathy as described here must be seen as preliminary. Further research is needed to explore the different types of apathy and determine whether other apathy phenomena may be identified. Different apathy phenomena would in turn help us to understand that there may be different underlying mechanisms and research could then be directed at understanding them. One potential area of study could look at the possibility that type of apathy is related to disease progression in HD. Given Simpson et al. (2015) found similar experiences in people with apathy in Parkinson's disease (PD), the relationship between disease progression and apathy type should also be extended to include other neurodegenerative conditions including HD, PD, and different forms of dementia. Alongside this future work, research should include the views and experience of healthcare professionals, a key (but necessary) omission in this thesis.

As well as different types of apathy, identifying some of the underlying reasons behind why people may be experiencing apathy is particularly important. Some research, including the work conducted in this thesis, has identified that apathy may be a protective factor in shielding people from the impact of neurodegeneration (e.g. Simpson et al., 2015). People may also experience apathy in neurodegenerative conditions as a result of external pressure, similar to Swaffer's (2015) description of prescribed disengagement in dementia. There may also be different underlying neural mechanisms at play, something that the field of neuropsychology continues to explore (see Le Heron, Holroyd, Salamone, and Husain, 2019) and which should be combined with experiential work, such as that conducted in this thesis.

Importantly, the identification of different types of apathy raises research questions around their clinical treatment and management. Current research that has looked at treating apathy has tended to focus on identifying pharmacological treatment (Berman, Brodaty, Withall, and Seeher, 2012) or recommended a wide range of treatment types, including pharmacological and psychosocial interventions (Krishnamoorthy and Craufurd, 2011). Little evidence exists for the effectiveness of these treatments, and given the lack of understanding around people's experience of apathy, it seems plausible that treatment outcomes will be difficult to determine. By understanding different types of apathy, any possible treatment can be better tailored to the individual and has the much-needed potential to improve quality of life.

It is also important to note that given the experience profiles of bewildered and empty apathy, both suggest different types of treatment may be more appropriate. In bewildered apathy, people tended to suffer from an identity crisis brought about by their struggle to match their authentic-self and apathy-self. This could be the main concentration of any psychosocial or psychological intervention. Treatments aimed at helping to resolve or come to terms with such conflicts may be important to develop and test in future work, perhaps using some techniques from acceptance and commitment therapy (Hayes, Strosahl, and Wilson, 2012). It is important to note here, however, that accepting this new self may come dangerously close to a form of prescribed disengagement, a phenomenon identified in dementia research (Swaffer, 2015), where people may be inadvertently pushed towards a path that rejects their pre-apathy life. Drawing the line between accepting one's new behaviours and pushing back against them should be the focus of research in managing people's bewildered apathy. Empty apathy, conversely, is likely to demand a different approach to management. Patients with this type of apathy may respond more to behavioural type treatments with active prompting and encouraging and establishing routines, such as the video reminders trialled by O'Neill et al. (2011).

By understanding that apathy phenomena are of different types and underpinned by different mechanisms, research can thus be directed at determining the most appropriate form of management in each case, whether this be psychological, psychosocial, behavioural, or even involve different types of pharmacological treatment (Berman et al., 2012; Theleritis, Siarkos, Katirtzoglou, and Politis, 2017). Developing effective methods to improve people's quality of life with apathy in HD and other neurodegenerative conditions must be at the forefront of all future research.

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10. Appendices:

10.1 Semi-structured interview schedule

Introduction:

- Go through PIS
- Go through consent
- Any questions? Then begin. *[NOTE: Not all questions to be used - some may be missed out if interviewee answers without prompt. Also, follow-up Qs not noted here may be used if interviewee is unable to answer a question due to misunderstanding/ needs more elaboration]*

Schedule:

First of all, I just want to remind you that if I ask anything you don't want to answer, then do tell me and we'll move on. If you need a break at any time for any reason, just tell me as well. Lastly, if you've just had enough and want to stop, tell me and we can either arrange another time to finish or we'll just finish.

Start Dictaphone.

1. [Personality/shift] So this research is looking at psychological symptoms in Huntington's disease (HD), in particular apathy [define as "a lack of motivation, a lack of emotion" only if necessary]. We want to understand a bit more about apathy and how you understand it. First of all, it would be really useful to get to know you a little bit better, what kind of person you are and that kind of thing. So can we start with; what sort of things do you like to do?
    a. How much do you do those things now?
        i. [Did you used to do them more/less?]
    b. Has the way you do those things changed?
    c. And how would you describe yourself more generally?

    d. [If difficult to answer, ask specific] Would you describe yourself as a happy person?
    e. And is that how you feel you are now?
    f. Do you think the HD has changed you at all?
    g. How do you react to stressful things?
        i. [Like if you've just had a bad day]
        ii. [Like hearing some bad news from someone in the family]
    h. Do you think you're a particularly active person?
        i. [If not, did you used to be?]
    i. Do you do things on your own?
        i. [Or wait for other people to start things maybe?]
    j. How would you describe your general mood?
    k. Have you noticed any differences in how you feel or react to things since being diagnosed?
    l. [Continue where/if interviewee continues]

2. [Temporal change/shift, also disease progression/history] As we've mentioned, a lot of people find that some parts of them change after a diagnosis such as HD. Do you think you've changed since being diagnosed?
    a. [If yes] How do you think you've changed?
    b. Has your personality changed?
    c. Do you feel any different now?
    d. Has the way you react to things changed?
    e. Do you get frustrated by things more now?
    f. Do you react to people any differently now?
    g. Have you noticed any other changes in yourself?
        i. [Perhaps it's more difficult to do things?]
        ii. [Have you noticed anything like colours being different?]

        iii. [Have you lost interest in things you used to like?]

    h. [If not] Why do you think that is?
    i. Do you feel any different now?

3. [Apathetic yes/no] OK, next question. A lot of people with neurological conditions, including HD, suffer from apathy. Do you think this applies to you?
    a. [If yes, elaborate] How do you think it applies to you?
    b. How would you describe apathy?
    c. Could you describe how it feels?
    d. Does it always feel the same?
    e. Does it affect your energy levels?
    f. Does it affect how you react to things?
    g. [If not, elaborate] Why do you think this does not apply to you?
4. [Emotional reaction] OK, a bit more general now. Do you get anxious or worried?
    a. [If yes] What do you worry about?
    b. Has that changed, do you think? [What you worry about]
        i. [Do you worry more or less now, do you think?]
    c. [If no] Is that a good thing? [Not worrying]
    d. Do you find that you have nothing to do sometimes?
    e. Does your mind ever just go blank?
        i. How does that make you feel at the time?
        ii. Do you feel any different thinking about it now?
5. [General] OK, I've just got a few slightly more general questions. Could I ask, generally, how much do you understand about HD?
    a. Do you want to know any more?
    b. What about things like household tasks?
        i. How do you find them?
        ii. [Do you find them different now?]

    c. What about self-care?
        i. [How do you find that?]
        ii. [Is it more or less difficult now?]
    d. How often do you see friends or family?
    e. What sort of things do you do with them?
    f. Does it tend to be you or them that organises all that?
    g. Is that more or less than before?

6. We're almost done now then. Thinking about what we've discussed today, is there anything else you think we might not have covered, related to apathy or how you feel?
    a. Is there anything else this has made you think about? [Continue if necessary]
7. OK, thank you very much, that's it for my questions. Thank you for taking part.

    [Debrief participant]

    - Ask participant if they have any questions related to the research (expect questions about when results published etc.).
    - Ask participant how they found the questions and if anything came up they were not expecting.
    - Check participant is OK and not overly distressed by anything that has been discussed. If participant is distressed, inform them that they can contact their clinical team and/or GP. If participant remains distressed, ask them if it's OK for me to contact their clinical team and/or GP.
    - Ensure no problems either related to the research or to their general well-being before finishing.
    - Remind about and schedule a second date for second part of study.

10.2 Table 1 – All papers:

Key: ↓ = lack/loss/decrease/reduction | → = leads to | + = plus | / = or

Description (of paper):

- Measure: paper that develops a novel measure of apathy
- Discussion: paper that discusses apathy conceptually, sometimes referencing a specific illness/injury
- Review: paper that reviews either the concept of apathy or the available measures of apathy (not necessarily a systematic review)

Papers are presented in chronological order. Papers have not been subjected to a quality assessment or ranked by a scoring system. The main aim of the systematic review is to assess the concept of apathy; this aim is not compatible with standard quality assessment tools and developing a bespoke scoring system would not necessarily add to the review. Additionally, one of the problems with assessing the papers, and assessing how well they articulate apathy, is that there is no standardised definition of apathy to compare the authors' definitions against. There is not a gold-standard to apply to the definitions presented and discussed in any of the papers. This means that the judgements of the measures are based on how well the paper explains the authors' concept of apathy and how well it seems to have been thought through and evaluated.

#	Year	Author(s) and title	Description	Field & journal	Definition	Concept	Evidence
1.	1982	Andreasen Negative Symptoms in Schizophrenia	Measure | Development of the SANS measure – includes apathy sub-scale. Developed for use with people with schizophrenia	Clinical General psychiatry	↓ of energy + ↓ of initiation of different tasks + ↓ of saddened or depressed affect + less attention to personal care + difficulty in maintaining employment/study + failure to complete chores + physically inert with ↓ of concentration + severe complaints of apathy [often not expressed, however] = apathy → severe social and economic impairment.	Limited concept – only described briefly Avolition-apathy Views apathy as a negative symptom of schizophrenia Presence of apathy is proxy for assessing negative symptoms – not individual syndrome Patient reporting of apathy contradictory – perhaps author unsure of self-report apathy	No evidence for apathy
2.	1990	Burns, Folstein, Brandt, and Folstein Clinical assessment of irritability, aggression, and apathy in Huntington and Alzheimer disease	Measure | Development of the IAS, includes apathy sub-scale. Developed with AD and HD.	Clinical Psychometric Mental illness	State of disinterest +/ ↓ of action = ↓ of emotion = apathy	Extremely limited conceptualisation. Loss of emotion key Not clear what precisely apathy is or how it differs to other mood disorders in content or identification	No evidence for apathy

3.	1990	Marin Differential diagnosis and classification of apathy	Discussion | Discusses the definition of apathy, suggests symptoms of the apathy syndrome, and explores differential diagnoses	Clinical Psychiatry	↓ of feelings + ↓ of emotions + ↓ of interest/concern + ↓ of motivation w/o ↓ consciousness/intellectual deficit/emotional distress = state of primary motivational impairment = apathy	Apathy conceptually discussed in clinical terms, primarily centring around loss of motivation with some acknowledgement that this is potentially problematic Focusses on clinical use/application of definition	Theoretical, descriptive evidence presented – no data or field evidence
4.	1991	Marin Apathy: A Neuropsychiatric Syndrome	Discussion | Discussion of the definition of apathy, plus differentiation between apathy as a syndrome and symptom	Clinical Neuropsychiatry Clinical neuroscience	Primary ↓ of motivation w/o ↓ consciousness/intellectual deficit/emotional distress = syndromic (primary) apathy ↓ of motivation w/ ↓ consciousness/intellectual deficit/emotional distress = apathy as a symptom (secondary to another syndrome) Deficit in goal-directed behaviour (relative to previous functioning + age/culture) + ↓ productivity + ↓ effort + ↓ initiation + ↓ persistence = apathy	Similar definition and issues to paper #3. Apathy is discussed as a syndrome and as a symptom, although both are referred to as a loss of motivation. Accounts for apathy without a neurological cause, i.e. apathy as primary illness/syndrome, although this is unclear – slightly unclear what is meant by primary syndromic and secondary symptom in relation to apathy.	Similar to #3. Theoretical evidence including specific section looking at definitions of a syndrome in relation to apathy. No data or field evidence.

5.	1991	Marin, Biedrzycki, and Firinciogullari Reliability and validity of the apathy evaluation scale (AES)	Measure | Development of the earliest apathy-specific scale, the AES, for use with all adults	Clinical Psychometric Psychiatry	↓ of motivation w/o ↓ consciousness/cognitive impairment/emotional distress	Apathy discussed, although not conceptually, and relatively briefly, particularly in comparison with other papers by Marin (#3+4). Describes apathy as a psychological response to major life stressors. Confusion between psychological and neurological cause at beginning is conceptually inconsistent.	No evidence presented
6.	1992	Starkstein, Mayberg, Preziosi, et al. Reliability, validity, and clinical correlates of apathy in Parkinson's disease	Measure | Development of AS, Starkstein's revised version of Marin's AES for use with people with PD	Clinical Psychometric Psychiatry	No concept or definition (although takes inspiration from Marin, 1990)	N/A	N/A
7.	1994	Cummings, Mega, Gray, et al. The Neuropsychiatric Inventory: Comprehensive assessment of psychopathology in dementia	Measure | Development of the NPI to assess behaviour in various neurological conditions	Clinical Psychometric Neurology	↓ spontaneity + ↓ activity = apathy	No concept of apathy given and very little description. Definition based on items included in measure rather than conceptual discussion; limited usefulness.	N/A

8.	1998	Levy, Cummings, Fairbanks, et al. Apathy is not depression	Discussion | Exploration of the overlap between apathy and depression in relation to different dementias and neurodegenerative conditions.	Clinical Neuropsychiatry	↓ of motivation w/o ↓ level of consciousness/cognitive impairment/emotional distress = apathy	Limited discussion of apathy and no conceptual discussion.	Useful discussion in terms of differentiating apathy and depression conceptually and symptomatically, but lack of theoretical exploration
9.	2000	Starkstein Apathy and withdrawal	Discussion | Paper explores what apathy is and whether it is a symptom or syndrome	Clinical Unpublished	↓ feeling + ↓ emotion + ↓ interest + ↓ concern + ↓ of motivation = apathy	Apathy definition discussed briefly, but lack of conceptual discussion. It is unclear how the author views apathy – definition presented quotes others without comment	Some evidence from literature, including empirical data. Limited conceptual discussion undermines data
10.	2001	Landes, Sperry, Strauss, and Geldmacher Apathy in Alzheimer's disease	Review | Summary of prevalence, neuropathology, and clinical impact of apathy in AD using a systematic review-like format.	Clinical Geriatrics	↓ of motivation + ↓ of initiation + poor persistence + ↓ of interest + indifference + low social engagement = apathy	Apathy briefly discussed, but no references for definition Conceptual discussion limited.	Empirical evidence presented but little acknowledgement of difference in understanding apathy

11.	2001	Starkstein, Petracca, Chemerinski, and Kremer Syndromic Validity of Apathy in Alzheimer's Disease	Discussion | Examination of proposed diagnostic criteria for apathy in patients with AD, based on Marin's criteria	Clinical Psychiatry	↓ feeling + ↓ emotion + ↓ interest + ↓ concern + ↓ motivation w/o ↓ consciousness/cognitive impairment/emotional distress = apathy	Apathy discussed but not at length and only Marin's definition quoted. Concept of apathy presented is not discussed. Refers to Stoic philosophy derivation of 'apatheia' but does not discuss in relation to modern derivative.	Presents novel empirical data to validate diagnostic criteria. Difficult to assess without further conceptual discussion
12.	2002	Robert, Clairet, Benoit, et al. The Apathy Inventory: assessment of apathy and awareness in Alzheimer's disease, Parkinson's disease and mild cognitive impairment	Measure | Development paper for apathy inventory (IA) for use with any demented and non-demented patients	Clinical Psychometric Geriatrics	↓ of motivation + ↓ of interest + ↓ of emotion = apathy	Some analysis of apathy conceptually but no opinion proffered. Authors do not state how they define apathy (definition here is taken from first lines of paper)	Scale seems well evidenced through statistical analysis but lack of discussion about the concept of apathy and how this measure relates to this.

13.	2002	Strauss and Sperry An informant-based assessment of apathy in Alzheimer disease	Measure | Development of the DAIR for use in people with probable or possible AD	Clinical Psychometrics Neuropsychiatry Neurology	↓ initiation of behaviour + poor persistence + ↓ interest + indifference + low social engagement + blunted emotion → ↓ motivation = apathy	Conceptually unclear. Authors open by describing loss of motivation as a synonym for apathy, in line with Marin's thinking. Later, describes the DAIR, and therefore apathy, as unidimensional, although this is not explained. Apathy discussed but lack of consensus in literature not articulated. Definition also presented as factual rather than on-going discussion.	Statistically sound psychometric evaluation, conceptual discussion lacking
14.	2003	Kolitz, Rodney, Canderploeg, and Curtiss Development of the Key Behaviors Change Inventory: A Traumatic Brain Injury Behavioral Outcome Assessment Instrument	Measure | Development of the KBCI for use with TBI.	Clinical Psychometrics Rehabilitation	N/A	Apathy is not discussed – it is referred to on occasion but not conceptualised	Evidence for measure presented, but no conceptual discussion

15.	2005	Starkstein, Ingram, Garau, and Mizrahi. On the overlap between apathy and depression in dementia	Measure | Mixed paper that develops the SCIA, as well as discusses overlap between apathy and depression	Clinical Psychometrics Neurology Psychiatry	↓ of motivation + ↓ of effort in everyday tasks + dependency on others to structure activity + ↓ of interest in learning new things + ↓ of concern about problems + flat affect + ↓ of emotional response = apathy	Definition here is derived from the discussion of domains in the measure. Clinically focussed, assuming clinician talking to patient. Slightly unclear exactly how the authors conceptualise apathy	Statistically sound, with some good justification of why the data presented is reliable.
16.	2005	Van Reekum, Stuss, and Ostrander Apathy: Why care?	Discussion | General discussion of apathy in various illnesses, with justification of why it is an important research and clinical issue	Clinical Neuropsychiatry	↓ motivation (+ missing 'spark' + missing 'get up and go') = apathy	Apathy is discussed at length in paper and several definitions presented. Lack of clear description of authors' definition of apathy. Questions the use of some scales in presenting clinically useful outcomes	Empirical data from literature presented but not always analysed
17.	2006	Kirsh-Darrow, Fernandez, Marsiske, et al. Dissociating apathy and depression in Parkinson disease	Discussion | Study evaluating the presence of apathy and depression in PD and discussing the dissociation of the two syndromes	Clinical Neurology	Primary ↓ of motivation + ↓ of interest + ↓ of effortful behaviour = apathy	Apathy defined in line with Marin's definition. No discussion of lack of consensus.	Statistically sound empirical data, but conceptual discussion limited

18.	2006	Levy and Czernecki Apathy and the basal ganglia	Discussion | Discussion of apathy and its neurological correlates, specifically how apathy affects the prefrontal-basal ganglia	Clinical Neurology	(Processing of external and internal factors → intention to act) + (planning and initiating action) = goal-directed behaviour; ↓ of goal-directed behaviour, therefore quantitative ↓ of self-generated, voluntary, purposeful behaviour = apathy Three subtypes; emotional, cognitive, auto-activation: ↓ of willingness to maintain action + ↓ of completion of actions + ↓ ability to evaluate actions = emotional apathy ↓ of manipulation of goals (↓ of working memory) + difficulty in generating rules + difficulty in changing tasks = cognitive apathy ↓ initiation of action/thought + ↓ of external response + ↓ of automatic initiation of thought = auto-activation apathy (severe apathy)	Extensive discussion of apathy. Purposefully presents an opposition to Marin's dominant definition; moves away from apathy as primarily loss of motivation, instead focussing on quantifiable reduction in GDB. Concentrates on possible neurological aspects of apathy. Different model of apathy than has previously been described – focusses on localisation rather than symptom identification. Conceptualisation of apathy relies purely on neurological theory and ignores the psychosocial and socioenvironmental factors that may have a role in apathy.	Neurological data presented as evidence for neurological location and identification of apathy.

19.	2006	Levy and Dubois Apathy and the functional anatomy of the prefrontal cortex-basal ganglia circuits	Discussion | Description of apathy based on a new model of apathy	Clinical Neurology Neurophysiology	(Processing of external and internal factors → intention to act) + (planning and initiating action) = goal-directed behaviour, ↓ of goal-directed behaviour, therefore quantitative ↓ of self-generated, voluntary, purposeful behaviour = observable, objective behavioural syndrome, independent of psychological interpretation = apathy	Extensive discussion of apathy and good discussion of the conceptualisation of apathy. Splits the underlying mechanisms of apathy into three types of "disrupted processing" (p916); emotional-affective, cognitive, and auto-activation. General lack of impact of socioenvironmental factors (as #18). Tries to move apathy away from psychological symptom identification (overly subjective) and towards objective measure of behaviour and neurology	Theoretical evidence presented, combined with neurological data from papers on neurological make-up of the brain
20.	2006	Richard Apathy does not equal depression in Parkinson disease: Why we should care	Discussion | Short editorial piece reinforcing Kirsch-Darrow et al.'s (2006) article on apathy and depression in PD	Clinical Neurology	↓ goal-directed speech + ↓ motor activity + ↓ emotion + indifference + neutral mood = apathy	Apathy discussed briefly with limited discussion of variability in literature.	Some evidence from #17 presented

21.	2006	Sockeel, Dujardin, Devos, et al. The Lille apathy rating scale (LARS), a new instrument for detecting and quantifying apathy: validation in Parkinson’s disease	Measure | Development of the LARS apathy scale for use in PD	Clinical Psychometrics Neurology Psychiatry	↓ of motivation/↓ of initiative + ↓ interest + ↓ participation in activities of daily living + early withdrawal from initiation + indifference + flat affect = apathy	Apathy discussed and authors acknowledge conceptual difficulty and variability in literature. Authors quote other definitions without then clearly laying out their own definition or interpretation of the definitions available.	Statistically sound measure development, but lack of conceptual discussion
22.	2006	Starkstein, Jorge, and Mizrahi The prevalence, clinical correlates and treatment of apathy in Alzheimer’s disease	Discussion | Article exploring the prevalence, clinical correlates, and treatment of apathy in AD with reference to the SCIA and other measures of apathy	Clinical Psychiatry	↓ goal-orientated behaviours + ↓ cognition →↓ motivation = apathy	Apathy discussed and apathy is conflated with diminished motivation. Authors’ conceptualisation of apathy unclear. Discussion of different diagnostic criteria but no acknowledgement of the difficulties this presents discussing different prevalence rates or definitions of apathy.	Some tentative evidence for possible treatment routes suggested, but independent conceptualisation of apathy not present
23.	2006	Starkstein, Jorge, Mizrahi, and Robinson A prospective longitudinal study of apathy in Alzheimer’s disease	Discussion | Study evaluating the presence of apathy and its relation to disease severity functional decline	Clinical Neurology Psychiatry	↓ goal-directed cognition + ↓ goal-directed behaviour → ↓ motivation relative to individual’s normal functioning = apathy	Apathy briefly discussed, but only one definition presented and no conceptual discussion	Statistical evidence for links between apathy, older age, depression, and worse disease progression and severity. Concept not evidenced

24.	2008	Leentjens, Dujardin, Marsh, et al. Apathy and Anhedonia Rating Scales in Parkinson's Disease: Critique and Recommendations	Review | Systematic review of apathy and anhedonia scales used and validated in PD	Clinical Neurology	↓ of motivation = apathy	Apathy very briefly discussed conceptually as paper focusses on measurements. Briefly presents Marin's diagnostic criteria but does not expand or explain them.	Evidence for and against measures of apathy presented; lack of conceptual discussion. Discussion of lack of consensus in field at end of article but no explicit explanation of these difficulties or how this affects the different measures of apathy.
25.	2008	Pedersen, Larsen, and Aarsland Validation of the Unified Parkinson's Disease Rating Scale (UPDRS) section I as a screening and diagnostic instrument for apathy in patients with Parkinson's disease	Measure | Validation of parts of the UPDRS for use in assessing apathy in PD	Clinical Neurology	↓ of goal-directed cognition and behaviour → ↓ of motivation = apathy	Apathy discussed briefly, again with only one definition presented. Some discussion of negative impact of apathy on patient and caregiver	Statistical validation of measure sound, but concept discussion lacking

26.	2008	Starkstein and Leentjens The nosological position of apathy in clinical practice	Discussion | Discussion of consensus in the field of apathy research and its necessity to further our understanding	Clinical Neurology Psychiatry	↓ goal-directed cognition + ↓ goal-directed behaviour → ↓ motivation (+ maybe ↓ emotion) = apathy	Apathy discussed at length, with good acknowledgement of lack of consensus in definition and conceptualisation. Concept developed, e.g. introducing time component to diagnostic criteria.	Directly addresses nosology of apathy using previous work and novel observations. Points out lack of clarity in field
27.	2009	Naarding, Janzing, Eling, van der Werf, and Kremer Apathy Is Not Depression in Huntington's Disease	Discussion | Evaluation of the overlap between apathy and depression in HD	Clinical Neuropsychiatry	Cognitive + motor + emotional components = apathy	Apathy discussed briefly but no conceptual discussion.	Statistical evidence for prevalence of apathy and distinction from depression, but conceptual discussion missing
28.	2009	Reyes, Viswanathan, Godin, et al. Apathy: A major symptom in CADASIL	Discussion | Examines frequency of apathy in SIVD, specifically CADASIL. Compares apathy and depression rates and symptom overlap	Clinical Neurology	Quantitative ↓ of voluntary goal-directed behaviour + ↓ interest + indifference + flat affect + ↓ of emotion = apathy	Apathy discussed by referencing other definitions without critical analysis. Levy and Dubois relied on heavily	Statistical analysis of data sound, but lack of conceptual discussion. Acknowledgement that conceptual discussion is lacking in the field

29.	2009	Robert, Onyike, Leentjens, et al. Proposed diagnostic criteria for apathy in Alzheimer's disease and other neuropsychiatric disorders	Discussion | Diagnostic criteria presented and discussed	Clinical Psychiatry	↓ of motivation + ↓ initiative + ↓ interest + blunt emotion + indifference + ↓ insight = apathy	Apathy discussed at length, broadly, and with specific reference to conceptualisation and diagnostic criteria. Conceptually, the criteria are like Marin's and Starkstein's and centre on a loss of motivation combined with other factors.	Good discussion for new diagnostic criteria, although formulation of criteria slightly lacking and largely unchanged from Marin or Starkstein (this is acknowledged). Validation of criteria needed. Still could be argued that this is a clinical description not a conceptualisation
30.	2009	Starkstein, Merello, Jorge, et al. The Syndromal Validity and Nosological Position of Apathy in Parkinson's Disease	Discussion | Validation of diagnostic criteria for apathy and relation to different types of depression and dementia	Clinical Neurology	Flat affect + ↓ goal-related behaviour + ↓ goal-related thought content → (↓ motivation + ↓ interest) = apathy	Apathy discussed but limited discussion of conceptualisation of apathy despite focus on diagnostic criteria	Novel empirical data presented, further reinforcing previous work describing apathy as a separate syndrome to depression. Conceptual discussion of apathy still somewhat limited

31.	2009	Tagariello, Giardi, and Amore Depression and apathy in dementia: Same syndrome or different constructs? A critical review	Discussion | Paper examines the overlap and differences between apathy and depression in dementia	Clinical Geriatrics Psychiatry	↓ goal-directed behaviour + ↓ goal-related thought content + flat affect ↓ motivation w/o ↓ consciousness, cognitive impairment, emotional distress = apathy	Apathy only briefly discussed, mainly with reference to Marin's definition. Paper notes that apathy's nosological position is still broadly unknown.	Evidence presented for apathy and depression to be separate constructs, but no discussion of difference conceptually
32.	2010	de Medeiros, Robert, Gauthier, et al. The Neuropsychiatric Inventory-Clinician rating scale (NPI-C): reliability and validity of a revised assessment of neuropsychiatric symptoms in dementia	Measure | Development of the NPI-C to assess validity of clinician-based rating scale in dementia.	Clinical Geriatrics Psychaitry	Apathy not discussed No concept or definition	Refers to #29 diagnostic criteria, but no discussion. States that apathy is identifiable by a knowledgeable clinician, but does not state how.	

33.	2010	Drijgers, Dujardin, Reijnders, et al. Validation of diagnostic criteria for apathy in Parkinson's disease	Discussion | Development and validation of Robert et al (2009) diagnostic criteria for apathy in PD.	Clinical Psychiatry Neurology	Apathy discussed in broad terms but no specific concept or definition	No concept or definition	Positive evidence for #29 diagnostic criteria, but no conceptual critique or discussion of the criteria
34.	2011	Bogart Is Apathy a Valid and Meaningful Symptom or Syndrome in Parkinson's Disease? A Critical Review	Review | Critical review of apathy in PD, specifically looking at impact of apathy.	Clinical Health psychology Neurology	↓ of motivation = apathy Either symptom or syndrome, ↓ of initiation → no response to stimuli = apathy	Apathy discussed both in terms of definition and conceptualisation Purposefully does not definitively conceptualise apathy Construct validity of apathy cannot be determined until a consensus has been reached on definition	Empirical and theoretical evidence presented for apathy's lack of standardisation. Suggestions of behaviours that could present as apathy but are symptoms of PD, therefore apathy may not be clinically useful
35.	2011	Chase | Apathy in Neuropsychiatric Disease: Diagnosis, Pathophysiology, and Treatment	Discussion | Overview of research into apathy with recommendations for future research	Clinical Neurology Toxicity	↓ of motivation + ↓ of interest + ↓ of concern + ↓ of volitional goal-directed behaviour + ↓ of emotional responsivity + ↓ cognitive activity → ↓ of emotional distress ↓ intellectual impairment + ↓ consciousness = apathy	Apathy discussed and identified primarily as a disorder of motivation. Acknowledges lack of consensus and presents multiple interpretations of apathy. Conceptual discussion slightly lacking	Presents evidence for lack of consensus but does not relate this to treatment options presented

36.	2011	Clarke, Ko, Kuhl, et al. Are the available apathy measures reliable and valid? A review of the psychometric evidence	Review | Looks specifically at a wide range of apathy measures available and gives an overview of their development.	Clinical Psychiatry	↓ of motivation + ↓ of initiation + change in affect/behaviour/cognition = a form of apathy [apathy as not just a single syndrome]	Concept of apathy not explicitly discussed by the authors other than pointing out that it is ill-defined	Evidence presented for measures of apathy rather than looking at concept of apathy
37.	2011	Krishnamoorthy and Craufurd Treatment of Apathy in Huntington's Disease and Other Movement Disorders	Discussion | Reflection on possible treatments for apathy in HD and other conditions.	Clinical Psychiatry Neurology	↓ emotional response +/ flat affect + ↓ in goal-directed behaviour cognitively and emotionally = apathy	Different definitions of apathy presented, mostly in a descriptive fashion. States that concept and nosology of apathy has changed over time but is unclear about lack of consensus, implying consensus has been reached. Acknowledges similarities between apathy and depression but does not offer any way to deal with this clinically	Treatments suggested based on current best evidence, but conceptual discussion of apathy lacking
38.	2011	Mulin, Leone, Dujardin, et al. Diagnostic criteria for apathy in clinical practice	Discussion | Evaluation of diagnostic criteria proposed by Robert et al. (2009).	Clinical Psychiatry Geriatrics	↓ of motivation for 4 weeks + ↓ goal-directed behaviour/↓ goal-directed cognitive activity/↓ emotion + functional impairments caused by apathy w/o symptoms/ conditions mimicking apathy = apathy	Apathy discussed mainly with reference to #29 diagnostic criteria, no original definition. Does slightly alter #29 diagnostic criteria without stating why Reported 94% presence of apathy in participants with major depressive disorder extremely high.	Presents reasonable empirical data supporting diagnostic criteria, but lack of conceptual discussion

39.	2011	Schrag Apathy and depression scales in Parkinson's disease: Are they good enough?	Discussion | Overview of the current validated apathy and depression scales available	Clinical Neurology	↓ of motivation for 4 weeks + (≥2 of ↓ goal-directed behaviour + ↓ goal-directed cognitive activity + ↓ emotion) + functional impairments attributable to apathy = apathy	Apathy definition very briefly discussed, with reference to #29 diagnostic criteria	Evaluation of measures useful, but no conceptual discussion of apathy
40.	2011	Starkstein and Brockman Apathy and Parkinson's Disease	Discussion | Discussion of possible treatment regimens for apathy based on treatment for other mood disorders	Clinical Neurology Psychiatry	↓ feeling + ↓ emotion + ↓ interest + ↓ concern + ↓ of motivation = apathy	Apathy defined and discussed clinically in terms of underlying mechanism and treatment possibilities, but not clear conceptually. Starkstein's diagnostic criteria referred to but not discussed	Conceptual discussion lacking
41.	2012	Apathy: A pathology of goal-directed behaviour. A new concept of the clinic and pathophysiology of apathy	Discussion | Conceptual discussion of apathy with specific reference to how apathy may occur neurologically.	Clinical Neurology	Quantitative ↓ of voluntary, adaptive, goal-directed behaviour w/o environmental or physical constraints GDB = processing of external and internal determinants that influence the intention to act, elaboration of the plan of actions, initiation, execution, feedback control. Three subforms of apathy: • Cognitive • Emotional-affective • Auto-activation	Apathy discussed at length with a new definition and conceptualisation of apathy presented and analysed. Concept relies entirely on neurological mechanisms to identify apathy and does not discuss psychosocial or socioenvironmental factors. Limitations but useful model for discussion	Presents interesting definition, concept, and model of apathy based on previous research and observations in the literature. Empirical evidence lacking but theoretical model explained

42.	2012	Mortby, Maercker, and Fortsmeler Apathy: a separate syndrome from depression in dementia? A critical review	Discussion | Overview of the papers that have explored the overlap between apathy and depression in various forms of dementia	Clinical Experimental Ageing	↓ of motivation w/ ↓ initiation + poor persistence + ↓ interest + indifference + ↓ social engagement + flat affect + ↓ insight w/o ↓ consciousness/ cognitive impairment/ emotional distress + behaviour compared relatively to previous functioning and cultural norms = apathy	Apathy discussed and multiple definitions presented. Good acknowledgement of various issues in the study of apathy and depression. Analysis of comparing of different definitions present. Slightly unclear what authors think apathy is, given the variety of definitions presented	Presents evidence based on literature that apathy is a separate construct to depression. Acknowledges lack of consensus, but does not present independent conceptualisation
43.	2012	Starkstein Apathy in Parkinson's Disease: Diagnostic and Etiological Dilemmas	Discussion | Critical review of recent studies looking at the phenomenology, diagnosis, underlying mechanisms, and treatment of apathy	Clinical Psychiatry Neurology	↓ of motivation + ↓ emotion = apathy	Apathy discussed and concepts of apathy analysed and countered. Slightly lacking in own definition and conceptualisation of apathy in favour of analysis and critique of other theories. Difficult to know where to position apathy nosologically with problems associated with each theory	Theoretical evidence against other definitions and models presented. No reinterpretation or novel conceptualisation offered

44.	2013	Arnould, Rochat, Azouvi, and Van der Linden A Multidimensional Approach to Apathy after Traumatic Brain Injury	Discussion | Discursive paper looking at apathy in traumatic brain injury (TBI). Frames apathy and discusses wide-ranging issues around definition and measurement of apathy	Clinical Neurology Psychology Psychopathology	↓ goal-directed cognition + ↓ goal-directed behaviour + flat affect → ↓ of motivation = apathy Four proposed dimensions of apathy = cognitive + affective + motivational + symptoms related to personal identity	Apathy discussed widely and conceptually. Good analysis of apathy literature to date. Slightly descriptive in places. Offers novel interpretation and conceptualisation of apathy in presenting a multidimensional framework of apathy that moves the discussion of apathy away from a descriptive, symptom-based diagnostic model to a model that considers underlying mechanism alongside symptoms	Novel model of apathy presented with evidence from literature to reinforce points made
45.	2013	Carvalho, Ready, Malloy, and Grace Confirmatory Factor Analysis of the Frontal Systems Behavior Scale (FrSBe)	Measure | Development and revision of the FrSBe (formerly FLoPS), broad applicability	Clinical Psychometrics Psychiatry	None	Apathy definition not discussed. No concept offered. Brief discussion of apathy index on FrSBe that suggests apathy involves difficulty with initiation, psychomotor retardation, spontaneity drive, persistence, loss of energy and interest, lack of concern about self-care, and/or blunted affective expression	No evidence presented

46.	2013	Santangelo, Trojano, Barone, et al. Apathy in Parkinson's disease: Diagnosis, neuropsychological correlates, pathophysiology and treatment	Discussion | Overview of various issues in the field of apathy, including definition, correlates and concomitants, and treatment. Not systematic review but very detailed	Clinical Neurology Psychology	↓ of motivation w/o ↓ consciousness/ cognitive impairment/ emotional distress = apathy / pathology of voluntary action or GBD [sic] arising from alterations occurring at the level of elaboration, execution and/or control of GBD [sic] = apathy	Apathy discussed and quotes from Marin and Levy and Dubois used as definition. No addition or analysis. Conceptualisation limited	Confusing presentation of conflicting evidence from different papers. No statement clarifying with which the authors agree
47.	2014	Cipriani, Lucetti, Danti, and Nuti Apathy and Dementia. Nosology, Assessment and Management	Review | Discusses apathy in relation to various illnesses and recommends way forward for apathy research	Clinical Neurology Psychiatry	Reduction of voluntary goal-directed behaviour + ↓ of motivation + ↓ of initiation = apathy [as an observable behavioural syndrome]	Apathy discussed with recognition that consensus is still a problem. Descriptive rather than analytical	Some evidence from literature presented but not used to build up a clear picture of apathy
48.	2014	Massimo, Evans, and Grossman Differentiating Subtypes of Apathy to Improve Person-Centered Care in Frontotemporal Degeneration	Discussion | Uses a model of GDB to apply targeted interventions to people with apathy in bvFTD, discusses tailored treatment of apathy, and introduces the PACT	Clinical Neurology Nursing	↓ of initiation / planning / motivation →↓ of self-generated/ voluntary GDB = apathy (based on Levy and Dubois, 2006)	Model of apathy presented is combination of Levy and Dubois' (2006) neurological/behavioural model, Brown and Pluck's (2000) GDB, and Roberts et al.'s (2009) diagnostic criteria. Authors state that apathy emerges when there is dysfunction at any stage of the GDB process and apathy is a pathology of GDB	Combination of literature useful, and evidence pulled from each paper well explained

49.	2014	Radakovic and Abrahams Developing a new apathy measurement scale: Dimensional Apathy Scale	Measure | Development of the DAS for use with patients with motor dysfunction	Clinical Psychometrics Psychology Psychiatry	(↓ of energy + ↓ of interests + ↓ of emotion → ↓ of motivation towards goal-directed behaviour) + constantly = apathy	Various definitions of apathy discussed. Apathy defined and conceptualised based on a combination of Marin and Levy and Dubois' models of apathy. Justification given where possible and explained in theoretical terms. Authors see apathy as multidimensional, in-line with the triadic factor view adopted by most authors	Good evidence-based conceptualisation using previous literature. Measure development well described. Focusses on measure development rather than conceptual development
50.	2015	Agueera-Ortiz, Gil-Ruiz, Cruz-Orduna, et al. A Novel Rating Scale for the Measurement of Apathy in Institutionalized Persons with Dementia: The APADEM-NH	Measure | Development of a novel apathy measure for people with dementia in nursing homes	Clinical Psychometrics Psychiatry Neurology Geriatrics	Persistent ↓ of motivation, feelings, emotions, and/or interests → significant ↓ in self-generated behaviours aimed at a goal = apathy	Apathy discussed, major definitions presented. Possibly over-extends Levy and Dubois' model, suggesting specific pseudo-diagnostic criteria where none have been yet suggested. Slightly unclear which definition the authors favour and how they are defining apathy for the purposes of the measure	Measure well developed statistically, but conceptualisation of apathy limited

51.	2015	Massimo, Powers, Evans, et al. Apathy in Frontotemporal Degeneration: Neuroanatomical Evidence of Impaired Goal-directed Behavior	Discussion | Exploration of apathy in bvFTD using novel computer-based reaction time assessment	Clinical Neurology Neuroscience Nursing	Quantitative ↓ of self-generated, voluntary and purposeful GDB, including at least initiation, planning, and motivation = apathy	Apathy defined in various ways and a clear statement given as to how the authors choose to view apathy – following and building on Levy and Dubois (2006). Explain that GDB is an essential part of decision making in life which facilitates intended outcomes and is impaired in apathy	Good discussion of apathy, although tentative in places – development of conceptualisation seems on-going. Unclear in places
52.	2015	Radakovic, Harley, Abrahams, and Starr A systematic review of the validity and reliability of apathy scales in neurodegenerative conditions	Review | Review of the available measures of apathy, specifically looking at neurodegenerative conditions	Clinical Psychiatry Geriatrics Psychology	↓ of energy + ↓ of interests + ↓ of emotion → ↓ of motivation = apathy, undesirable syndrome/symptom	Various definitions of apathy are presented with some comparison between other mood disorders suggested. Lack of firm statement of what authors believe apathy to consist.	Evidence presented for reliability of apathy measures but conceptual discussion lacking
53.	2015	Weiser and Garibaldi Quantifying motivational deficits and apathy: A review of the literature	Review | Systematic review of the measures available to assess apathy with future direction for research suggested	Clinical Psychiatry	The broad concept of apathy is mentioned but no discussion of specific definitions. Refers to Robert et al.’s (2009) diagnostic criteria but does not outline or explain.	Definitions and concepts are not discussed	Evidence presented for reliability of apathy measures but no conceptual discussion

54.	2016	Fitts, Massimo, Lim, et al. Computerized assessment of goal-directed behaviour in Parkinson's disease	Measure | Reporting of development of a novel measure of apathy that involves using the Starkstein Apathy Scale and a reaction test	Clinical Neuropsychol	Apathy = reduction in GDB, see number 48 above for description of GDB (same authors)	Relatively little conceptual discussion of apathy.	Interesting concept, relating reaction time to initiation, planning, and motivation parts of GDB and apathy (not included in measures as no novel items for analysis).
55.	2017	Lanctot, Aguera-Ortiz, Brodaty, et al. Apathy associated with neurocognitive disorders: recent progress and future directions	Discussion | Discursive article that extensively discusses the current research into apathy from a variety of perspectives	Clinical Neurocog	Apathy = primarily ↓ motivation	Cognitive, affective, behavioural dimensions all part of same syndrome = apathy.	Notable as pushes for further work defining as a syndrome, not a symptom
56.	2017	Ang, Lockwood, Apps, et al. Distinct subtypes of apathy revealed by the Apathy Motivation Index	Measure | Development of the AMI measure of apathy and reporting of subtypes of apathy	Research	Apathy = disorder of motivation characterised by reduced action initiation and GDB.	Divides apathy into a three-factor model that consists three distinct types of apathy; generally apathetic, emotionally apathetic, behaviourally/socially apathetic	Points out that apathy occurs in neuro- disorders and healthy people. Interesting measure with useful subtypes.

57.	2017	Gelderblom, Wuestenberg, McLean et al. Bupropion for the treatment of apathy in Huntington's disease	Treatment | Phase 2b trial into the efficacy of a pharmacological treatment of apathy in HD.	Clinical Neurocog	Apathy = absence of motivation, lack of initiative and drive, emotional indifference. Apathy is subdivided into deficient emotional-affective function, cognitive function, or auto-activation.	Little conceptual study of apathy.	Interesting conclusion that drug ineffective but trail involvement effective. Implications for future talking therapy-based treatment.
58.	2017	Radakovic, Davenport, Starr, et al. Apathy dimensions in Parkinson's disease	Discussion | Testing of the Dimensional Apathy Scale for use in PD.	Clinical Neurocog Psychiatry Geriatrics	No development over paper #52, above – same authors.	No development over paper #52, above – same authors.	Reports that the DAS is effective in PD – originally developed in a health population.
59.	2018	Valentino, Alessandro, Amboni, et al. Apathy in Parkinson's disease: differences between caregiver's report and self-evaluation	Discussion | Looks at the relationship between caregiver and self-report of apathy using the AES.	Neurology	Apathy = diminished goal-directed speech, motor activity, and emotions	Three sub-types of apathy following Levy and Dubois; apathy = combination of auto-activation, emotional-affective, and cognitive apathies.	Significant discrepancies exist between caregiver and self-report of apathy.

60.	2018	Santangelo, D'Iorio, Maggi, et al. Cognitive correlates of pure apathy in Parkinson's disease	Discussion | Assesses apathy while controlling for symptoms of PD in apathetic patients.	Neurology	Apathy = primarily ↓ motivation	Apathy is a distinct psychiatric syndrome characterised by simultaneous diminution in the cognitive and emotional concomitants of GDB.	Interesting conclusion that supports the idea that neurological damage worsens apathy and executive function deficit.
61.	2018	Radakovic and Abrahams Multidimensional apathy: evidence from neurodegenerative disorders	Discussion | Presents a novel framework of apathy, the Dimensional Apathy Framework	Clinical Behaviour	No development over paper #52, above – same authors. Development of work into multi-dimensional aspect of apathy rather than definition itself	Concept based on three sub-types of apathy; initiation, executive, and emotional apathy, all moderated by a person's self-awareness	Novel framework that develops further understanding of apathy using data from a broad range of literature
62.	2018	Robert, Lanctot, Aguera-Ortiz, et al. Is it time to revise the diagnostic criteria for apathy in brain disorders?	Discussion | Revised diagnostic criteria for apathy, updated Robert et al. 2009	Psychiatry	Apathy = lack of motivation that persists over time and causes impairment	Three dimensions of apathy; deficits in GDB, goal-directed cognitive activity, and emotions	Updated diagnostic criteria that are further reported below

63.	2018	Henstra, Feenstra, van der Valde, et al. Apathy is associated with greater decline in subjective, but not in objective, measures of physical functioning in older people without dementia	Discussion | Reporting of a study that evaluated association between apathy and physical functioning in older people without dementia	Geriatrics Gerontology	Apathy = lack of motivation that is not solely attributable to diminished level of consciousness, cognitive impairment, or emotional distress.	Apathy = reduction in GDB, goal-directed cognition, and emotional expression	People with apathy tended to report lower activity levels even when they remained the same. A decline in functioning usually followed, however.
64.	2018	Caravaggio, Fervaha, Menon, et al. The neural correlates of apathy in schizophrenia: An exploratory investigation	Discussion |	Clinical Neuropsychol Biological	Defines apatheia briefly Apathy = clinical syndrome, cognitive and behavioural disturbance in drive and motivation rather than emotional expression.	Apathy = deficit in self-initiated GDB Also, apathy = global score from Avolition-Apathy subscale of SANS	Partial reliance on SANS, a particularly old measure, slightly unusual. Global score of apathy difficult to rely on.
65.	2019	Carrozzino Clinimetric approach to rating scales for the assessment of apathy in Parkinson's disease	Review | Systematic review that uses a clinimetric approach to measures rather than a psychometric approach	Clinical Neuropsychol	Also defines apatheia briefly and points out that it was seen as a virtuous state. No specific definition of apathy	Apathy is characterised by a constellation of behavioural, cognitive, and emotional symptoms	Points out problems with psychometric testing of measures of apathy, particularly in relation to detecting change over time

10.3 Table 2 – Abridged versions of diagnostic criteria:

Marin, 1991		**Starkstein et al., 2001**		**Robert et al., 2009**		**Robert et al., 2018**	
Lack of motivation that is not attributable to intellectual impairment, emotional distress, or diminished level of consciousness (drowsiness and/or diminished attentional capacity)		Lack of motivation relative to the patient's previous level of functioning		A. Loss of or diminished motivation in comparison to the patient's previous level of functioning		A. A quantitative reduction of goal-directed activity in either behavioural, cognitive, emotional, or social dimensions in comparison to the patient's previous level of functioning in these areas.	
A. Lack of motivation, relative to the patient's previous level of functioning		Presence, with lack of motivation, of at least one symptom belonging to each of the following three domains:		B. Presence of at least one symptom in at least two of three following domains for a period of at least four weeks, present most of the time:		B. Presence of at least one symptom in at least two of three following domains for a period of at least four weeks, present most of the time:	
A1. Diminished GDB:	Lack of productivity, time spent in activities of interest, initiative, perseverance	Diminished GDB:	Lack of effort	B1: Loss of, or diminished, GDB:	Loss of self-initiated behaviour	B1. Behaviour and cognition: Loss of GDB/ cognition as evidenced by:	Reduced activity level/requires prompting
							Less persistent in maintaining conversation
	Behavioural compliance/depend				Loss of environment-stimulated behaviour		Makes fewer choices
							Reacts less to good/bad news

	ency on others to structure activity,		Dependency on others to structure activity				Less interest in wellbeing and personal image
	Diminished socialisation or recreation	Diminished goal-directed cognition	Lack of interest in new things	B2. Loss of, or diminished, goal-directed cognition	Loss of spontaneous ideas and curiosity	B2 Emotion: Loss of emotion as evidenced by:	Less spontaneous emotions regarding self
							Less emotional reaction to positive or negative events
			Lack of concern		Loss of environment-stimulated ideas and curiosity		Less concerned about impact on others
							Shows less empathy
							Less demonstration of emotional reactions
B. Lack of motivation is not attributable to intellectual impairment, emotional distress, or diminished level of consciousness		Diminished concomitants of GDB	Unchanging affect	B3. Loss of, or diminished, emotion	Loss of spontaneous emotion	B3: Social interaction	Less initiative in proposing social activity
							Participates less in social activity
							Less interest in family members

				Loss of emotional responsiveness		Less initiation of conversation
						Prefers to stay at home
		Lack of emotional responsivity to positive or negative events	Symptoms A and B cause clinically significant impairment		Criterion C: Symptoms A and B cause clinically significant impairment in personal, social, occupational, or other important areas of functioning	
C. Emotional distress is absent or is insufficient to account for the lack of motivation	The symptoms cause clinically significant distress or impairment		Symptoms A and B are not exclusively explained or due to physical disabilities, motor disabilities, diminished level of consciousness, or substance		Criterion D: Symptoms A and B are not exclusively explained or due to physical disabilities, motor disabilities, diminished level of consciousness, direct effects of a substance, or major changes in a patient's environment	
	The symptoms are not due to a diminished level of consciousness or substance					

10.4 Table 3 – Validated measures of apathy:

#	Measure	Author(s)	Items	Items used	Comment
1.	Scale for the Assessment of Negative Symptoms (SANS)	Andreasen, 1982	4	Grooming and hygiene Impersistence at work or school Physical anergia Global rating of avolition-apathy	Subscale, clinician administered to patient with input from family, developed for people with schizophrenia, variant has 4 items with subjectivity item dropped
2.	Irritability-Apathy Scale (IAS)	Burns, Folstein, Brandt, and Folstein, 1990	5	Interest in everyday events Time spent doing nothing (i.e. sitting in a chair w/o TV etc.) Active in day to day activities Busyness throughout day Withdrawal level	Subscale, clinician administered to carer, limited instruction suggests open-ended questions, assessed patient with either AD, HD, or dementia
3.	Apathy Evaluation Scale (AES-S/I/C)	Marin, Biedrzycki, and Firinciogullari, 1991	18	Interest Completes things Own initiative is important Interested in new experiences Interested in learning new things Little effort into things Intensity in life Finishing things is important Spends time doing things that are interesting (to them)	First full scale, commonly used as gold standard (although this is contentious), clinician administered to patient or carer, or self-report

				Someone else must initiate activities Lack of concern about problems Has friends Meeting friends is important Excited by good things that happen Accurate understanding of problems Completing things is important Initiative Motivation	
4.	Apathy Scale (AS)	Starkstein, Mayberg, Preziosi, et al., 1992	14	Interested in new things Interested in anything Concerned about condition Puts effort into things Always looking for something to do Plans/goals for future Motivation Energy for daily activities Someone tells them what to do every day Indifference Unconcerned about things Need a push to start things Neither happy nor sad, just in between	Full scale, adapted version of Marin's AES, simplified version of the AES for use with PD, clinician administered to patient

				Apathetic	
5.	Apathy Inventory (IA)	Robert, Clairet, Benoir, et al., 2002	3	Emotional blunting – affection and emotion level Lack of initiative – initiates conversation/decision Lack of interest – interest in others plans, interest in friends/family, enthusiastic about interests/profession	Full scale, clinician administered to either patient or carer, delivered verbatim but prompts if necessary, used with AD, PD, or MCI
6.	Dementia Apathy Interview and Rating (DAIR)	Strauss and Sperry, 2002	16	Indifference Important to succeed in things Sits and does nothing Less active Keeps busy throughout day Initiates activities Enthusiastic about things Full range of emotion As reactive to things as used to be Starts conversations Less spontaneous Interested in friends/family Suggests things to do Enjoys things like did before illness Concerned about others' feelings Cares less about finishing things	Full scale, clinician administered to carer (although paper conflicts on this point), administered as an interview, used with probable/possible AD

7.	Key Behaviors Change Inventory (KBCI)	Kolitz, Rodney, Vanderploeg, and Curtiss, 2003	8	Four positive, four negative items	Subscale, clinician administered to patient, used with traumatic brain injury
8.	Structured Clinical Interview for Apathy (SCIA)	Starkstein, Ingram, Garau, and Mizrahi, 2005	Int	Questions related to: Lack of motivation relative to previous functioning Lack of effort to perform activities Dependency on others to structure activity Lack of interest in learning new things Lack of concern about personal problems Unchanging/flat affect Lack of emotional response to +ve or -ve events	Full scale, only used in this paper, does not seem to have been used since, sparse details, clinician administered structured interview with patient. Measure in development and being tested in stroke and PD (personal communication).
9.	Lille Apathy Rating Scale (LARS)	Sockeel, Dujardin, Devos, et al., 2006	33	Everyday productivity – day-to-day life Interests – keep yourself occupied Initiative – do things on own Novelty seeking – find new things Motivation/voluntary actions – easy to do things Emotional responses – easily emotional? Concern – problems cause worry Social life – lots of friends Self-awareness – think about own actions	Full scale, nine domains, clinician administered to patient, verbatim, although prompts allowed if necessary
10.	Neuropsychiatric Inventory (NPI)	Medeiros, Robert, Gauthier, et al., 2010	9+3	Interest Motivation Difficult to engage in conversation/chores	Subscale, nine initial items and three items related to perceived severity of apathy, second most used apathy scale despite being subscale, clinician administered to patient

				Apathy or indifference present Four questions above lead to: Spontaneity Initiation Comparative reduction in affection/emotion Contributes less to chores Less interested in plans/activities Less enthusiastic about interests Signs of not caring	
11.	Frontal Systems Behavior Scale (FrSBE)	Carvalho, Ready, Malloy, and Grace, 2013	14	Speaks only when spoken to Lack initiative, motivation Neglects personal hygiene Does nothing Incontinence Lost interest in things Does not finish things Unconcerned and unresponsive Lacks energy Is interested in sex Cares about appearance Gets involved spontaneously Does things without reminders	Subscale (originally developed 2005 by Malloy and Grace and called FLOPS), clinician administered to patient, 1-5 Likert-type scale, rated on before-and-after illness onset

				Starts conversations	
12.	Dimensional Apathy Scale (DAS)	Radakovic and Abrahams, 2014	24	Time based answers to: Need encouragement Contact friends Express emotions Do new things Concerned about family Staring into space Consider others when doing something Plan days in advance Bad news make patient feel bad Focusses until task finished Lack motivation Struggle with empathy Set goals Try new things Unconcerned about others Act on thoughts during day Difficulty with demanding tasks Keep busy Easily confused when multitasking Become emotional easily when watching TV	Full scale, dimensional approach unusual, clinician administered to patient,

				Lack of concentration Spontaneous Easily distracted Indifferent	
13.	Apathy in Dementia, Nursing Home (APADEM-NH)	Aguera-Ortiz, Gil-Ruiz, Cruz-Orduna et al., 2015	26	**Deficit of thinking** Reacts to name Reacts to person appearing Reacts to family/friend Touches people around them Change position when uncomfortable Expresses pleasure at smell/taste/touch/sound Initiates fun activity Respond to uncomfortable situation Interested in health Interested in appearance Interested in family/friends Interested in leaving residence for activities Interested in new things **Emotional blunting** Shows affection Reacts positively to acknowledgement of completed task	Full scale, developed specifically for use in a nursing home, clinician administered to professional caregiver (unclear if this precludes family members)

				Reacts when feels attacked/upset Different emotions to different meals Talks about others' feelings Show emotion when hear a story Show emotion at new activity **Cognitive inertia** Engages in new activity Once given tools to perform activity, performs immediately Persists in activity until completion Asks for help if can't do something Choose easily between options Talks to people	
14.	Apathy Motivation Index (AMI)	Ang, Lockwood, Apps, et al.	18	**Behavioural activation** I make decisions firmly and without hesitation When I decide something, I make effort easily I don't like to laze around I get things done without being reminded When I decide something, I am motivated When I have something to do I do it immediately **Emotional sensitivity** I feel sad or upset when I hear bad news	Full scale, developed with only healthy participants, no illnesses. Development took place alongside use of LARS and authors report it is appropriate for use in healthy or clinical populations – future research should be undertaken to prove this.

				After a decision, I wonder about my choice In the last 2 weeks, I care deeply about others' opinion of me I feel awful if I say something insensitive I feel bad when I hear someone has an accident If I realise I have been unpleasant, I feel guilty **Social motivation** I start conversations with random people I enjoy doing things with people I have just met I suggest activities for me and my friends to do I go out with friends weekly I start conversations without prompt I enjoy choosing what to do	

10.5 PRISMA 2009 checklist for systematic review

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	35
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	Not appropriate in thesis format
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	35
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	37
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	38
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	40

Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	38
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	39
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	40
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	43
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	43
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	43
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	44
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	43
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	43
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	44

RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	41+42
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	198
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	43
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	198
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	N/A
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	N/A
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	44
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	59
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	72

Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	73
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	72

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(7): e1000097.

For more information, visit: www.prisma-statement.org.

10.6 Definitions (listed alphabetically)

AD – Alzheimer’s disease

ADL – Activities of Daily Living

AES – Apathy Evaluation Scale

AMI – Apathy Motivation Index

APA – American Psychiatric Association

APADEM-NH – Apathy in Dementia Nursing Home

AS – Apathy Scale

BCE – Behaviour, Cognition, Emotion

bvFTD – behavioural variant Frontotemporal dementia

CADASIL – Cerebral Autosomal Dominant Arteriopathy with Subcortical Infarcts and Leukoencephalopathy syndrome

DA – Discourse Analysis

DAF – Dimensional Apathy Framework

DAIR – Dementia Apathy Interview and Rating

DAS – Dimensional Apathy Scale

DSM – Diagnostic and Statistical Manual of Mental Disorders

FrSBE – Frontal Systems Behaviour Scale

FTD – Frontotemporal dementia

GDB – Goal-Directed Behaviour

GDC – Goal-Directed Cognition

GT – Grounded Theory

HD – Huntington’s disease

HIV – Human Immunodeficiency Virus

HYMS – Hull York Medical School

IA – Apathy Inventory

IAS – Irritability-Apathy Scale

ICD – International Classification of Disorders

IPA – Interpretative Phenomenological Analysis
KBCI – Key Behaviours Change Inventory
LARS – Lille Apathy Rating Scale
MCI – Mild Cognitive Impairment
NA – Narrative Analysis
NHS – National Healthcare Service
NPI – Neuropsychiatric Inventory
OED – Oxford English Dictionary
PD – Parkinson’s disease
PFC – Prefrontal Cortex
PSP – Progressive Supranuclear Palsy
SANS – Scale for the Assessment of Negative Symptoms
SCIA – Structured Clinical Interview for Apathy
TA – Thematic Analysis
TBI – Traumatic Brain Injury
TLF – Treasury of the French Language