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Comparing autism and OCD within a Compulsive and Repetitive Trait framework: do Free Will beliefs predict clinical symptoms?

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Abstract

Background Although a range of evidence suggests links between OCD and autism, there remains a lack of clarity on how symptoms may be related between these disorders. Repetitive traits are key components in both OCD and autism. Understanding the functions and origins of these traits is crucial. Repetitive traits in OCD are ego-dystonic, therefore related to distress. However, the nature of repetitive traits in autism is less clear. Historically, they were assumed to be ego-syntonic, therefore opposed to distress. However, recent evidence indicates ego-dystonic and ego-syntonic properties of repetitive traits may be demonstrated in autism. The main aim in the present thesis, therefore, is to investigate the relationship between mood and repetitive traits in autism and OCD. These findings would indicate whether disorders such as autism and OCD may be better understood within a Compulsive and Repetitive Trait (CaRT) framework. A pilot investigation is also put forward to investigate whether free will beliefs – an unstudied concept in autism research – may offer further insight into a CaRT framework.

Method A cross-sectional questionnaire method compared adults with autism, OCD and neurotypical peers on OCD traits, Repetitive Behaviours and Free Will beliefs.

Results Repetitive Behaviours were comparable in number and frequency between the OCD and autism groups, with higher positive mood in the autism group. OCD traits were highest in number and severity for the OCD group, although significantly higher in the autism compared to the control group. Groups did not differ on the presence of CaRTs due to social context. Strong correlations were identified between OCD traits and Repetitive Behaviours, despite no correlations being found between mood associated with these traits. No differences in free will beliefs were demonstrated

between the groups, although there was some indication of the significance of Scientific Determinism beliefs in autism.

Conclusions The research presented appears to support the usefulness of a CaRT framework to compare symptomology between autism and OCD. Mood appears to be an important factor in distinguishing between CaRTs. Comparable free will scores indicate clinical behaviours (CaRTs) may be relatively independent of free will beliefs. Limitations are discussed, which may have masked stronger evidence, such as the unrepresentative nature of the samples.

Keywords: autism, OCD, repetitive, mood, ego-dystonic, ego-syntonic, free will beliefs.

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Glossary of key terms

Autism – Autism Spectrum Disorder, a neurodevelopmental disorder with two main traits: social-communication deficits; and Repetitive Behaviours.

Compulsion – Typically, compulsions are clinically defined within the boundaries of OCD. The DSM-5 defines compulsions as repetitive traits performed to alleviate anxiety caused by intrusive obsessional content. However, compulsions may also be viewed generally as behavioural urges.

Compulsive and Repetitive Trait (CaRT) – Traits within a framework proposed within this thesis. These traits combine the assessment of OCD traits (measure of repetitive traits within OCD research) and Repetitive Behaviours (measures of repetitive traits within autistic and learning disabilities research) to compare disorders such as OCD and autism within a model based on assessment of repetitive traits.

Diagnostic Statistical Manual-5 (DSM-5) – The most recent version of a handbook, published by the American Psychiatric Association, used worldwide to guide the classification and diagnosis of mental disorders.

Ego-dystonic – A trait which is inconsistent with the individual's sense of self. It is therefore unwanted and unpleasant.

Ego-syntonic – A trait which is consistent with the individual's sense of self; one in which they derive pleasure from.

Free will – Potentially linked to various other phenomena (such as volition, desire and control), Free Will beliefs have two main definitions within psychological literature: agency (causing something to happen); and moral responsibility.

Neurotypical – A trait of an individual without a clinical disorder. Accordingly, a neurotypical person is an individual without a clinical disorder.

OCD – Now classified within an umbrella category of Obsessive-Compulsive and Related Disorders (DSM-5, APA, 2013), this disorder is widely known as Obsessive-Compulsive Disorder.

OCD traits – Also regarded as obsessive-compulsive traits, these are clinically defined as highly repetitive activities undertaken as an attempt to alleviate the anxiety caused by preceding obsessional content (i.e. intrusive thoughts).

Obsessive-compulsive traits – (see OCD traits)

Repetitive trait – Often referred to in psychological literature as Repetitive Behaviours, they can be more accurately be termed traits as they may consist of thoughts as well as actions.

Repetitive Behaviour – Specific repetitive traits which have originated from the study of individuals with autism and/or intellectual disability.

Higher-order Repetitive Behaviours – Cognitively complex types of Repetitive Behaviours, such as Insistence on Sameness.

Lower-order Repetitive Behaviours – Repetitive Behaviours which are based more on motor and sensory responses as opposed to complex cognitions.

Chapter 1. Background

1.1. Research Interest

As a practitioner, I regularly witnessed behaviours across children and adults with autism which appeared to be OCD traits but were always regarded as part of the phenomenology of autism by professionals. However, a missed OCD diagnosis would potentially deny an individual the opportunity to the most appropriate therapies. Therefore, it seems necessary to understand whether OCD traits are present in autism and, if so, what the similarities and differences are between obsessive-compulsive and repetitive traits in autism.

Initial searches of empirical literature revealed many links between the two disorders. However, on a symptom level (e.g. the similarities and differences between repetitive traits), there appeared to be a lack of research. Repetitive traits in both OCD and autism appear to have evolved separately, in isolation. Ego-dystonic and ego-syntonic origins of these traits, however, appeared to be fundamental. OCD traits are ego-dystonic (unwanted), whilst repetitive traits commonly researched in autism and learning disabilities (referred to as Repetitive Behaviours) appear to be ego-syntonic (i.e. part of the individual's sense of self and, therefore, wanted). Initially, the interest for the research was to compare repetitive traits in OCD and autism within a single framework; a Compulsive and Repetitive Trait framework. This framework was intended to compare the OCD traits and Repetitive Behaviours across both disorders, whilst making item-level mood assessments to attempt to understand the ego-syntonic and ego-dystonic origins of these traits.

Additionally, my interest has always been in philosophy and, particularly, in issues relating to free will. It seemed striking to me this major tenet of philosophical enquiry has received little attention in psychological investigation. It was my aim to contribute to the synthesis between philosophy (specifically free will) and clinical psychology (specifically traits in autism). Despite a lack of empirical research identifying the effect of self-reported free will beliefs in autism, it was proposed these beliefs may improve the understanding of clinical traits, specifically Compulsive and Repetitive Traits for the purposes of the present thesis.

1.2. Rationale: Comorbidity Between Obsessive-Compulsive Disorder and Autism

There is much uncertainty about the nature of repetitive traits across clinical groups. Even the relationship between traits within Obsessive-Compulsive and Related Disorders (OCD, American Psychiatric Association, 2013) seems unclear (Abramowitz & Jacoby, 2014). Whilst a range of evidence suggests links between OCD and autism (e.g. Anholt et al., 2010; Bejerot, Nylander, & Lindström, 2001; Delorme et al., 2007; Deramus, 2009; Hollander, King, Delaney, Smith, & Silverman, 2003; Ivarsson & Melin, 2008; Lehnhardt et al., 2013; Russell, Mataix-Cols, Anson, & Murphy, 2005), the precise nature of the relationship between these disorders remains elusive. As repetitive traits are central to both disorders (DSM-5: APA, 2013), comparing these traits between autism and OCD may help to clarify the relationship between the two disorders generally.

Part of the lack of clarity in the relationship between OCD and autism may have origins in theoretical assumptions relating to self-awareness. A certain level of introspection is

required in ego-dystonic traits (e.g. OCD); the individual must be somewhat aware the traits are inconsistent with their sense of self. However, self-awareness can be limited in autism (Williams, 2010). Deficits in social communication and social interaction (DSM-5, APA, 2013), a precursor to self-knowledge (Taylor, Peplau, & Sears, 2003, p.101), appear to lead to a lack of social understanding in the autistic mind (Marris, 1999). This factor may complicate the comparison between OCD and autism, but appears to require further investigation: if self-awareness is central to differences between autism and OCD, it is also possible free will may play a role in the production of repetitive traits. There is a lack of empirical research identifying the role of self-reported free will beliefs in both autism and OCD. An understanding of the relationship between these beliefs and Compulsive and Repetitive Traits may further improve the overlap in symptomology the two disorders.

This thesis aims to address these issues by comparing symptoms in autism and OCD within a single model, using evidence taken from the fields of repetitive traits in both OCD and autism research. A framework against which OCD and autism can be compared, will them allow additional phenomenological comparisons (e.g. beliefs in free will) to be applied. Defining the subtle differences between OCD and autistic traits in individuals with comorbid OCD and autism would allow for more accurate therapeutic support, aimed at traits with potentially distinct origins. Bentall (2004) argues this “cluster of symptoms” approach, which attempts to understand an individual in relation to a holistic view across a comprehensive set of traits (rather than as categorical labels of disorders), to be a much more accurate way of clinically defining an individual and, consequently, more accurately predicting therapeutic targets.

1.3. Research Aim and Questions

The main aim of the study was to investigate the relationship between repetitive traits, both within and between autism spectrum disorders and Obsessive-Compulsive Disorder (OCD). The secondary aim was to investigate whether free will beliefs are related to compulsive and repetitive traits. This investigation was set out to be achieved through a series of chapters.

Chapter 2 provides a background to the justification for comparing repetitive traits between OCD and autism. This was achieved through outlining theories of repetitive traits in OCD (including issues related to classification of the disorder as a potential dimensional construct) and evidence of the nature of repetitive traits in autism. Whilst arguments are presented in this chapter related to the problems of the concept of comorbidity, a wide range of evidence is presented related to comorbidity between the two disorders. This evidence suggests validity in comparing traits between OCD and autism.

Chapter 3 is a systematic review evidencing comorbidity between OCD and in adults with autism, to identify more specific evidence related to the sample group in the overall study. As there was sufficient evidence indicating an overlap between the two disorders, Chapter 4 looks in detail at evidence of the function and type of repetitive behaviours in autism. Research in this field indicates a measure of repetitive trait in autism literature, referred to as Repetitive Behaviours. The evidence base indicates a variety of different types and functions of Repetitive Behaviours. Frequently categorised as lower- and higher-level repetitive traits, they span from relatively simple motor-sensory behaviours (lower-level) to more cognitively complex – and

potential autism-specific – higher-level traits, such as Insistence on Sameness. Chapter 4 also synthesised existing literature on neuroanatomical and neuropsychological evidence related to repetitive traits in autism.

In Chapter 4, the evidence presented revealed a major theme of ego-syntonic and ego-dystonic origins of compulsive and repetitive traits across both autism and OCD. This indicated the potential importance of mood correlates, with ego-dystonic traits related to unwantedness (and therefore negative moods) and positive (or neutral) mood being related to ego-syntonic traits, by its opposing theoretical nature. This differentiation appears to provide a framework in which to distinguish between potentially different (or similar) repetitive traits both within, and between, OCD and autism.

As mood appeared to be implicated in compulsive and repetitive traits, the main systematic literature review (presented in Chapter 5) was performed to investigate the evidence of mood and compulsive and repetitive traits in adults with autism. Evidence for both ego-dystonic and ego-syntonic repetitive traits in adults with autism indicated the validity in a framework, hereon in referred to as Compulsive and Repetitive Traits (ego-dystonic and ego-syntonic traits, respectively). This forms a theoretical model, which enables an empirical test of the relationship between ego-syntonic and ego-dystonic repetitive traits in and between autism and OCD.

Chapter 6 presents a systematic literature review of psychometric properties of measures of OCD and repetitive behaviours, which informed the selection of the most appropriate assessment tools for use in the empirical investigation. These tools consisted of all items from the Yale-Brown Obsessive Compulsive Scale-Self Report

(Baer, 1992) and the Repetitive Behaviour Questionnaire (Moss, Oliver, Arron, Burbidge & Berg, 2009). Chapter 7 is the empirical investigation, consisting of design, results and discussion, comparing self-reported compulsive and repetitive traits in samples of adult participants with OCD, autism and neurotypical controls.

Whilst there is a lack of direct evidence of the relationship between compulsive and repetitive traits within and between autism and OCD, the evidence presented throughout these series of chapters indicates the validity in comparing compulsive and repetitive traits both within and between adults with autism and adults with OCD. Firstly, there is strong evidence of comorbidity between OCD and autism (Chapter 3). Furthermore, there is evidence, in autism, of potentially both ego-dystonic and ego-syntonic repetitive traits (Chapters 4 and 5). This evidence has been presented within one overall model, comparing the measures of OCD traits and autism-related Repetitive Behaviours, to form a Compulsive and Repetitive Trait (CaRT) framework.

The research questions related to this study were:

1. Is there a relationship between OCD traits (existing measures of OCD symptoms) and Repetitive Behaviours (existing measures of autism-related repetitive traits), clinically (i.e. within and between OCD and autism samples)?
2. Are Repetitive Behaviours associated with positive mood, specifically in autism?
3. Are OCD traits associated with negative mood, specifically in OCD?

Finally, Chapter 8 describes a correlational design investigation to test the secondary overall aim, comparing self-reported free will beliefs against this CaRT framework. The research question relating to this study was:

> How are free will beliefs associated with Compulsive and Repetitive Traits?

The first three questions test the validity of the conceptual framework; the Compulsive and Repetitive Trait framework. The final question is a novel investigation (free will beliefs) to offer a new perspective on this framework.

Chapter 2. Nature and Function of Compulsive and Repetitive Traits in Obsessive Compulsive Disorder and Autism.

Background

To test the validity in comparing Obsessive-Compulsive Disorder (OCD) and autism within a phenomenological framework, the nature of OCD symptoms is first presented, including consideration of whether OCD is a dimensional framework. Following this is evidence of the nature of repetitive behaviours from the field of autism research including evidence from neuroscience and cognitive literature. This presents the background to the systematic literature review of the evidence of overlapping symptomology between autism and OCD, considering issues relating to comorbidity generally.

2.1. Nature of Obsessive Compulsive Disorder symptoms

Obsessions are recognised as repetitive and persistent thoughts, impulses, or urges that cause anxiety; compulsions are repetitive behaviours or thoughts undertaken as an attempt to alleviate the anxiety caused by an obsession (APA, 2013). The result of the repetitive compulsion becomes life-consuming; rituals often take hours each day, usually with the individual fearing “dire consequences if the action is not performed” (Davison, Neale, & Kring, 2004, p. 157). OCD is defined by its ego-dystonic nature, meaning that the thoughts are not consistent with their concept of self and are, therefore, recognised by the individual as being unwanted, intrusive and distressing.

OCD is a heterogeneous disorder, with a wide range of symptoms (obsessions and compulsions) varying from one person to the next. Whilst clinical OCD measurement

tools list anywhere between 39 and 62 distinct traits (e.g. Kyrios, Bhar, & Wade, 1992; Steketee & Freund, 1993), researchers tend to classify distinct subtypes of OCD. Typical subtypes consist of washing, checking, symmetry and sexual content (Auoizerate et al., 2004; Mataix-Cols, Conceicao do Rosario-Campos, & Leckman, 2005). The current classification of OCD as a spectrum disorder reflects the complexity of interpreting OCD as a unitary disorder. Distinct, but related conditions, such as body dysmorphia (excessive worry about flaws in personal appearance), trichotillomania (hair-pulling), hoarding (excessive collecting and inability to discard) and excoriation (skin-picking) disorder are deemed sufficiently comparable to fall alongside OCD within an umbrella diagnostic classification (Bartz & Hollander, 2006).

Obsessive-compulsive disorder (OCD) is a debilitating condition, marked in all sufferers by distress (caused primarily by the unwanted nature of the symptoms) and inflexibility to time-consuming rituals. The restricted and repetitive nature of OCD symptoms is clear: in extreme cases the individual can literally be trapped by their compulsions, surrounded by the collections of their hoarding obsession and unable to leave their ritual for hours on end. Typically, OCD directly impacts on an individual's quality of life, negatively affecting employment, social, emotional and family outcomes (Coluccia et al., 2016). OCD can also indirectly limit an individual's life opportunities, with avoidance behaviours now recognised as an important measure of affliction (e.g. Storch et al., 2010).

OCD is now recognised within a range of heterogeneous disorders under the umbrella category "obsessive-compulsive and related disorders" (DSM-5, APA, 2013). Symptoms are required to be accompanied by a significant degree of dysfunction or distress, as

subclinical symptoms are part of typical development (Stein, 2002). Whereas it was once believed to be a rare condition, OCD is now established as one of the most common psychiatric conditions, with a prevalence around 0.6% to 3.1% of the general population (Stein, Forde, Anderson, & Walker, 1997) and a lifetime prevalence of around 3.5%, generally affecting women slightly more than men (Angst et al., 2004). OCD is considered to have a bimodal onset and most individuals develop the disorder in late adolescence or early adulthood (Davison et al., 2004). The median age of onset appears to be around 18 years, with prevalence rising exponentially with increasing age (Angst et al., 2004). Around 0.25% of children develop OCD (Heyman et al., 2001), typically around the age of 11 years (Taylor, 2011). Compared to late-onset OCD, early-onset OCD has been reported to be more common in males, demonstrate higher global severity and higher frequency of symptoms, more likely to be associated with other obsessive-compulsive disorders (particularly tic disorder) and appears to be more heritable (Taylor, 2011).

2.2. Theories of Obsessive Compulsive Disorder

There is strong evidence OCD is heritable, not just through genetic evidence, but also the stronger methodology of twin-studies (Leckman et al., 2010; Samuel & Nestaldt, 1997). In terms of the origins of OCD in individuals, the role of stress has always been implicated in OCD. The long-held suggestion (in early classical conditioning theory) of traumatic experiences triggering the disorder has been replaced by the idea exposure to stress (such as a stressful life event) is responsible for the onset of OCD (de Silva & Marks, 1999).

Abramowitz and Houts (2002) argue compulsive acts lead to a reduction in fear and uncertainty. The relationship between obsessions and compulsions appears to be little more than basic behavioural processes at work: compulsions are negatively reinforced in the presence of an obsessional fear. They appear to be unrelated to general intellectual or executive functioning. Whilst intrusive thoughts are ubiquitous (reported to occur in 90% of the general population), the researchers argue the propensity to misunderstand these thoughts as threatening (or related to a negative event) appears to be the feature specific to OCD.

2.2.1. Psychodynamic theories.

Early psychodynamic theory indicated emphasised the role of "hypermorality" in OCD (see Moritz, Kempke, Luyten, Randjbar & Jelink, 2011). OCD was said to result from holding back latent aggression to others, the suppression of which consequents in fantasies in the form of obsessional content. The individual is said to create a coping strategy (defence mechanism) by over-reacting to aggressive impulses due to feelings of over-responsibility i.e. hypermorality. Intrusive OCD content (e.g. sexual, aggressive or blasphemous) arises. These traits are egodystonic, as the individual recognises the immorality of the cognitive process.

General criticisms of psychodynamic theory hold: suppressed content (e.g. unconscious processes) are largely falsifiable and untestable, ergo unscientific and possibly meaningless. Whilst Moritz et al. (2011) demonstrate support (via medium to very strong effect sizes) for measures of latent aggression and distrust in OCD (though lack of support for measures of social desirability), they also report scientific evidence is largely equivocal for psychodynamic theories of OCD. Whilst cognitions arising from

this theory may be relevant, they would form part of a wider picture of the pathway model of OCD.

Modern psychodynamic theory appears to overlap more with cognitive-behavioural theories. As Kempke (2007) explains, such theories have progressed from conflict between hate and love, to the relationship between cognitive-affective schemas relating to the self and others. Whilst cognitive-behavioural models may largely explain how OCD is maintained, rather than how it originates, the field has produced a range of cognitions believed to be fundamental to OCD. Cognitive models largely suggest biases in thinking which cause OCD, as opposed to neuropsychological deficits.

2.2.2. Cognitive-behavioural theories

Cognitive models concerning perception of the self may not just emphasise the role of negative appraisal in reinforcing (cognitive-behavioural) OCD traits, but also the ego-dystonic (opposing the sense of self) elements, which are a key feature of the disorder. Aardema et al. (2017) indicated whilst fear-of-self perceptions appear not to be a specific feature of OCD in adults – negative perceptions were also consistent with non-OCD clinical participants in this study – unwanted repugnant thoughts and impulses do appear to be a significant feature in OCD.

A metacognitive model has been put forward to explain the product of intrusive thoughts, and the subsequent way they are actioned in OCD. Myers et al. (2017) tested the thought fusion metacognitive model, which concerns three processes: beliefs that thoughts alone can cause (or signify) an event to happen (Thought-Event Fusion); beliefs that intrusive thoughts can cause an individual to perform an unwanted act

(Thought-Action Fusion); and beliefs that thoughts/feelings can transfer into objects (Thought-Object Fusion). In a clinical investigation, Myers et al. (2017) reported how these three processes appear to be somewhat independent to each other in individuals with OCD. These thoughts are said to be appraised negatively by an individual and, following this, a second domain is activated, whereby an individual will attempt to regulate the distress caused, either by actions (rituals) and/or continued metacognitive plans. The strength of this model is in its ability to distinguish between OCD-related thoughts (as described above) and non-metacognitive beliefs in OCD: for example, Myers et al. (2017) argued beliefs such as perfectionism and responsibility are present in OCD as a by-product of metacognitive beliefs, rather than directly related to OCD. However, there still appears to be a need to identify the extent of predictive factors in the disorder, with Myers et al. (2017) emphasising the great deal of variance in measures of metacognitive belief in OCD samples.

There is evidence difficulty in making decisions generally may be a significant factor in OCD. Using a simple task taken from behavioural economic theory in adults with OCD, Pushkarskaya et al. (2017) demonstrated difficulty in planning may differ between individuals with different OCD traits. For example, Pushkarskaya et al. (2017) reported increased inconsistent choices in participants with OCD, but not Hoarding Disorder, under laboratory conditions. The researchers claim subjectivity (i.e. subjective goal values) is the critical factor in difficulties of such value-based decision-making in OCD. Such subtle understanding of decision-making processes appears to be important to lead to greater knowledge of other related constructs deemed important within OCD, such as indecisiveness, pathological doubt, increased deliberation and general avoidance of decisions.

2.2.3. Cognitive biases.

A range of cognitive biases have been suggested throughout cognitive-behavioural research. Some of the more prominent biases are discussed below, with a view to explaining their impact on the knowledge of OCD generally.

The cognitive theory of OCD, which argues individuals with OCD misinterpret the significance of normal intrusions, has led to the obsessive beliefs questionnaire-44, which implicate the importance of six cognitive domains in the onset of OCD: overestimation of threat; excessive responsibility; perfectionism; need for certainty; and both the importance, as well as the control of thoughts. However, Tibi et al. (2018) recently reported mixed evidence still of the significance of the relationship between cognitions and OCD symptoms, with this theory failing to be supported in longitudinal analysis (over a two-year span).

Overactive performance monitoring is claimed to be a key feature in the production of OCD, whereby individuals with the disorder are prone to experience excessive concern (which they feel the need to alleviate), following identifying inadequacy and error in everyday activities. This theory has been supported neurologically, where participants with OCD have been reported to demonstrate larger brain amplitudes in response to emotional faces (compared to controls), indicative of the role of affective functions within the fronto-striatal network in OCD (Roh, Chang & Kim, 2016). Overactive performance monitoring has been supported in physiological assessments: Yoris et al. (2017) reported how, whilst OCD participants outperformed matched non-OCD clinical (panic disorder) and healthy control groups in self-monitoring their own heartbeats, they also demonstrated poorer performance and awareness of these detections. Thus,

this seemingly OCD-specific feature of over-monitoring is marked by a lack of awareness/confidence in their biological activities. There is consistent evidence for the relationship between doubts about actions and OCD severity (e.g. Martinelli, Chasson, Wetterneck, Hart & Björgvinsson, 2014).

Perfectionism, whereby an individual sets unrealistically high expectations and self-evaluations, has been reported to be highly prevalent in OCD (Pinto et al., 2017). Perfectionism has been reported to be a major factor in “not just right experiences”, believed to underlie OCD traits such as symmetry, counting and repeating, with an absence of “perfect certainty” associated with doubt in OCD (Pinto et al., 2017).

Whilst guilt has been long indicated in OCD literature to be a significant feature of the disorder, Chiang, Purdon and Radomsky (2016) reported fear of guilt may be a significant, and specific, feature of OCD. Whilst the Fear of Guilt Scale is awaiting assessment in a clinical OCD group, psychometric properties of the scale in relation to OCD symptoms in 2407 non-clinical participants point to the promising nature of the measure. In addition to significantly predicting OCD symptom severity, strong internal consistency (α = 0.92-0.94), strong convergent validity (r = 0.39-0.62 for various measures of OCD) and strong divergent validity (e.g. r = 0.35-0.49 for measures of depression) suggest fear of guilt may be an important cognitive construct in OCD.

Melli, Aardema and Moulding (2016) reported fear-of-self to be the only unique predictor of obsessionality in adults with OCD with OCD, compared with measures of anxiety and depression. Furthermore, measures of fear of self uniquely and majorly predicted unacceptable thoughts. Similarly, guilt sensitivity has also reported to be a

significant predictor of checking-related OCD traits, independent of depression, disgust, anxiety and stress (Melli, Carraresi, Poli, Marazzati & Pinto, 2016).

The problem with many of the cognitive-behavioural research is its correlational nature. Whilst much of the evidence is consistent with OCD theory, the studies reported above cannot definitively claim to show causes of OCD symptoms. These cognitive beliefs are likely to be only part of the full neuropsychological pathway contributing to OCD, with other moderating and mediating variables contributing to the disorder. As OCD is consistently reported as a heterogeneous disorder, it is possible the range of cognitive biases may be further complicated when separating out the effect of dimensions or subtypes within OCD.

2.3. Obsessive Compulsive Disorder as Dimensional Disorder

OCD is no longer classified as an anxiety disorder for the DSM-5 (APA, 2013), although there are still some proponents who argue the importance of considering OCD as a disorder of anxiety (e.g. Starcevic & Janca, 2011). There is still support for anxiety disorder to be grouped with OCD, based on links via increased co-morbidity and familial data (Bienenu et al., 2012) and clinical utility (Stein et al., 2010). However, OCD is reported to differ from other anxiety disorders with earlier age of onset, complex comorbidity, and severity of obsessions and compulsions (Murphy, Timpano, Wheaton, Greenberg & Miguel, 2010). Instead, it is now recognised as an umbrella of disorders – Obsessive Compulsive and Related Disorders (OCRD) – with five disorders, diagnostically linked by “repetitive thoughts and behaviours and phenomenological and neurobiological similarity to OCD” (Krzanowska & Kuleta, 2017). Other disorders within OCRD in the DSM-5, as well as OCD, are body dysmorphic disorder, hoarding

disorder, trichotillomania, and skin picking disorder. Only disorders were included within the OCRD category if they are marked by distress caused by ego-dystonic features (i.e. unwanted, opposing to the individual's sense of self). A major criticism about this OCRD category is the way the disorders were seemingly arbitrarily put together, rather than supported by clear evidence, a problem which still appears to exist (Krzanowska & Kuleta, 2017).

This change for the DSM-5 did not take full heed of the recommendations by Hollander et al. (2009), a planning committee formed to suggest future directions for the conceptualisation of OCD. This research committee indicated the potential validity in an even wider range of disorders, which can fit within an OCD spectrum, referencing various links (phenomenology, comorbidity, course of illness, brain circuitry, familial/genetic factors, and treatment response) between impulse control disorders, autism, Tourette's syndrome, trichotillomania and Parkinson's disease. Autism, for example, appears to be linked to OCD not just in their symptomological similarities (repetitive traits in particular), but also in comorbidity, genetic heritability rates of traits across both disorders, and potentially dysfunction across similar neuroanatomical regions. Despite the strong emphasis on the utility of understanding such "related" disorders within a unified spectrum, Hollander et al. (2009) themselves indicate the lack of clarity on how to group disorders on scientifically sound principles.

Among various recommendations for diagnostic improvements before the DSM-5 was created, Leckman et al. (2010) strongly emphasised the difficulties in distinguishing between OCD content and similar, but distinct, traits which are part of different disorders. Examples given include obsessive ruminations in hypochondriasis, obsessive

preoccupations in anorexia nervosa, or ruminative thoughts in depression. However, the researchers also stress the necessity for a hierarchical approach to understanding OCD and nosologically relevant disorders.

A systematic review and cluster analysis by Lochner and Stein (2006) concluded a spectrum of obsessive-compulsive disorders may lie on three clusters: reward deficiency, including disorders such as pathological gambling, trichotillomania and Tourette's; impulsivity, including disorders such as eating disorder, compulsive shopping and kleptomania; and somatic disorders, including body hypochondriasis and dysmorphic disorder. The researchers argue the compulsive-impulsive framework has been largely demonstrated to be an overly simplistic heuristic, and OCD is better explained in a more relational way to other disorders. Whilst these groupings have some degree of basis across neuroimmunology, neuroanatomy, neurochemistry and hereditability, Lochner and Stein (2006) reported further evidence is needed to identify endophenotypes within these clusters and disorders – whereby disorders with tics, for example, may be recognised differently though specific neurobiological markers.

However, proponents against a spectrum framework argue arbitrary decisions appear to be generally made to include "related" disorders within an OCD spectrum (e.g. trichotillomania, kleptomania, pathological gambling/shopping, binge eating, Tourette's syndrome, hypochondriasis and body dysmorphic disorder - BDD), based on assumptions of superficial similarities (shared repetitive thoughts and behaviour patterns). Abramowitz and Hourt (2002) reviewed evidence of links between OCD and different related disorders and report evidence of links with both hypochondriasis and

BDD appear sufficiently strong to be potentially classified within an OCD spectrum. The authors do not argue against the concept of a spectrum per se, but warn caution against claiming a shared continuum on merely incidental behavioural correlation alone.

2.3.1. Dimensions by symptom subtypes.

Many investigations have attempted to address the heterogeneity of OCD, with symptom dimensions acting as distinct phenotypes of OCD. Support for this concept is the associations found between certain OCD symptom dimensions and three obsessive beliefs (responsibility and threat estimation; perfectionism and intolerance of uncertainty; and importance and control of thoughts). Brakoulias et al. (2014) identified specific positive correlations between: importance and control with unacceptable thoughts/taboos; responsibility/threat with doubt/checking; and perfectionism/intolerance of uncertainty with symmetry/ordering OCD dimensions (see also Martinelli et al., 2014). Whilst no associations were found for contamination/cleaning, the same finding was also described for the hoarding dimension, adding further weight to the idea hoarding may be somewhat divergent within the “OCD spectrum”.

Factor analytical investigations have indicated the potential validity in distinguishing between OCD patients whose traits form subtypes (e.g. contamination worries or obsessions of symmetry and ordering). Factor analysis has typically revealed the strongest evidence for the validity of the following subtypes of OCD, based on symptoms: symmetry/ordering; contamination/cleaning; unacceptable thoughts; doubt/checking; and hoarding. Leckman et al. (2010) suggest other statistical analyses

are more valid in understanding the apparent heterogeneity in OCD, with latent class modelling indicating the greater relevance in defining OCD sufferers by the severity of their symptoms. However, Leckman et al. (2010) argue the clinical utility of OCD is perhaps increased by understanding OCD as a dimensional construct.

The relevance of understanding OCD as dimensional constructs is supported by evidence these constructs appear to be underpinned by specific patterns of neuropsychological functioning. In a meta-analysis of ten systematically studies (*N* = 628), Bragdon, Gibb and Coles (2018) reported small negative correlations for: the symmetry/ordering domain and overall neuropsychological functioning, executive functioning performance, memory, cognitive flexibility, visuospatial ability and verbal working memory; obsessing/checking traits and memory and verbal memory. Furthermore, a large effect size was reported in the symmetry/ordering domain and attention, verbal working memory and visuospatial ability. Pedron, Ferrao, Gurgel and Reppold (2015) have also reported further unique neuropsychological patterns associated with OCD symptom dimensions. The researchers describe positive correlations between hoarding dimension severity and cognitive flexibility, visual processing and logical reasoning, in addition to negative correlations with problem-solving capacity. Pedron et al. (2015) also found contamination/cleaning severity to be positively associated with executive functioning, inhibitory control and attentional control.

Obsessional dimensions have also been suggested. Demonstrating good psychometric properties, Garćia-Soriano, Belloch, Morillo and Clark (2011) identified six first-order factors, within two second-order factors, from the self-reported Obsessional Intrusive

Thoughts Inventory (OITI). One second-order factor was reported to contain the aggressive, sexual, religious, immoral and religious obsessive intrusive thoughts, whilst the other factor consisted of contamination, doubts/checking, symmetry/order and superstitions. It is notable however, the OITI contains some "miscellaneous" obsessions, (such as superstitions), which are frequently omitted in Y-BOCS-SC studies, whilst the OITI itself omits hoarding traits (Garćia-Soriano et al., 2011). These independent obsessional symptom factors may provide further clarity into what has traditionally been seen as a heterogeneous construct.

Distinct neuropsychological functioning within symptom dimensions has been revealed by Kashyap, Kumar, Kandavel and Reddy (2017). After controlling for depression and OCD severity, medication and other symptom dimensions the following neuropsychological patterns were found for the five identified factors: contamination/washing related to poorer attention/working memory, visuo-spatial construction and increased planning abilities; doubts/checking associated with poorer alternation learning; symmetry/ordering related to poorer verbal fluency; forbidden thoughts with better visuospatial scanning and working memory; and hoarding factor with worse verbal recall and increase performance of visuospatial working memory.

Such evidence would indicate the possibility of these domains being served by different neural circuitry, which would be a strong indicator of the validity in a dimensional framework in OCD. There is evidence different OCD symptom dimensions are mediated by separate neural networks, as increased activity in emotion processing areas in individuals with washing- and hoarding-related OCD to related content (Phillips & Mataix-Cols, 2004). Additionally, increased activation of the amygdala

(involved in processing of fear and anxiety), has been reported for individuals with aggression/checking and sexual/religious OCD traits, whilst those with other OCD traits appear not to demonstrate increased activation in this area (Via et al., 2014). Furthermore, Lázaro et al. (2014) have reported evidence indicating neuroanatomical differences between OCD subtypes, including: harm/checking dimension demonstrating reduced fractional anisotrophy (FA) in the corpus callosum, left anterior cingulate gyrus and caudate nucleus; and contamination/washing trait dimension decreased FA in left midbrain, lentiform nucleus, insula and thalamus, plus increased diffusivity measures in the anterior lobes of the cerebellum and the pons. Whilst exploration of neural networking still requires much further study, such evidence provides compelling indication of the necessity to consider OCD as a dimensional, rather than a categorical construct.

Murphy, Timpano, Wheaton, Greenberg and Miguel (2010) found four symptom dimensions appear to be the most commonly identified solution: contamination obsessions/cleaning compulsions; aggressive, sexual, religious and somatic obsessions, with cleaning compulsions; symmetry/exactness and “just right” obsessions with ordering/arranging/counting compulsions; and hoarding obsessions and compulsions. There is evidence these subtypes are stable temporally and cross-culturally, and have biological/neurological and comorbidity patterns (Prabhu et al., 2013). They also note the strong evidence demonstrating the differences between early- and late-onset OCD (e.g. in terms of gender and comorbidity differences).

2.3.2. Early- versus late-onset subtypes.

Noting the lack of meaningful and replicable findings from the study of a genetic basis of OCD, Prabhu et al. (2013) attempted to address the understanding of the heterogeneity of OCD through the relationship between clinical characteristics and OCD symptom dimensions. It was reported sexual/religious, aggression and symmetry subgroups were marked by earlier age of onset. Fear of contamination was higher in females, with higher family loading, and poorer insight and functioning.

In a systematic review and meta-analysis consisting of 4650 participants with OCD (2162 for early-onset and 4650 for late-onset) across 27 studies, Taylor (2011) claims strong evidence for early- and late-onset as two distinct subtypes of OCD. Accounting for around 78% of all OCD, early-onset OCD was described to differ from late-onset OCD with regards to: higher proportion of males; higher severity of OCD and increased prevalence of most OCD symptoms; higher comorbidity with tics and other "OCD-spectrum" disorders; and larger rates of prevalence in families (first-degree relatives).

However, other investigations have reported identical factor structures of OCD traits in early- and late-onset OCD (Grover et al., 2018), with comparable responses to treatment and, furthermore, rates of 80% early-onset OCD reported in adult OCD samples (see review by Grados, Labuda, Riddle & Walkup, 1997).

2.3.3. Dimensions by Subtypes of Cognitive Biases

Subtypes of OCD appear to be clearly distinguished by specific cognitive appraisals. However, what has been traditionally been assumed to be homogenous dimensions, now have been indicated to be multi-faceted. For example, OCD contamination fear

has been indicated to be distinguished by two different motivational dimensions: harm avoidance and disgust avoidance (Melli, Chiorri, Carraresi, Stopani & Bulli, 2015). Whilst these two factors may co-exist, being potentially strongly correlated (r = 0.62), Melli et al. (2015) report these two subscales as demonstrating good convergent and discriminant validity, showing different patterns of correlation with other OCD traits.

The cognitive trait of perfectionism has been reported to be associated to specific subtypes of OCD, including ordering, checking, cleaning and hoarding (Pinto et al., 2017). Shame has been reported to be associated with specific OCD dimensions; Wetterneck, Singh and Hart (2014) identified a positive correlation between shame and harm, but not unacceptable thoughts. Additionally, a significant relationship was reported by the researchers between shame and symmetry, argued to be related to previous findings of associations between symmetry and perfectionism.

2.3.4. Evidence from comorbidity and subtypes in Obsessive Compulsive Disorder.

Further support for the validity of dimensional constructs within OCD has been demonstrated by variations in comorbidities, which have been described for different OCD symptom dimensions. In a large (N = 1000) cross-sectional analysis of OCD patients, Torres et al. (2016) assessed lifetime axis I comorbid disorders using the Structured Clinical Interview for DSM-IV Axis I Disorders. No comorbidities were identified for between axis I disorders and the symmetry-ordering dimension. However, the following independent associations were identified: aggressive dimension with posttraumatic stress disorder (PTSD), separation anxiety, impulse-control disorder and skin-picking; sexual/religious dimension with mood disorder,

panic disorder/agoraphobia, social phobia, separation anxiety, non-paraphilic sexual disorder, somatoform disorder, body dysmorphic disorder and tic disorder; contamination/cleaning dimension with hypochondriasis; and hoarding dimension with depressive disorders, specific phobia, PTSD, impulse-control disorders, ADHD and tic disorders.

2.3.5. Problems and limitations with dimensional classification of OCD

Whilst clinically useful (e.g. by informing therapeutic targets), there are a number of criticisms over such classifications of OCD, starting with fundamental principles aimed at any artificial (and often arbitrary) way of constructing cognitive disorder. Many of these studies are correlational in design, therefore any causal links cannot be firmly established. Also, many of the studies lack the accuracy and sensitivity to assess family-links, with the hereditability potentially being understated. Significantly, it has been demonstrated a sizeable proportion of OCD patients do not necessarily fit into reported subtypes, perhaps around a quarter (Matsunga, Hayashida, Kiriike, Maebayashi & Stein, 2010).

Murphy et al. (2010) argue the study of subgroup has typically be flawed. Whilst hoarding-related OCD has been repeatedly indicated to be a specific subgroup, other attempts to define subgroups by anxiety, depression and suchlike appears to be generally hampered by studies lacking adequate sample sizes. Clearly, understanding any possible reliable subtypes and spectrum conditions is a huge task, requiring more clarity over shared underlying mechanisms, genomic investigations and aetiologies.

Significant problems with the dimensional constructs generally put forward in OCD research has been highlighted by Hasanpour et al. (2017), using a more thorough set of clustering algorithms. These results revealed the most appropriate best fits, statistically, were not by type of symptom, but on severity of symptoms (similar to Leckman et al., 2010). The more severe patients were reported to have an earlier age of onset, more females, lower education levels, higher depression, higher avoidance and lower insight. Whilst it is plausible symptom severity may be a significant factor of distinguishing between subgroups, the clinical utility (e.g. type of intervention), may not be as high as symptom-based dimensions.

2.3.6. Summary

Psychodynamic theory appears to have clear flaws, through partly untestable claims. However, modern forms of the theory have provided a framework which has supported cognitive-behavioural models. These models have produced a range of cognitive biases, which have generated a body of evidence to indicate the significance – and specificity – to the nature of OCD. However, cognitive-behaviour models are largely correlational, and there is little substantive evidence to explain the causes of the disorder. Further work is needed to understand the OCD pathway (psychological and neural). Clarity needs to be given to the validity of OCD as a unitary disorder, with explicit links to related disorders, or a dimensional disorder with clearly defined and valid subtypes.

Whist there is a wealth of evidence described above indicating the validity of understanding the heterogeneity, there is still argument of a lack of consensus for the clinical utility and theoretical relevance of grouping various “OCD-related” disorders as

an OCD spectrum (e.g. body dysmorphic disorder, hoarding disorder, trichotillomania, Tourette's disorder), perhaps on the over-reliance on comparing repetitive behaviours (Stacevic & Janca, 2011).

There is much stronger evidence for understanding OCD as a dimensional symptom construct, with dimensions seeming to display specific neuropsychological patterns, and seemingly subserved by distinct neural circuitry. However, understanding as categorical symptom approaches may be flawed as individuals generally present with overlapping symptomologies; symptom dimension approaches may be flawed due to the lack of consensus over measuring, scoring and analysing, leading to varying structures (Pinto et al., 2017). The mediating/moderating effect of all the factors (e.g. age of onset, subtype, severity of OCD, comorbid disorders) is still unclear and much more investigation is clearly needed to understanding the actual phenomenology of OCD.

For now, the evidence seems to indicate OCD is best understood as a dimensional construct or spectrum. However, whilst there appears to be little doubt over the heterogeneity of OCD, greater understanding may be identified by investigating issues related to comorbidity between OCD and other disorders. As autism appears to be significantly prevalent as a comorbid disorder with OCD (Hutton et al., 2008; Lehnhardt, 2013; Stein et al., 1997), it may be necessary to make greater comparisons between the two disorders.

Empirical research undertaken over the last two decades has provided a good understanding of OCD as a standalone disorder. However, current research is

attempting to understand the nature of OCD when it is present in comorbid clinical populations (Hollander, Kim, Braun, Simeon, & Zohar, 2009; Russell et al., 2005; Scahill et al., 2006; Wahl, 2009), such as autism. Recent indications of OCD as a spectrum disorder offers a new challenge, particularly when attempting to understand the phenomenological link between subtypes, which may in fact "fail to meet standard guidelines for developing subtypes" (Rowsell & Francis, 2015). It is likely a greater understanding of comorbidity issues will lead to a better understanding of the disorder. As autism appears to be significantly prevalent as a comorbid disorder with OCD (Hutton et al., 2008; Lehnhardt, 2013; Stein et al., 1997), it may be necessary to make greater comparisons between the two disorders.

2.4. Autism Spectrum Disorder

Autism is a neurological developmental disorder marked by social and communication difficulties, alongside the presence of Repetitive Behaviours (DSM-5: APA, 2013). Predominantly, the severity of symptoms affects an individual's ability to comprehend and function in the social world. It is a lifelong disorder which affects the life opportunities of individuals in many ways. Whilst those with milder symptoms regularly go on to lead normal lives, individuals with more exaggerated traits may typically become the highest of achievers, or forgo a life of high dependency on social support (Cooper, 2012). Intelligence is a major protective factor mediating between these two life outcomes, particularly when obsessional temperaments (Soderstrom & Gillberg, 2002) can be channelled into a positive career-led focus (Baron-Cohen, 2008; Wheelwright & Baron-Cohen, 1998). Regardless of the level of intervention, significant impairments in daily living and communication often remain throughout the life of an individual with autism (Magiati, Tay, & Howlin, 2014). Overall, quality of life appears to

be generally poorer for individuals with autism (Chiang & Wineman, 2014; Henninger and Taylor, 2012), with typical disadvantage in employment (Mavranezouli et al., 2014) and health (Howlin & Moss, 2012).

Evidence strongly suggests autism is a highly heritable disorder (e.g. Tick, Bolton, Happé, Rutter, & Rijsdijk et al., 2016), which occurs in around 1.16% of the general population (Baird et al., 2006) including cross-culturally (e.g. Wakabayashi, 2006; Ruta et al., 2012). Levels of comorbid intellectual disability in autism appears to be around 70% (La Malfa, Lassi, Bertelli, Salvini, & Placidi, 2004). Much like OCD, autism is a highly heterogeneous spectrum disorder. Across the autism spectrum, individuals with autism present with a hugely varied pattern of social and communicative limitations, and a "spiky profile" of cognitive abilities and intelligence (Happé, 1994, p. 1469). Fundamentally, it seems a processing bias for non-social information appears to lead to a variety of social difficulties (Jeste & Nelson, 2009), whilst clearly enabling some individuals to excel in areas such as mathematics, physics, engineering and computing (Baron-Cohen, 2008; Wheelwright & Baron-Cohen, 1998).

"Restricted/Repetitive Behaviours" appear to be so fundamental to the phenomenology of autism, their presence is one of only two symptom categories required for a diagnosis of an autism spectrum disorder, alongside social communication deficits (DMS-5: APA, 2013). These repetitive traits range from lower-level repetitive traits such as stereotyped motor movements and abnormal reactivity to sensory stimuli, to higher-level (more cognitively complex) traits such as insistence on sameness and fixated interests (Bodfish, 2007; Turner, 1999). Repetitive Behaviours are claimed to have distinct origins from repetitive traits in OCD. OCD traits are ego-

dystonic (inconsistent with the self), therefore unwanted and unpleasant. However, Repetitive Behaviours in autism have been suggested to be a fundamental part of the self, i.e. ego-syntonic (Baron-Cohen, 1989; Rice, 2009), to such an extent the DSM-5 states they may become a source of "pleasure or motivation", whether by education or employment. However, more recent research has indicated some of these repetitive traits may be more ego-dystonic than traditionally assumed (e.g. Barber, 2015; Cath Ran, Smit, van Balkom, & Comijs, 2008; Saddington, 2013), which may reconcile the links between autism and OCD.

There are still significant gaps in scientific understanding of autism, particularly in relation to how it affects females (Van Wijngaarden-Cremers et al., 2014) and knowledge of how the disorder presents in later life (Happé & Charlton, 2012). Anxiety is widely acknowledged to be a significant issue in autism, although White et al. (2014) contend the aetiology of anxiety in autism is still little understood. Regardless, to understand the validity of comparing autism and OCD symptom, the evidence of comorbidity between the two disorders is critical.

2.5. Evidence of Comorbidity between Obsessive-Compulsive Disorder and Autism

Whilst early research suggested obsessive-compulsive behaviour has a distinct pattern in OCD compared to autism (McDougle, 1995), later evidence from a range of fields indicates considerable overlapping symptomologies between the two disorders. Such evidence includes increased autistic traits in individuals with OCD (Bejerot et al., 2001; Ivarsson & Melin, 2008), similar patterns of obsessions and compulsions in individuals with OCD and with autism (Russell et al., 2005) and increased rates of OCD in parents of children with autism (Hollander et al., 2003). Similarly, other studies have reported

comparable severity of obsessive-compulsive symptoms in autism and OCD groups (Deramus, 2009; Russell et al., 2005), increased prevalence of autistic traits in OCD (Anholt et al., 2010; Bejerot et al., 2001), a high incidence of compulsive behaviour in autism (Simons, 1974) and similar profiles of shared executive dysfunction in unaffected relatives of both OCD and autistic individuals (Delorme et al., 2007).

There is evidence that OCD is significantly prevalent in adults with autism. Comorbid diagnoses of OCD in samples of individuals with autism have been reported anywhere between 3.7% (Hutton et al., 2008) and 50% (Lehnhardt, 2013), compared to the expected prevalence of between 0.6% to 3.1% in the general population (Stein et al., 1997). Barber (2015) reported 40% of OCD adult participants were found to display clinical levels of autism traits. However, Barber (2015) also warned against making a causal assumption based on similarities in clinical profiles: similar executive functioning between OCD and autism appeared to be moderated by variables such as depression and anxiety.

Such evidence has ignited interest in understanding the overlapping nature of the two disorders specifically (see review by Fischer-Terworth & Probst, 2009), and comorbidity issues in autism generally. The evidence is so compelling, a category of "autism-related obsessive-compulsive phenomena" has been suggested as part of the clinical symptomology of autism (Fischer-Terworth & Probst, 2009). It is intriguing why this category has not been retained in the current diagnostic criteria (DSM-5: APA, 2013) in view of this increasing evidence. Fischer-Terworth and Probst (2009) claimed little is understood regarding the similarities and differences between obsessive and compulsive symptoms in OCD and autism, which still appears to be true. Despite these

increasing links, it has been reported that OCD diagnoses can be missed, particularly in populations involving comorbid psychiatric (Wahl, 2009), neurological (Hollander et al., 2009) and developmental disorders (e.g. Russell et al., 2005).

The evidence presented in this chapter gives an indication of some overlap between autism and OCD generally. Whilst most psychological research in autism (including what has been reported in this chapter) relates to childhood samples, the overall body of work in this thesis is aimed at the adult age range. The next section provides a systematic review of evidence of repetitive trait symptomology overlap (comorbidity) in autistic adult samples.

Chapter 3. Systematic Review of Comorbidity between Obsessive Compulsive Disorder and Autism in Adults

Background

Understanding comorbidity is important, as one can only truly understand any disorder by delineating all the mechanisms which affect its pathogenesis. We must not look at comorbidity rates alone, as there are a number of factors which increase the likelihood of comorbidity, factors which, Rutter (1997) argues, artificially raise the comorbidity if we extrapolate from a given samples.

Referral factors may generally be a cause of such increased artefactual comorbidity (Rutter & Caron, 1991). Firstly, referral likelihood is increased by the combined likelihood of each disorder separately. Additionally, there may be inherent referral biases in any study, for example: individuals may be more likely to be referred if there are family problems; or participants referred to a researcher known to be investigating comorbidity.

As there are many factors involved in psychopathology (e.g. two disorders may share the same underlying risk factors, or different disorders may contain the same/similar diagnostic traits), Rutter and Caron (1991) suggest a dimensional approach – rather by distinct categories – may be a more valid interpretation. This recommendation is line with a shared symptomological framework of OCD and autism, whereby an understanding of the whole of the individual (i.e. “cluster of symptom approach”; Bentall, 2004), could be a more accurate way of recognising disorder.

As Rutter (1997) argues, comorbidity is only a statistical phenomenon, devoid of any particular meaning; its relevance instead, is what comorbidity indicates about the underlying mechanisms. Sufficient comorbidity, therefore, between OCD and autism may implicate the relevance of a different perspective on understanding the disorders, which again may point towards the relevance of a shared framework to understand OCD and autism comorbidity.

3.1. Method

3.1.1. Research aim and questions.

The aim of the systematic literature review was to investigate existing evidence of the comorbidity between OCD and autism.

The research questions related to this study were:

1. Are there overlaps in symptomology between autism and OCD, whereby repetitive traits are significantly related between the disorders?

3.1.2. Search strategy.

To include studies from a large pool across psychology research, articles were retrieved using EBSCO (PsycINFO, PsycARTICLES, MEDLINE and CINAHL Complete – four search engines which would produce a thorough yield of psychological research) for published articles related to comorbidity between OCD and autism. Limiters were placed to ensure mainly that studies of adult participants were identified. No studies were excluded based on date, to include as much evidence as possible.

3.1.3. Inclusion criteria.

Articles were included where the sample consisted of either: 1) comorbid autism and OCD participants; 2) OCD participants with an outcome related to autism symptoms; or 3) autistic participants with an outcome related to OCD symptoms.

3.1.4. Exclusion criteria.

Based on the research aims and inclusion criteria, studies were excluded if they fell into the following criteria:

- Participants were below the age of 18 years (as they are not within the adult age range of the final empirical investigation);
- Single case studies, or studies involving fewer than 10 participants, due to reduced reliability of such evidence when generalising to the wider population
- Indirect reference to the overlap/comorbidity (i.e. the study makes no meaningful contribution to understanding the issues of comorbidity or symptom overlap between the two disorders).
- Participants with intellectual disabilities/learning disabilities, due to other comorbidity issues.

3.1.5. Search terms

Search terms as described in Table 3.1 were used, with the final search being completed on 14th May 2019.

Table 3.1. *Search terms used for comorbidity systematic search using PsycINFO, PsycARTICLES, MEDLINE and CINAHL Complete.*

Any of the following terms for autism as "select a field"	Combined with "AND"	The following term for Repetitive Behaviour as "abstract"	Combined with "AND"	The following term for mood as "abstract"
OCD or obsessive compulsive disorder		autis* or Asperger* or "pervasive developmental disorder" or pdd		comorb* or overlap* or shared

* indicates the word was truncated to allow for multiple word endings
Nb. Special limiters placed for PsycARTICLES: all articles for participants aged 18 years and over; human; exclude book reviews; exclude non-article content. Special limiters placed for CINAHL Complete: human; English language; all adult. Special limiters placed for MEDLINE: human; English language; all adult. Special limiters placed on PsycINFO: all articles for participants aged 18 years and over; English language; human.

3.2. Selection strategy

For more thorough identification of evidence, the abstracts (rather than the titles) of all yielded articles identified were screened for relevance against the inclusion and exclusion criteria outlined in 3.1.3 and 3.1.4. Eight articles were included in the final systematic review, as outlined in the PRISMA (Moher, Liberati, Tetzalff, & Altman, 2010) study selection flow chart (see Figure 3.1).

3.3. Data analysis and synthesis

The approach to writing narrative syntheses as recommended by Popay et al. (2006) was followed in this review. This was the most appropriate approach to take due to the mix of methods, participants and different outcome measures reported in the heterogeneous articles yielded. First, textual descriptions via tabulation were created for each of the included studies, to facilitate a simple comparison of the key information from these studies (see Table 3.5). Groupings were also identified (see Table 3.4), to form the layout of the narrative synthesis. The methodological quality of the studies are illustrated in the appraisal Table 3.6, which indicates the "weight" (Popay et al., 2006, p.16) to be given to the evidence within each study. The critical appraisal questions used were those recommended by Aveyard (2010, p.103) and assess the robustness of the studies, used to guide the narrative synthesis, as suggested by Popay et al., (2006).

Finally, with regards to robustness, the approach taken in this systematic investigation has been laid out by Aveyard (2010) – see Table 3.6. This recommendation was followed as it lends itself more reliably to the evidence presented via the methodologies in the included articles. These appraisal methods, in addition to the summaries (Table 3.5), are in line with the recommendations of robustness by Popay et al. (2006): Weight of Evidence – EPPI Approach; Best Evidence Synthesis; and use of validity assessment. In general, precedence has been given to the more relevant and higher quality evidence through a larger narrative weighting.

Table 3.2. *Selection of tools and techniques in developing synthesis as recommended by Popay et al. (2006).*

Name of tool/technique	Comments in relation to current synthesis	Should, and has, this tool/technique be applied here?
Textual descriptions	With a relatively small number of studies included in this systematic review, it would be useful to extract the key information from each study in a systematic way. For this to be achieved, only the answers to the questions as set out in Table 3.6 should be included, with outcome information only provided if relevant specifically to the research question (i.e. comparison of symptoms or diagnoses between adults with autism and OCD).	Yes.
Groupings and clusters	This would enable ease of comparison for the reader, to improve facilitate ease of scrutiny. See Table 3.4.	Yes.
Transforming data: constructing a common rubric	With a range of different statistical methods used across the different studies, care must be taken to provide effect sizes as a common rubric wherever possible.	Yes.
Translating data	With a relatively narrow range of qualitative evidence, this seems unnecessary.	No.
Tabulation	A range of tables, including summaries of studies (Table 3.5), quality appraisal (Table 3.6) and tools used to guide the syntheses approach (e.g. Table 3.4), provide further clarity to the reader and reviewer.	Yes.
Vote-counting as a descriptive tool	Whilst the utility of this method is debatable, it would add some overall clarity to simplify the evidence through an overall number of studies rejecting the null hypothesis (that OCD and autism symptoms do not overlap), versus the number of studies which fail to reject the null hypothesis. It would be prudent to weight these votes, with a scoring of 3 for high quality studies, scoring of 2 for medium and 1 for low.	Yes

Table 3.3. *Selection of tools and techniques for exploring relationships between studies as recommended by Popay et al. (2006).*

Name of tool/technique	Comments in relation to current synthesis	Should, as has, this tool/technique be applied here?
Graphs, frequency distributions, funnel plots, forest plots and L'Abbe plots.	The studies vary across a range of methods, therefore, comparative data would be difficult to achieve and relatively unclear.	No. Whilst this may provide some utility to the reader, it is not strictly necessary, particularly as the information is provided within the text and tables.
Moderator variables and subgroup analyses.	As above, this can be difficult to achieve	Yes. Despite the issue raised with the lack of comprehensive data, a section will be included to raise the issue of different moderating variables identified through different designs/samples etc.
Developing conceptual models.	This approach seems unnecessary due to the limited range of evidence yielded from the research question and subsequent search method.	No.
Concept mapping.	This would provide the reader with more clarity on the relationship between the evidence from the different studies and enable a theoretical assessment of the research questions.	Yes.
Translation as an approach to exploring relationships.	Specialised approach, may be more of value to exploring relationships. Can be subjective and lack a certain degree of transparency.	No. Popay et al. (2016) recommend against this approach in the full context of a narrative synthesis. The general approach is taken through regardless.
Qualitative case descriptors.	Comparison between studies on methods used as an explanation of the potential reason for any different outcomes.	Yes. The narrative review includes a comparison between different outcomes based on varying methods used.
Investigator triangulation and methodological triangulation.	This is difficult to achieve by the single reviewer as in this investigation.	Yes. A critical approach is taken to identify any differences in outcomes as a consequence of the viewpoint any different disciplinary approach affecting the evidence.

3.4. Results

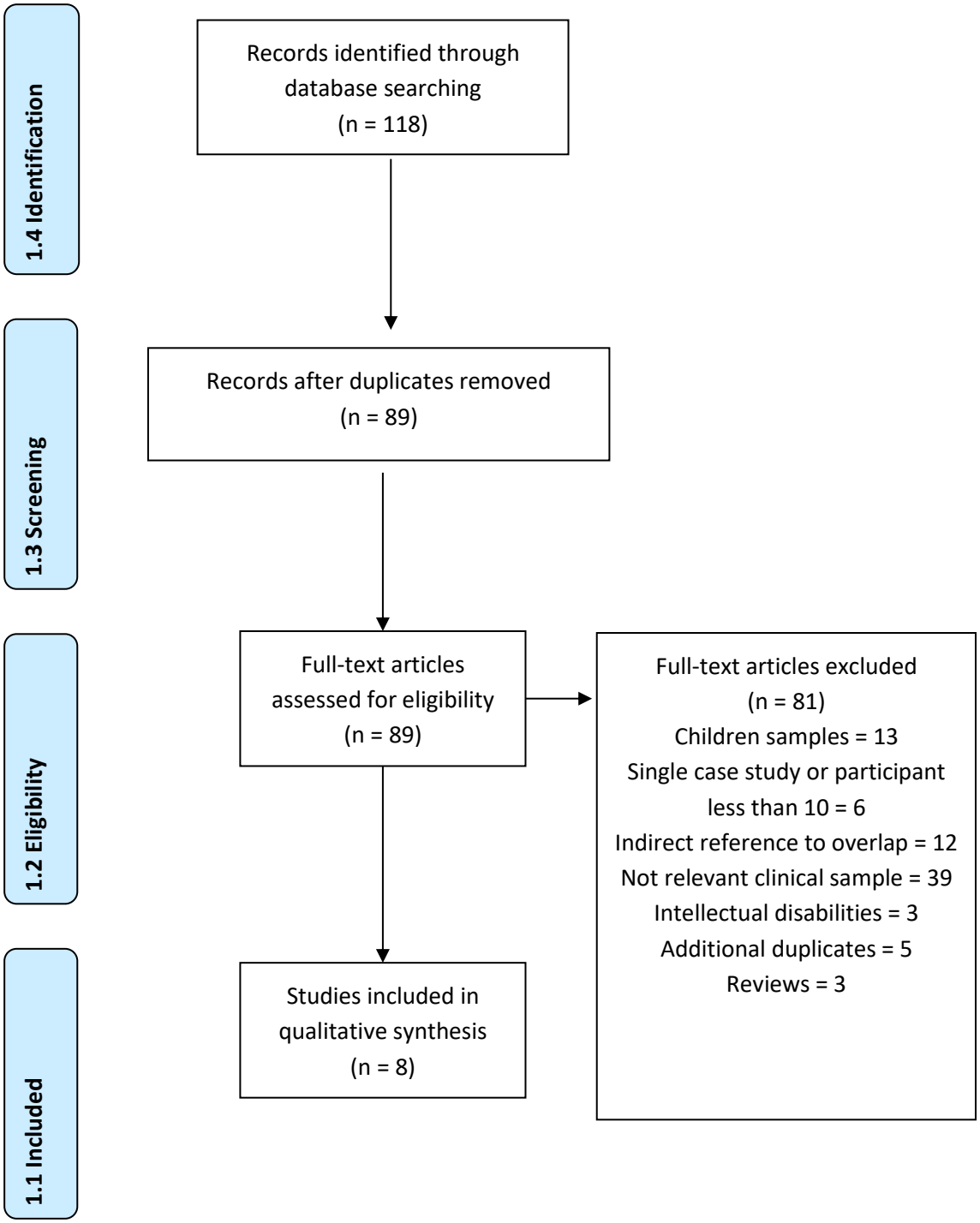

Figure 3.1. PRISMA study selection flow chart (Moher et al., 2009).

Overview of the literature

A summary of the main findings is presented in Table 3.5. The eight included studies consisted of 919 participants: 584 with OCD; 27 with comorbid autism and OCD (including 12 with comorbid autism and OCD or Social Anxiety Disorder); 198 with autism spectrum disorder; and 110 with Hoarding Disorder. As recommended by Popay et al. (2006), grouping characteristics were identified to facilitate the narrative synthesis (see Table 3.4). These groupings indicated studies were best described within either "focus of report" or "population". The decision was made to group studies according to focus of report, as this would give the most theoretic clarity within the review.

Table 3.4. *Grouping characteristics for comorbidity systematic literature review studies.*

Group according to:	
Focus of report	Population
Comorbidity rates (Pertusa, de la Cruz, Alonso, Menchón, & Mataix-Cols, 2012; Roy, Prox-Vagedes, Ohlmeier and Dillo, 2015; Rydén & Bejerot, 2008)	Autism and OCD in psychiatric sample (Rydén & Bejerot, 2008)
Symptom overlap studies (Anholt et al., 2009; Boerema, de Boer, van Balkom, Eikelenboom, Visser, & van Oppen, 2019; Cath Ran, Smit, Balkom and Comijs, 2008; Pertusa et al., 2012, Wikramanayake et al., 2018)	OCD in Asperger's syndrome sample (Roy et al., 2015)
Neuroanatomical (Tsuchiyagaito et al., 2017)	Autism symptoms in OCD patients (Anholt et al., 2009; Boerema et al., 2019; Wikramanayake et al., 2018)
	Comorbid OCD and ASD (Cath, 2008; Tsuchiyagaito et al., 2017)
	Separate OCD and autism group (Pertusa et al., 2012)
	Asperger's and/or high-functioning autism (Roy et al., 2015)
	Hoarding disorder (Boerema et al., 2019; Pertusa et al., 2012)

Table 3.5. *Characteristics of comorbidity systematic literature review studies.*

Reference	Design	Participants	Statistical tests	Outcome measure	Results and notes	Quality appraisal (see Table 3.6)
Anholt et al. (2009)	Cross-sectional questionnaires	1) 109 OCD patients (DSM-IV confirmed). 39% males, mean age 37.5 years, SD 10.0). 2) 87 neurotypical controls (53% males, mean age 37.6 years, SD 13.6). 72% OCD patients receiving medication.	Pearson's correlations to assess the interrelationships between the scales. Chronbach's alpha to measure internal consistency.	Comparison of difference between Y-BOCS-assessed OCD traits, Autism-Quotient (AQ) assessed autism traits and ADHD interview-assessed ADHD traits.	Chronbach's alpha of all scales were reasonable to high. AQ total scores were identified to be related to severity of OCD symptoms (*r* (109) = 0.56, Zr = 0.63). The attention switching AQ subscale had the highest correlation with OCD traits, between (*r* (109) = 0.42, Zr = 0.45) and (*r* (109) = 0.45, Zr = 0.48). AQ subscales social skills (*r* (109) = 0.45; Zr = 0.48) and attention switching (*r* (109) = 0.27; Zr = 0.28) were significantly correlated with Y-BOCS severity, as well as all OCD dimensions. OCD participants demonstrated higher AQ scores than controls (*F* (1, 193) = 68.8, d = 1.19). 4.2% of OCD participants scored above AQ cut-off point, compared to 1.2 % controls. AQ did not predict hoarding symptoms. Hoarding symptoms – only two items on the Y-BOCS – were associated with autism symptoms, but lower than other dimensions. Hoarding symptoms were associated with inattentiveness and increased age. AQ-measured attention to detail scores demonstrated low correlation with OCD and didn't predict symptoms or severity of the disorder.	High.

Reference	Design	Participants	Statistical tests	Outcome measure	Results and notes	Quality appraisal (see Table 3.6)
Boerema et al. (2019)	Cross sectional between subjects comparison of comorbidities and demographic differences between OCD-only and OCD-hoarding participants.	Entire set of a sample of 419 adults with OCD (The Netherlands Obsessive Compulsive Disorder Association). Two subgroups identified: 58 participants with OCD-hoarding (mean age 38.02 years; 44.8% male); and 349 non-hoarding OCD participants (mean age 36.53 years; 44.1% male).	Odds ratios calculated through logical regression analyses.	Relevant to the present review, Autism Quotient scores were compared between the groups, alongside Hoarding symptoms and OCD traits (via the Y-BOCS).	Mean AQ scores in hoarding participants 124.21 (SD = 16.34) versus mean 112.62 (SD = 15.79) non-hoarding participants. Independently of OCD, hoarding symptoms in the OCD-hoarding participants significantly associated to higher Autism Quotient (AQ) scores (b = 0.71, p < .001, OR = 2.09 (95% CI: 1.34, 3.26), in addition to more severe anxiety, and depression, whilst unrelated to ADHD, PTSD and childhood trauma. Association between hoarding and autism remains when controlling for severity of OCD symptoms, relationship between hoarding and anxiety or depression does not. Hoarding symptoms associated with early-age onset of OCD, more severe and higher scores of OCD symptoms (across all trait categories; disappears when controlling for number of OCD traits.	High.
Cath et al. (2008)	Cross-sectional questionnaires	4 groups of adult participants: 12 participants with comorbid autism and OCD or social anxiety disorder (SAD); 12 participants with OCD, 12 participants with SAD; 12 neurotypical controls.	Pearson's correlations to assess the interrelationships between the scales, between groups.	Comparison of difference between Autism Quotient, Y-BOCS, Liebowitz Social Anxiety Scale, Beck Anxiety Inventory and novel egodystonia questions related to OCD behaviours.	Lower OCD symptom severity in comorbid OCD/autism versus pure-OCD participants (large effect size, over 1 standard deviation; *d* = -1.00), though higher than controls (large effect size, over 1 standard deviation; *d* = 1.92). No significant differences on the novel ego-dystonic measures of repetitive traits between the autism-comorbid OCD participants and OCD-only participants. No significant differences identified in the type of OCD traits reported by the two groups.	Medium to high.

Reference	Design	Participants	Statistical tests	Outcome measure	Results and notes	Quality appraisal (see Table 3.6)
Pertusa et al. (2012)	Between-group comparison of traits/symptoms between clinical participants.	221 patients were recruited from clinics in England (60.6%) and Sweden: 64 autistic (mean age 30.7 years; 47.8% males; 98.4% Caucasian); 52 with hoarding disorder (mean age 49.4 years; 23.1% males; 90.4% Caucasian); 31 with nonhoarding OCD (mean age 39; 38.7% males; 90.3% Caucasian); 19 with anxiety disorders (mean age 37.8 years; 31.6% males; 94.7 Caucasian); and 55 neurotypical controls (mean age 37.4 years; 50.9% males; 100% Caucasian).	Continuous data (e.g. personal characteristics) analysed using t-tests and ANOVAs. Nonparametric Spearman correlations were used to analyse correlations between the Eyes Test, AQ and Saving Inventory-Revised, with linear regression analyses being calculated on these data.	Differences in Autism-Spectrum Quotient, "Reading the Mind in the Eyes" (theory of mind measure), alongside hoarding severity (Saving Inventory-Revised) between the groups and OCD traits (Dimensional Y-BOCS).	Mean total Autism Quotient (AQ) scores significantly higher in the autistic versus OCD and Hoarding Disorder groups. AQ scores were comparable between the OCD and Hoarding Disorder groups, which were higher than neurotypical controls. Autistic traits specifically related to OCD traits in Hoarding Disorder participants: AQ scores exclusively predicted by OCD traits scores in this group (Adjusted R^2 = .59; β = .80; *p* = .006).	High.
Roy, Prox-Vagedes, Ohlmeier and Dillo (2015)	Within-subject design of psychiatric reports (interviews) of adults with Asperger's syndrome.	50 adults (mean age 36.46 years) with Asperger's syndrome (68% males).	Comorbidities are presented as percentages of the group.	Evidence of psychiatric comorbidities of participants.	OCD reported in 14% of the sample. No other conclusive evidence was presented within the study: reference only was made to previous reports of potential differences between OCD and autism.	Low.

Reference	Design	Participants	Statistical tests	Outcome measure	Results and notes	Quality appraisal (see Table 3.6)
Rydén and Bejerot (2008)	Naturalistic study. Between subjects comparison of comorbidities (and other factors) in autism versus non-autistic psychiatric patients.	All autism-diagnosed patients within a psychiatric unit (*n* = 84; 54% male; mean age 30 years) and 34 psychiatric patients without autism or ADHD (47% male; mean age 34 year). Exclusion criteria included IQ < 70. A significant proportion of all the participants had received a prior diagnosis of OCD.	T-tests were used to test the significance of differences between the two groups.	Differences in demographic factors, psychiatric comorbidity and personality traits.	Comparative comorbidity of OCD in autistic psychiatric participants (23%) and control psychiatric patients (16%). Many other comorbid disorders were reported by the autism group including social phobia (17%). Depression only significantly different rate of comorbidity between the two groups (49% autism and 68% non-autistic patients), although higher rates may be masked by difficulties in assessing in autistic samples. Participants with autism demonstrated lower levels of social and occupational functioning compared to non-autistic psychiatric participants, despite comparable levels of education. Lack of insight significantly higher in the autistic sample.	High.

Reference	Design	Participants	Statistical tests	Outcome measure	Results and notes	Quality appraisal (see Table 3.6)
Tsuchiyagaito et al. (2017)	Comparison of CBT outcomes plus structural MRI comparison between two groups.	37 adults with OCD: 15 with comorbid autism (mean age 29.53 years, 27% females); 22 with OCD only (mean age 34 years, 77% females).	1. Multilevel linear mixed model at three time points (before, during and after 11 week CBT intervention) of Y-BOCS scores. 2. T-tests to compare group differences in whole brain volumes.	Clinical change as a consequence of treatment, as measured by Y-BOCS scores.	Participants with comorbid autism and OCD responded less well to Cognitive Behavioural Therapy than participants with non-comorbid OCD: significant Y-BOCS difference between both the mid (mean difference = 5.48; small effect size, *d* = 0.96) and post-treatment responses (mean difference = 6.16; small effect size, *d* = 1.07) between these two groups. Higher reports of disability, daily impairment and comorbid depression and/or other anxiety disorders in the autism-comorbid group. Structural MRI evidence: comorbid-autism OCD participants demonstrates significantly smaller grey matter volume in the left occipital lobe (visuospatial processing). Left superior frontal gyrus smaller in the OCD-only group compared to the comorbid autism group. Response to treatment attributed specifically to neural circuitry (abnormalities in the dorsolateral prefronto-striatal circuit, responsible for "executive" functions such as planning) across all participants.	High
Wikramanayake et al. (2018)	Within-subjects comparison of high versus low autism symptom comparisons in OCD sample on demographic factors related to comorbidity and treatment response.	73 adults with OCD (from 106 consecutive referrals to OCD clinic). 53.4% females. Mean age between 44.47.	T-tests to compare high- versus low-AQ scoring samples.	1. Association between OCD and autism symptoms. 2. Clinical change as a consequence of treatment, as measured by Y-BOCS scores.	Positive significant correlation between total Y-BOCS and total AQ scores. No significant difference in the total Y-BOCS, compulsions or obsessions between high-AQ and low-AQ samples. AQ measure of Attention Switching specifically related to OCD: high-AQ participants reporting increased impairments compared to the low-AQ participants (both significantly higher than controls). Lower insight into OCD symptoms in high-versus low-AQ score participants, with a large effect size (effect size = -0.65 [-1.13, -0.15], *p*=0.1).	Medium.

Table 3.6. *Critical appraisal questions by Aveyard (2010, p103) for comorbidity systematic literature review studies.*

Study	**Journal quality**	**Clear research question, appropriate for research?**	**Valid research method?**	**Sufficiently large sample?**	**Appropriate sample?**	**Data collection appropriate to the research method?**	**Appropriate statistical test?**	**Quality – overall appraisal**
Anholt et al. (2009)	Good. Peer-review with impact factor 3.341	Yes.	Yes.	Yes.	Mostly. Participants were not screened for autism.	Yes.	Yes.	High.
Boerema et al. (2019)	Good. Peer-review with impact factor 3.786	Yes.	Yes.	Yes.	Yes. There were no significant differences in age, gender and education levels in OCD-hoarding and OCD without hoarding participants.	Yes.	Yes.	High.
Cath et al. (2008)	Quite Good. Peer-review with impact factor 1.06	Yes.	Yes.	Relatively small sample size (n = 12) in each group, and lack of sub-group comparison.	Yes, although may have benefited from an autism-only sample too.	Yes.	Yes.	Medium to high.
Pertusa et al. (2012)	Good. Peer-review with impact factor 5.004	Yes.	Yes.	Yes.	Yes. Screening not an issue as comorbidity addressed as a fundamental part of the research question. Whilst the hoarding disorder group was significantly older, with more females, this was reported to statistically not affect the results.	Yes.	Yes.	High.

Study	**Journal quality**	**Clear research question, appropriate for research?**	**Valid research method?**	**Sufficiently large sample?**	**Appropriate sample?**	**Data collection appropriate to the research method?**	**Appropriate statistical test?**	**Quality – overall appraisal**
Roy et al.(2015)	Quite good. Peer-review with impact factor 0.43	Yes.	Partly – validity impaired by lack of statistical analyses.	Yes.	Yes. Screening not an issue as comorbidity addressed as a fundamental part of the research question.	Yes.	No. Percentages, but no statistical analyses used.	Low.
Rydén and Bejerot (2008)	Good. Peer-review with impact factor 1.84	Yes.	Yes.	Yes.	Partly. High rates of female participants, possibly due to the psychiatric setting used (potentially different demographic). Screening not an issue as comorbidity addressed as a fundamental part of the research question.	Yes.	Yes.	High.
Tsuchiyagaito et al. (2017)	Good. Peer-review with impact factor 3.532	Yes.	Yes.	Small sample size (*n* = 12) in autism + OCD group.	Yes.	Yes.	Yes.	High
Wikramanayake et al. (2018)	Good. Peer-review with impact factor 1.337	Yes.	Yes.	Partly, not for statistics involving the subgroup OCD plus high AQ traits without autism diagnosis (*n* = 8)	Yes. Consecutive referrals with diagnoses confirmed by in-depth clinical assessment.	Yes.	Lack of statistical analyses (e.g. null hypotheses testing) for AQ versus OCD traits.	Medium.

3.4.1. Comorbidity rates.

Comparable results were identified across both high quality studies investigating comorbidity of OCD in autistic adult psychiatric samples. Whilst Pertusa et al. (2012) reported 26.6% comorbidity of OCD in a sample of 64 participants from specialist autism clinics, Rydén and Bejerot (2008) similarly reported 23% comorbidity with OCD across 84 autism-diagnosed patients. This sampling method would appear to reduce the likelihood this sample is a true representation of the wider population. This is emphasised by Rydén and Bejerot's (2008) finding of statistically comparable rates of comorbid OCD in the autism sample and the control sample of psychiatric patients without autism. Male and female differences were reported: whilst the presentation of autism features was comparable, females were identified to show more attention and emotional issues and borderline personality traits. However, it is possible the female participants are captured within this psychiatric setting as missed diagnoses of autism may be more likely in females, therefore only come to clinical attention through other routes (e.g. psychiatric settings). Regardless, the comorbidity is much larger than found in the population generally; therefore, comorbidity between autism and OCD appears meaningful even if depression and psychiatric diagnoses are mediating variables.

A study by Roy et al. (2015) presented evidence of psychiatric comorbidities of adult participants with Asperger's syndrome. However, comorbidities are presented as percentages of the group, rather than statistically analysed against a null hypothesis. Whilst OCD was reported in 14% of this sample, no other conclusive evidence was presented within the study.

3.4.2. Symptom overlap studies.

Four studies used cross-sectional questionnaires to compare symptom overlaps between autism and OCD (Anholt et al., 2009; Boerema et al., 2019; Cath et al., 2008; Pertusa et al, 2012). Each of these studies recruited OCD participants to measure autism symptomology using the Autism-Spectrum Quotient (AQ), alongside OCD traits via the Yale-Brown Obsessive Compulsive Scale (Y-BOCS; Anholt et al., 2009; Boerema et al., 2019; Cath et al., 2008), or Dimensional Y-BOCS (Pertusa et al, 2012), in addition to hoarding symptoms (Boerema et al., 2019l Pertusa et al., 2012).

Considerable overlaps between OCD and autism symptoms across these studies indicate relatedness between the disorders. OCD participants appear to display higher AQ scores than controls ($F(1, 193) = 68.8$, d = 1.19) (Anholt et al., 2009): with 4.2% of OCD participants scoring above the AQ cut-off point, compared to 1.2 % controls. Furthermore, AQ total scores may be related to severity of OCD symptoms (r (109) = 0.56, Zr = 0.63), with the Attention Switching AQ subscale demonstrating the highest correlation with OCD traits, between (r (109) = 0.42, Zr = 0.45) and (r (109) = 0.45, Zr = 0.48). However, the AQ measure of “attention to detail” does not appear to predict OCD symptoms or severity (Anholt et al., 2009), indicating the “just right” phenomena in OCD and Insistence on Sameness in autism may be unrelated.

Hoarding symptoms, specifically, may be related to autism symptom. Although Anholt et al. (2009) reported hoarding scores to be associated with autism symptoms, they were not found to predict hoarding symptoms, although the use of Y-BOCS as a measure of hoarding contains only two hoarding items. Comparable mean total AQ scores have been reported between participants with OCD and with Hoarding

Disorder, whilst autism traits specifically in Hoarding Disorder participants appear to be related to OCD symptoms (Pertusa et al., 2012): AQ scores exclusively predicted by OCD traits scores in this group (Adjusted R^2 = .59; β = .80; *p* = .006). Boerema et al., (2019) reported hoarding symptoms to be significantly associated with higher AQ scores, independently of OCD (b = 0.71, p < .001, OR = 2.09 (95% CI: 1.34, 3.26). This association remained even when controlling for severity of OCD symptoms, whilst the relationship between severe anxiety and depression with hoarding symptoms in this group did not.

In a lower quality, mainly due to the small sample size, but highly relevant study, Cath et al. (2008) suggested individuals with autism-comorbid OCD may experience OCD traits as equally unwanted (ego-dystonic), regardless of fewer obsessive traits reported. No significant differences were found on the novel ego-dystonic measures of repetitive traits between the autism-comorbid OCD participants and OCD-only participants and no differences in the type of OCD traits reported by the two groups (although small sample size). Comorbid OCD and autism participants were identified to report lower OCD symptom severity than pure-OCD participants (large effect size, over 1 standard deviation; *d* = -1.00), though higher than controls (large effect size, over 1 standard deviation; *d* = 1.92); mainly due to lower obsession scores in the comorbid autism group compared to the OCD-only group (large effect size, over 1 standard deviation; *d* = -1.10). Overall, the presence of some differences between the autism-comorbid and the OCD-only groups is indicative of a valid autism subtype in OCD.

Despite a lack of useful statistical analyses (in particular descriptive statistics for AQ scores and null hypotheses testing), Wikramanayake et al. (2018) nevertheless

provided more evidence indicating the relevance of an autism-subgroup within OCD: AQ scores were reported to be lower than in previously published comparative group of autistic but non-OCD participants. Positive significant correlations were reported between the total Y-BOCS and total AQ scores, although there was no significant difference in the total Y-BOCS, compulsions or obsessions between the high-AQ (>25) and low-AQ (<26) samples. 28.7% of the group received an autism diagnosis, of which had been previously undiagnosed. The AQ measure of Attention Switching was specifically related to OCD, with the high-AQ participants reporting impairments in this domain compared to the low-AQ participants, who in turn reported significantly more than neurotypical controls. Participants with high-AQ scores demonstrated lower insight into OCD symptoms than low-AQ scorers, with a large effect size (effect size = -0.65 [-1.13, -0.15], p=0.1).

3.4.3. Neuroanatomical.

Whilst slightly less relevant, Tsuchiyagaito et al. (2017) demonstrated variable response to CBT in patients with comorbid OCD and autism, compared to OCD-only participants. With a significant reduction in Y-BOCS scores, at both the mid (mean difference = 5.48; small effect size, d = 0.96) and post-treatment responses (mean difference = 6.16; small effect size, d = 1.07), between these two groups, the researchers suggested to be related to the potentially mediating effects of the higher reports of disability and daily impairment in the comorbid autism and OCD group. Comorbid depression and/or other anxiety disorders was significantly higher in the autism-comorbid OCD group compared to OCD-only group, although this has been suggested to be specifically related to the OCD symptoms (Boerema et al., 2019). Structural MRI comparisons indicated response to treatment was attributed

specifically to neural circuitry across all groups: abnormalities in the dorsolateral prefronto-striatal circuit (responsible for “executive” functions, such as planning), predicted response to treatment, regardless of autism symptoms. However, Tsuchiyagaito et al. (2017) emphasised there are potentially many moderating factors in comorbidity between OCD and autism, such as comorbid depression and anxiety.

3.4.4. Moderator variables and subgroup factors

Some moderating subgroup factors were addressed across the studies, specifically hoarding disorder in OCD (Boerema et al., 2019; Pertusa et al., 2012) and Asperger’s and/or high-functioning autism within autism (Roy, Prox-Vagedes et al., 2015). Also, the moderating effect of anxiety and depression has also been reported (Boerema et al., 2019; Cath et al., 2008; Tsuchiyagaito et al., 2017).

However, the evidence here provides little insight into any differences in autism symptoms across subtypes of OCD, specifically none for age of onset (early versus late) and subtype of OCD (e.g. germ phobia, magical thinking). Furthermore, little statistical evidence is provided for age, gender or culture as mediating factors.

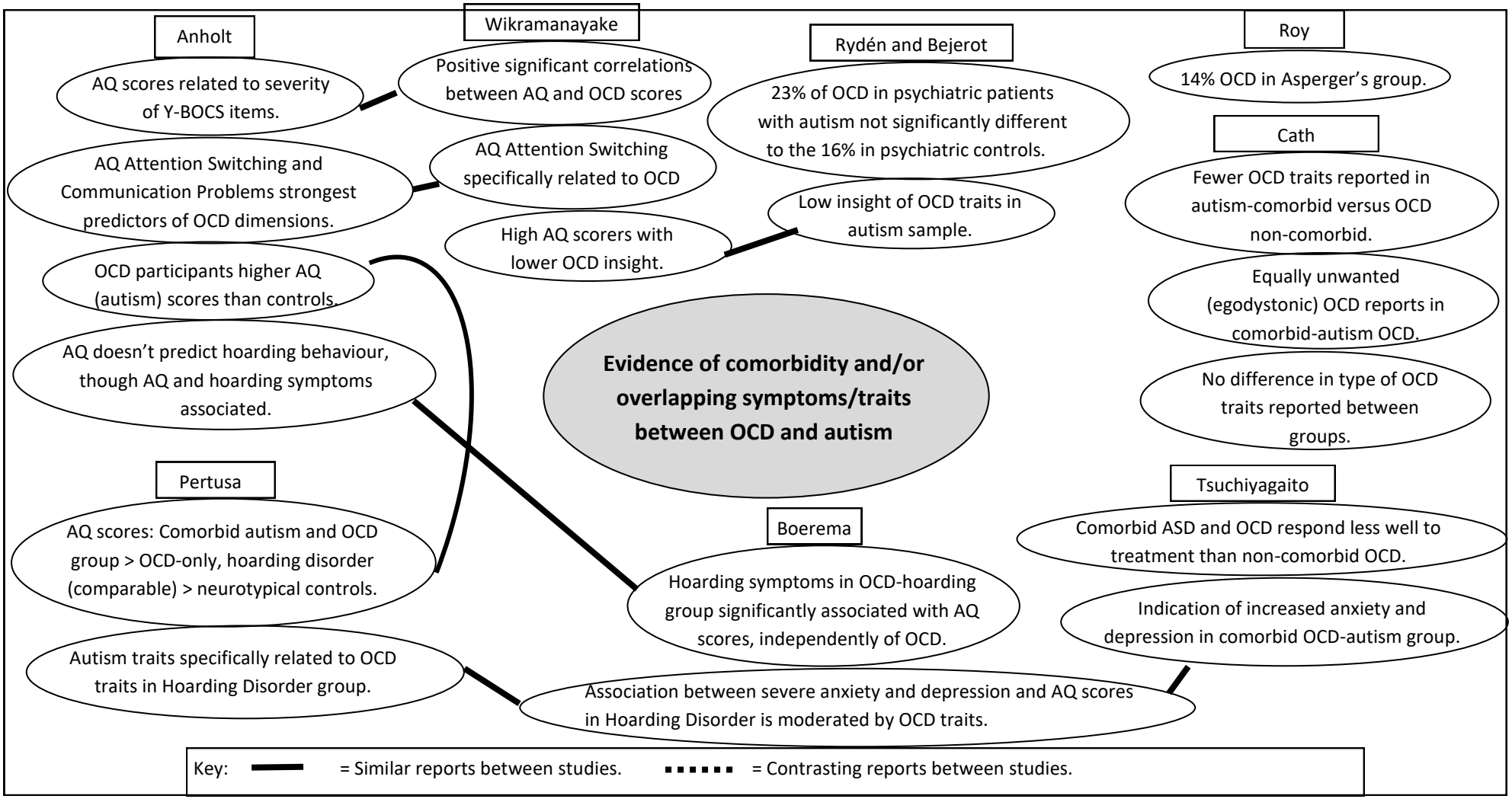

Figure 3.2. Conceptual mapping following study outcomes for comorbidity systematic literature review studies.

3.5. Summary

The evidence of an overlap between OCD and autism is strong. Vote counting identified seven of the eight studies indicated a degree of overlap between autism and OCD, whilst only one study (Tsuchiyagaito et al., 2017) provided no such evidence. More significantly, large effect sizes were demonstrated, which were: the relationship between autism scores and severity of OCD symptoms in OCD participants (Anholt et al., 2009); lower OCD scores in autism-comorbid versus OCD-only groups (Cath et al., 2008); a negative correlation insight into OCD symptoms and autism scores in OCD participants Wikramanayake et al., 2018); and decreased response to Cognitive Behavioural Therapy in autistic versus OCD participants (Tsuchiyagaito et al., 2017).

Figure 3.2 illustrates a clearer perspective of the results identified in this review. Consistently reported is the positive relationship between Autism Quotient scores and OCD traits (Anholt et al., 2009; Wikramanayake et al., 2018) and higher AQ scores in OCD participants compared to neurotypical controls (Anholt et al., 2009; Pertusa et al., 2012). An association between Hoarding disorder and Autism Quotient scores was consistent in the two studies investigating this subtype of OCD (Anholt et al., 2009; Boerema et al., 2019). Additionally, anxiety and depression has been reported to be significantly increased when autism is comorbid with OCD (Tsuchiyagaito et al., 2017), with indication also anxiety and depression is related to hoarding disorder in OCD (Boerema et al., 2019).

Overall, the presence of similarities, notably the significant relationship between AQ scores and OCD traits (Anholt et al., 2009; Boerema et al., 2019; Pertusa et al., 2012; Wikramanayake et al., 2018) and comparable presentation of OCD traits in comorbid

autism participants (Cath et al., 2008), alongside differences in the severity of AQ or OCD scores in subgroups between OCD-only, Hoarding Disorder and comorbid OCD and autism groups, indicates the potential relevance of an autism subtype of OCD.

However, the evidence presented here is still limited, generally failing to identify any causal relationships of symptomological overlap between autism and OCD. There is some evidence for some mediating factors, such as anxiety and depression (Boerema et al., 2019; Tsuchiyagaito et al., 2017) and low insight of OCD traits (Rydén & Bejerot, 2008; Wikramanayake et al., 2018). However, it lacks a comprehensive account for the effect of moderating and mediating variables (such as age, gender, intelligence, other comorbidities). Whilst there is indication of relatedness between OCD and autism, the absence of accounting sampling various subgroups (e.g. late- versus early-onset OCD) within both disorders confounds a comprehensive account at this stage. As argued by Rutter (1997), understanding these underlying mechanics is critical to the validity of comparing these two disorders – see section 3 (Background).

In conclusion, the evidence indicated by the narrative synthesis suggests it may be both valid and necessary to continue to assess the possible overlap/comorbidity between OCD and autism. The next chapter will review repetitive traits in greater detail to further identify the potential validity of understanding the overlap between autism and OCD.

Chapter 4. Background: Repetitive Traits in Typical Development, Obsessive-Compulsive Disorder and Autism

To establish whether it is valid to compare OCD and autism within a symptomological framework (based on repetitive traits), the function and type of repetitive traits across these disorders must be identified and understood. Understanding repetitive traits generally, through typical development, is necessary before understanding these symptoms in a clinical sense.

4.1. Repetitive Traits in Typical Development

"Repetition is an important aspect of all levels of behaviour."

(Ridley, 1994, p. 222)

Repetitive traits – to some extent – are ubiquitous in everyday life (Keren, Boyer, Mort, & Ellam, 2010), from daily routines to cultural rituals (Boyer & Liénard, 2006). They are an important stage in early typical development (Stahl & Pry, 2005), peaking roughly between the ages of 2 and 4 years (Evans et al., 1997). Accordingly, Lewis and Kim (2009) warned that care must be taken to interpret the phenomenology of repetitive traits across neurological disorders. It has been argued that diagnosis must be understood at a behavioural level: symptoms must reflect an underlying cause rather than co-occurring by chance (Happé, 1994).

4.2. Comparing Repetitive Traits in Obsessive-Compulsive Disorder and Autism

Overview

OCD is characterised by interacting symptoms of repetitive traits (i.e. thoughts and/or behaviours), known respectively as obsessions and compulsions. Obsessions are intrusive thoughts, urges or images initially causing anxiety or distress (APA, 2013). Compulsions are the repetitive traits – rigidly applied rules (Worden & Tolin, 2014) – which are enacted as an attempt to alleviate the anxiety created by obsessions. Diagnosis is dependent on significant levels of severity (including interference, distress and lack of control; Goodman et al., 1989a), with time spent in excess of one hour per day (DSM-5, APA, 2013), but in extreme cases more than eight hours, or constant intrusion.

Repetitive Behaviours – a term used in autistic research as a measure of repetitive traits – are regarded so fundamental to autism their presence is one of the two criteria for diagnosis in children (APA, 2013). Whilst Repetitive Behaviours become less pronounced in adults with autism – to such an extent diagnosis in this population is not dependent on the presence of these symptoms (APA, 2013) – it is suggested Repetitive Behaviours do not become irrelevant during later life, rather experience tends to encourage adults to suppress them, to fit in with social norms. This is perhaps not too dissimilar from the passive avoidance (e.g. Storch et al., 2010; Worden & Tolin, 2014), which may mask underlying OCD features in some sufferers. Evidence does suggest Repetitive Behaviours are significant in adults with autism (e.g. Chowdhury, Benson, & Hiller, 2010; Esbensen, Seltzer, Lam, & Bodfish, 2009; Howlin, Goode, Hutton, & Rutter, 2004; Piven, Harper, Palmer, & Arndt, 1996; Seltzer et al., 2003), with some studies even indicating that later-life changes (decreases) in this domain

may be less pronounced compared to social domains (Fecteau, Mottron, Berthiaume, & Burack, 2003; Lord, Bishop, & Anderson, 2015).

Repetitive Behaviours are not unique to autism; they are prominent symptoms across mental and neurological disorders (Ridley, 1994), meaning co-morbidity is an issue. However, it appears there are autism-specific patterns of Repetitive Behaviours, differing to traits demonstrated in other groups, such as individuals with language disorder (Barrett, Prior, & Manjiviona, 2004), developmental disorder (Goldman, Wang, Salgado, Greene, Kim, & Rapin, 2009) and general learning disabilities (Bodfish et al., 2000) as well as age and ability matched controls (Bodfish et al., 2000; Goldman et al., 2009). Whilst Repetitive Behaviours in individuals with autism may be inversely correlated with age and adaptive skills (Esbensen et al., 2009) as well as level of functioning (Cuccaro et al., 2003; Szatmari et al., 2006), there is also evidence severity of Repetitive Behaviour may be associated with severity of autistic symptoms (Bodfish et al., 2000). An autism-specific pattern of Repetitive Behaviours, with an emphasis on "insistence on sameness" and sensory symptoms (Barrett et al., 2015), may be in part a reflection of the autistic condition.

4.2.1. Type of repetitive traits in Obsessive-Compulsive Disorder.

OCD is recognised as a heterogeneous disorder, with a wide range of repetitive symptoms varying from one person to the next. For example, the most widely used clinical tool for OCD, the Yale-Brown Obsessive Compulsive Scale (Goodman et al., 1989b), suggests 58 traits grouped under eight categories for obsessions (aggressive, contamination, sexual, hoarding/saving, religious, symmetry, somatic and 10 miscellaneous) and seven for compulsions (cleaning/washing, checking, repeating,

counting, ordering/arranging, hoarding/collecting and seven miscellaneous items). Whilst most individuals with OCD may demonstrate multiple obsessions and compulsions (Hollander & Evers, 2004), it is commonly recognised OCD traits tend to cluster amongst certain themes, such as: fear of contamination/cleaning; obsession over symmetry and ordering; obsessional checking and hoarding; and sexual/religious obsessions (Auoizerate et al., 2004; Mataix-Cols et al., 2005).

4.2.2. Repetitive Traits in Autism

Repetitive traits in autism are commonly referred to as Repetitive Behaviours. The symptom category called "restricted and repetitive behaviours" are characterised by their "repetition, rigidity, invariance, and inappropriateness" (Turner, 1999). They are clinically defined as repetitive motor movements, insistence on sameness (routines and patterns of behaviour), highly restricted interests and hyper- or hypo-reactivity to sensory input (APA, 2013). Many researchers loosely subdivide the broad range of repetitive traits in autism into two categories of "lower-level" movement behaviours and more complex "higher-level" behaviours (e.g. Bodfish, 2007; Turner, 1999), referring to the complexity and degree of cognitive processing. A range of factor-analytical investigations have been identified through the process of reviewing relevant literature on repetitive traits in autism as part of the first two years of the present thesis process. An overview of the main findings of these factor-analytical investigations has been reproduced in Table 4.1. This appears to validate the general support for the dichotomous distinction between lower- and higher-level repetitive traits, classified in a continuum based on increasing cognitive complexity.

Lower-level repetitive traits are defined as “repetitions of movement” (Turner, 1999). These behaviours include stereotyped movements, repetitive manipulation (including ordering) of objects, repetitive types of self-injurious behaviours, dyskinesias and tics. Whilst lower-level repetitive traits appear to be significantly prevalent in autism (e.g. Bodfish et al., 2000; Goldman et al., 2009), even when matched by age, gender and level of functioning (Bodfish et al., 2000), they are not unique to the disorder and may be related to developmental age (Carcani-Rathwell, Rabe-Hasketh, & Santosh, 2006; Militerni, Bravaccio, Falco, Fico, & Palmero, 2002) and/or level of functioning (Richler, Huerta, Bishop, & Lord, 2010). Higher-order repetitive traits are cognitively more complex. Such traits include insistence on sameness, circumscribed (narrow) interests, rituals, routines and compulsions (Bodfish, 2007; Turner, 1999). It is claimed such traits may be specific to autism (e.g. Carcani-Rathwell et al., 2006), with insistence on sameness and circumscribed interests, in particular, reflecting the narrow processing style of autism (Baron-Cohen, 2008).

Table 4.1. *Factor analytical studies of the categories of repetitive traits in autism, organised in levels of proposed cognitive complexity.*

Cognition

Lower level ◄——————————————————————————————► Higher level

Bourreau, Roux, Gomot, Bonnet-Brilhault, and Barthélémy et al. (2009)	Restricted behaviours	Sensorimotor stereotypies	Modulation insufficiency		Reaction to change	
Smith et al. (2009)	Repetitive and stereotyped motor behaviour – simple	Repetitive and stereotyped motor behaviour – complex		Insistence on sameness		Intense preoccupations
Szatsmari et al. (2006)	Motor Behaviours			Insistence on sameness		
Lam and Aman (2007)	Stereotypic behaviour	Self-injurious behaviour		Ritualistic/ sameness behaviour	Compulsive behaviour	Restricted interests
Lam, Bodfish, and Piven (2008)	Repetitive motor behaviour			Insistence on sameness		Circumscribed interests

An OCD-related category has been suggested within the autism condition. The DSM-IV (APA, 2000), recognised "autism-related obsessive-compulsive phenomena (AOCP)" within the autism spectrum, based on similarities in repetitive symptomology between the two disorders. It has been argued these symptoms are so fluid they can transform into clinically relevant OCD (Fischer-Terworth & Probst, 2009). Whilst this category has not been retained in the current DSM-5 (APA, 2013), it seems supported by a significant body of evidence demonstrating various links between the two disorders (e.g. Anholt et al., 2010; Bejerot et al., 2001; Hutton et al., 2008; Hollander et al., 2003; Ivarsson & Melin, 2008; Russell et al., 2005). Whilst autism and OCD are both diagnostically recognised as spectrum disorders (DSM-5, APA, 2013), it is quite probable AOCP reflects not just a category of repetitive symptomology in autism, but a link between the two disorders. There appear to be differences in the type of repetitive traits between autism and OCD, although it is pertinent to investigate this distinction on a phenomenological level.

Whilst repetitive behaviours are reported to be relatively stable across early development (e.g. Joseph, Thurm, Farmer & Shumway, 2013) they are not held to be diagnostically critical in adults with autism; the DSM-5 (APA, 2013) removes this symptom as a requirement in this population. It is unclear whether such traits are no longer relevant to the disorder, or whether they have purely become masked or supressed in the older population. Accordingly, the relevance of repetitive behaviours, so central to diagnosis in children with autism, is unclear in adults. It may be important, therefore, to use the combined literature of repetitive behaviours across the lifespan to inform our knowledge of repetitive behaviours generally, before linking them specifically to autistic adults.

Gender differences have been identified in repetitive behaviours. Whilst there is indication severity of repetitive behaviours are comparable between autistic girls and boys (McFayden, Albright, Muskett & Scarpa, 2019), there are reports of differences in presentation, including increased higher-order repetitive behaviours in girls, consisting of compulsiveness, sameness, restricted and also self-injurious behaviours (Antezana et al., 2019), and narrower range of socially expressed restricted interests (McFayden et al., 2019).

Evidence from tests on executive functioning demonstrates a specific processing style in autism. Repetitive behaviours may be product of this. Therefore, as Baron-Cohen (1989) proposed, core repetitive behaviours in autism may not be ego-dystonic (unwanted and opposed to the individual's sense of self), but instead may be a wanted feature of the disorder.

The preference should be on the phrase "repetitive traits" over "repetitive behaviours", to include clarity on cognitive content to be included in repetitive phenomenology. However, repetitive behaviours will be referred to in this section as this is typically in line with the reported phraseology in the field of such traits in autism.

4.3. Nature of Repetitive Behaviours

Repetitive behaviours are defined to be regularly occurring interests or activities which interfere with daily functioning/performance (Martínez-González & Piqueras, 2018). Repetitive behaviours in adults with autism have scarcely been measured. For this purpose, Barrett et al. (2015) developed a self-reported measure, the Repetitive

Behaviour Questionnaire-2 Adults. A two-factor solution was identified, consistent with the majority of previous literature on autistic children samples: repetitive sensory motor behaviours (RMB) and insistence on sameness (IS). RMB consists of simple repetitive movements, such as such as pacing or finger movements. IS consists of a range of more complex behaviours, such as routines, preference of special objects and "just right" phenomena. Barrett et al. (2015) found repetitive behaviours to correlate with autistic symptoms. Overall, no gender differences were identified and autistic participants self-reported all three subscales (sensory, RMB and IS) significantly higher than neurotypical controls. In this sample of adults of typical intelligence, autistic participants were found to report higher levels of IS compared to RMB.

Support for this two-factor solution is consistent, across repetitive behaviour scales and cross-culturally, such as Georgiades, Papageorgiou and Anagnostou's (2010) low-order (stereotyped movements and self-injurious behaviours) and high-order (compulsions, rituals, sameness and restricted behaviours) solution in a Greek children and adult autistic sample using the Repetitive-Behaviour Scale-Revised (Bodfish et al., 2000). However, there is support for the inclusion of more categories, such as Martínez-González and Piqueras's (2018) six-factor solution, consisting of: stereotypies; self-injuries; compulsions; rituals; sameness; and restricted behaviours. Where three-factor solutions have been identified, self-injurious behaviours have typically been separated out from low-level stereotypies and higher-level sameness behaviours (e.g. Mirenda et al., 2010).

In autistic samples, repetitive behaviours have been reported to be negatively correlated with IQ, sensory processing difficulties, adaptive functioning and behaviour

problems, and positively correlated with autistic symptoms generally (Inada et al., 2015). Such results indicate the potentially complex pathway contributing to repetitive behaviours in autism, possibly through various moderating and mediating factors.

Whilst repetitive behaviours are common across development and other disorders (Boyer& Liénard, 2006; Evans et al., 1997; Happé, 1994; Keren et al., 2010); increased severity has been reported in children and adults with autism (Evans, Uljarević, Lusk, Loth & Frazier, 2017), in addition to suggestions of autism-specific patterns of insistence on sameness traits (Barrett et al., 2015).

4.3.1. Low-order repetitive behaviours.

4.3.1.1. Stereotypies.

The term "stereotypy" is used interchangeably in the literature on repetitive behaviour to define the low-level repetitive behaviours such as repetitive motor and vocal behaviours. Stereotypies have been reported to be significantly increased in children with autism, even from a very early age (Matson & Dempsey, 2009). However, autism traits have been reported not to be associated with stereotypy behaviour – which are common across low-cognitive groups – which indicate these behaviours are not specific to autism (Goldman et al., 2009).

Recent investigations have attempted to look at differing functional aspects to these behaviours by looking at the sensory or vestibular effects which may occur as a consequence. A review on the literature on stereotypies in autism has indicated a high prevalence, as high 88%, significantly higher than in other developmental disabilities and seemingly comparable across children and adults (Chebli, Martin & Lanovaz,

2016). Relative resistance to intervention indicates these traits may be fundamental to autism and non-socially driven (Cividini-Motta, Garcia, Livingston & MacNaul, 2018; Prefontaine, Lanovaz, McDuff, McHugh & Cook, 2019). Vocal stereotypies have been reported to be similar to behavioural stereotypies in autism, appearing not to be socially mediated but a consequence of sensory stimulation (Ahearn et al., 2007).

Cunningham and Schreibman (2007) reviewed literature on stereotypies of children with autism to determine various function of these behaviours. The researchers propose motor and vocal stereotypies have a differential effect on learning: research indicates a negative effect between motor stereotypies and skill development; verbal stereotypies demonstrating a positive association with treatment outcome. The researchers conclude the sensory and auto-reinforcing effects of stereotypies have a wealth of empirical evidence to support it, but there is also a variety of studies indicating these behaviours appear to be socially reinforced and, as such, a more comprehensive model of stereotypy behaviours needs to be identified in future research.

4.3.1.2. Sensory.

Repetitive behaviours have been described as a response to sensory processing difficulties. For example, Schulz and Stevenson (2019) reported a positive correlation between sensory hypersensitivity and repetitive behaviours. Although this relationship was not found to be specific to autism – it was consistent and comparable across all participants – it is still significant as sensory abnormalities are widely accepted to be a key feature in autism spectrum disorders (e.g. Tomchek & Dunn, 2007). This perhaps explains why stronger relationships have been identified between sensory

hypersensitivity and repetitive behaviours in autistic participants (Schulz & Stevenson, 2019).

Neurological evidence presents a compelling case for the relationship between sensory processing and repetitive behaviours in autism. Wolff et al. (2017), for example, reported an association between cerebellar and corpus callosum in autistic infants and, longitudinally, repetitive behaviours in later development. This effect was reported to be uniquely contributed by sensory responsiveness, holding even when controlling for cognitive and social abilities.

Regardless of their specificity to autism, the evidence of the effect of arousal on repetitive behaviours in autism offers a differential pathway from the types of repetitive behaviours which may be more cognitively driven (e.g. Insistence on Sameness). This has been supported by Wigham, Rodgers, South, McConachie and Freeston (2015), who reported mediating effects of anxiety and Intolerance of Uncertainty in autistic participants. The pathway model established by Wigham et al. (2015) indicated a direct path between sensory under-responsiveness to both repetitive motor and sameness behaviours in autism, and between sensory over-responsiveness (but not under-responsiveness) and sameness behaviours. A sequential pathway has also been identified through Intolerance of Uncertainty and anxiety in autism, consistent with more recent findings by Glod, Riby and Rodgers (2019). Furthermore, Glod et al. (2019) identified an autism-specific direct relationship between sensory hyper-responsiveness and sensory motor repetitive behaviours in young children with autism. Clearly a network of repetitive behaviours through

moderators consisting of sensory processing and arousal may be fundamental to certain (low-level) repetitive behaviours.

4.3.2. High-order repetitive behaviours.

Repetitive behaviours are generally ubiquitous in the population and typical development (Keren et al., 2010; Ridley, 1994). However, high-order repetitive behaviours have regularly been reported to be potentially specific to autism. Stereotyped and ritualistic/sameness behaviours have been reported as the most typical repetitive behaviours in autistic pre-schoolers, whilst also appearing to be associated with autistic severity (Fulceri et al., 2016). Furthermore, these ritualistic/sameness behaviours were not predicted by non-measures of non-verbal IQ, indicating this trait may be a distinctive feature of autism, something supported in other studies (e.g. Barrett et al., 2015; Hanson et al., 2016; Georgiades et al., 2010).

4.3.2.1. Interests.

Children with autism have been reported to demonstrate decreased measures of cognitive control, specifically when exposed to interest-related material (Bos et al., 2019), indicative of not only high the impact on their day-to-day life, but also the potential specific processing of such information in autism.

Turner-Brown, Lam, Holzclaw, Dichter and Bodfish (2011) identified 95% of children with autism have been reported by their parents to demonstrate “circumscribed interests”, with 70% demonstrating more than one. Circumscribed interests in children with autism were high in the domain of “folk physics” (56%), consistent with Baron-Cohen & Wheelright (1999). Anthony et al. (2013) found children with autism did not

have fewer interests than their typically developing peers, although their interests were generally more intense.

4.3.2.2. Sameness.

Sameness behaviours (whereby an individual has a desire/need to maintain the environment through strict and seemingly arbitrary rules; Turner, 1999) have been regularly reported to be a specific feature of autism, separate to repetitive motor behaviours (e.g. Barrett et al., 2015; Hanson et al., 2016; Georgiades et al., 2010), which themselves appear to be a feature of other disorders. Using a psychometrically sound measure of sameness (Behaviour and Sensory Interests Questionnaire), Hanson et al. (2016) reported sameness behaviours in autistic children to affect: limited food; changes at school; order; changes in play; routes; placing objects; arranging personal; carry atypical; and atypical counting.

Two-factor models, consisting of repetitive sensory motor behaviour and insistence on sameness, have been consistently reported (e.g. Georgiades, Papageorgiou & Anagnostou 2010; Matson, Boisjoli & Dempsey, 2009; Mooney, Gray, Tonge, Sweeney & Taffe 2009; Szatmari, White & Merikanga, 2007), whilst sameness has been demonstrated to be a robust construct also in thee-factor (Lam, Bodfish & Piven, 2008), four-factor solutions (Chung & Park, 2013), and even five-factor solutions (Scahill et al. 2014). Using the Repetitive Behaviour Questionnaire-2, Leekam et al. (2007) demonstrated sameness to explain 36.9% of the variance, whilst motor behaviours explained 17.1% of the total variance.

In a large scale investigation of the effect of individual differences in repetitive behaviours across the autism spectrum, Hus, Pickles, Cook, Risi and Lord (2007) demonstrated Insistence on Sameness specifically to be "relatively independent of gender, race, diagnosis [autism/Asperger's/PDD-NOS], chronological age, nonverbal and verbal IQ, and autism symptoms," as assessed by both the Autism Diagnostic Interview-Revised and the Autism Diagnostic Observation Schedule. This is in stark comparison to more low-level repetitive (motor and sensory) behaviours which Hus et al. (2007) claim are negatively correlated with verbal and nonverbal IQ as well as severity of autism symptoms.

Sameness behaviours appear to affect cognitive flexibility in autism. Bos et al. (2019) reported decreased cognitive control in autistic participants specifically when presented with interest material; this association between cognitive control and sameness was not demonstrated for noninterest material.

Anxiety has been reported to be associated with sameness behaviours in autism. Factor, Condy, Farley and Scarpa (2016) identified an association between anxiety and sameness (but not other repetitive behaviours) in children with autism, with social motivation appearing to mediate between sameness and anxiety. Anxiety has been associated with Insistence on Sameness measures (i.e. routines, rituals, hoarding, dislike of change) but not with the measure of repetitive motor behaviours (Rodgers et al., 2012; Stratis & Lecavalier, 2013), which was not explained by age or language level (Lidstone et al., 2014). Other evidence also indicates a potential mediating role of effort control between sameness and anxiety in autism (Uljarevíc, Richdale, Evans, Ying Chai & Leekam, 2017). These mediators are perhaps critical, as other research has

failed to identify a significant link between sameness and anxiety in autism (e.g. Mason, 2018).

Further differences between sameness and repetitive motor behaviours have also been reported in the effect of sensory stimulation on their onset. Wigham et al. (2015) identified a potential pathway from under-responsiveness to sensory stimuli and all repetitive behaviours, whilst sensory over-responsiveness was identified to be specific to Sameness but not repetitive motor behaviours. It was also suggested Intolerance of Uncertainty and anxiety appear to mediate the path to Insistence on Sameness through both under- and over-responsiveness.

Consistent with being a spectrum disorder, autistic symptoms are recognised to be heterogeneous in autism. Using various methods of statistical analyses to "test for categorical versus continuous variation" of various traits, Ingram, Takahashi and Miles' (2008) concluded their data seemed to demonstrate sameness (as well repetitive sensory motor behaviours and language acquisition) fits a dimensional approach, with subgroups being based on social interaction/communication, intelligence and physical phenotype.

4.3.2.3. Stress and anxiety.

Stress, but not executive functioning, has been reported to be a mediator between autistic traits and repetitive behaviours (García-Villamisar & Rojahn, 2015). However, anxiety, a prevalent feature of autism, appears to have a complex relationship with repetitive behaviours in the disorder. Rodgers, Glod, Connolly and McConachie (2012), for example, found a differential moderating effect of anxiety in high-anxious and low-

anxious children with autism. Repetitive behaviours were reported to be more prevalent in children with high-anxiety, whilst also associated specifically with Insistence on Sameness traits (and not with simple motor repetitions). Conversely, in autistic children with low-anxiety, measures of anxiety were related to repetitive motor-sensory behaviours. It would appear Insistence on Sameness would appear to be specifically related to anxiety and autism.

4.3.2.4. Obsessions and compulsions.

Whether obsessions and compulsions in autism should be viewed within the consideration of comorbid OCD disorder, or as potential distinct repetitive phenomenology, they should be included within this review to attempt to portray a comprehensive assessment of repetitive behaviours in autism. There is some validity in this approach, since compulsions, for example, have been categorised as a higher-order repetitive behaviours in autism, along with rituals, sameness and restricted behaviours using the Repetitive Behaviour Scale-Revised (Georgiades et al., 2010). It would also be in line with Fischer-Terworth and Probst's (2009) conclusion from a systematic review on OCD traits and repetitive behaviours in autism, which proposed a subtype of OCD "Autism-related obsessive-compulsive phenomena". In this subtype, the researchers argue, repetitive behaviours in autism appear to be ego-syntonic and comorbid autism and OCD traits ego-dystonic, based on a lack of distress in the former, and associated distress in the latter. It has been suggested, whilst individuals with autism may exhibit less severe obsessions and compulsions, they may regard them as equally inappropriate and intrusive as individuals with Obsessive Compulsive Disorder (Cath et al., 2008).

4.3.2.5. Compulsions.

Compulsions are categorised as a feature within repetitive behaviours. For example, the Repetitive Behaviour Scale-Revised (RBS-R; Bodfish, Symons & Lewis, 1999), a commonly utilised tool used to measure repetitive behaviours in autism, includes six items to measure compulsive behaviours. However, there is little research directly on compulsions in autism, with much research comparing with compulsivity in OCD. Neuroanatomically, compulsions and rituals (as well as difficulties with minor change and complex motor mannerisms) have been identified to potentially be a result of abnormal functional relationships between the caudate and other brain areas (Sears et al., 1999).

Compulsions measured by the RBS-R have recently identified compulsive, ritualistic and sameness behaviours to be negatively correlated with an emotional false-memory task (Solomon et al., 2019). Gender differences within the autism spectrum have been identified too, with increased compulsive, sameness and restricted repetitive behaviours indicated in females (Antezana et al., 2019). Whilst Boyd, McBee, Holtzclaw, Baranek and Bodfish (2009) no evidence of a relationship between sensory processing difficulties and executive dysfunction in autism, they did report sensory processing issues to be correlated with compulsions as well as stereotypies in autism.

Relating to OCD compulsions in autism, clinical levels of OCD in high-functioning children with autism may be over 50% and whilst individuals with OCD may demonstrate more obsessions, some measures of compulsions have been reported to be comparable in OCD and autism (Deramus, 2009). However, compulsions in autism may be distinct to OCD compulsions. Wu et al. (2012) reported poor consistency for

the compulsion severity subscale using the Children's Yale-Brown Obsessive Compulsive Scale. This suggests compulsive repetitive behaviours displayed by individuals with autism are non-OCD repetitive behaviours (e.g. not ego-dystonic), and perhaps compulsive reports are separate to OCD comorbidity.

4.3.2.6. Obsessions.

The Children's Yale-Brown Obsessive Compulsive Scale has been reported to demonstrate a four-factor solution in children with autism, consisting of obsessions, higher-order repetitive behaviours, lower-order repetitive behaviours, and hoarding (Anagnostou et al., 2011). Obsessions in autism may be more heterogeneous than comparable traits in OCD (Zandt, Prior & Kyrios, 2007). The researchers identified many categories of obsessions appear to be more highly prevalent in OCD than in autism, but individuals with autism appear to display significantly more miscellaneous obsessions.

Whilst there is evidence of increased OCD traits in autism (e.g. Anholt et al., 2010; Bejerot et al., 2001; Hutton, Goode, Murphy, Le Couteur, & Rutter, 2008; Hollander et al., 2003; Ivarsson & Melin, 2008; Russell et al., 2005), there is also evidence obsessions in autism are distinct to those in OCD. As the cognitive-behavioural theory of OCD claims the individual's sense of responsibility for causing/preventing harm is central, Ekman and Hiltunen (2018) found individuals with autism to demonstrate comparable measures of responsibility belief (obsessions) to controls, and significantly lower than OCD participants. It appears, therefore, obsessions in autism is distinct to obsessions in OCD and perhaps, as the authors claim, related to specific thinking in autism, which seems to be systematic (Baron-Cohen, 2008) and predominantly non-

social (Baron-Cohen & Wheelwright, 1999). Measures of obsessions in children with autism have indicated obsessions generally based in non-social "folk physics" (Baron-Cohen & Wheelwright, 1999).

It has been suggested there is a causal relationship between "compulsive/ritualistic behaviour" in autism and "the urge to repeatedly do things" in OCD. Ruzzano, Borsboom and Geurts (2015) reported "concern for dirt", "continual washing" and "thoughts due to a higher power" were not connected in autism. Whilst the perceived connection between continual counting and repeating as a compulsive behaviour was not upheld by the empirical data, the causal relationship between the autism symptom "compulsive/ritualistic behaviour" and the OCD symptom "the urge to repeatedly do things" was supported by clinical evidence, with "causal exploration [suggesting] that sensory interests may actually lead to OCD type compulsion continual checking" (Ruzzano et al. 2015).

4.3.3. Biological basis of repetitive behaviours in autism.

Review articles have highlighted inconsistencies between studies in accurately detecting regional brain differences in autism. Widespread volumetric differences have been reported in the brain of individuals with autism including the cerebellum, amygdala, corpus callosum, frontal, temporal and parietal lobes, thalamus and the brainstem (Anagnostou & Taylor, 2011). Widespread areas of the brain have been reported to demonstrate abnormal cortical thickness, in correlation with severity of repetitive behaviours in autism (Zabihi et al., 2019). Increased volumes have been consistently reported in total brain, cerebral hemispheres, caudate nucleus and decreased size of corpus callosum size, midbrain and cerebellar vermal lobules

(Stanfield et al., 2008). A review of neural circuitry in autism by Belger, Carpenter, Yuvel, Cleary and Donkers (2011) indicated the effect of two distinct brain regions significant to the disorder, with neural sites associated with repetitive behaviours include the anterior cingulate cortex, dorsolateral prefrontal cortex, caudate nucleus and the dorsal striatum. A review of 206 imaging studies by Pina-Camacho et al. (2011) reported that repetitive behaviours are associated with fronto-striato-cerebellar dysfunction.

Findings of neural dysfunction across various parts of the autistic brain supports the hypothesis that autism is largely a dysfunction of connectivity (e.g. Minshew & Keller, 2010). Functional imaging research has indicated the role of the cerebellum in the multiple functions of executive control, language and motor behaviour (Penn, 2006). It may be, therefore, a decisive neural region in restricted and repetitive behaviours. Repetitive behaviours may also be associated with the cerebellum in autism (Cheung et al., 2009; Penn, 2006). There is also evidence of neuroanatomical structural abnormalities in autism in the caudate nuclei, multiple frontal and temporal regions as well as the cerebellum (Rojas et al., 2006).

However, there has also been widespread neurological dysfunction linked to repetitive behaviours in autism. This evidence may not only emphasise Minshew and Keller's (2010) view of general function of connectivity, but also indicate the heterogeneous forms of repetitive traits in autism. Associations between weak connectivity in a posterior cingulate cortex network and repetitive behaviours in autism have been described (Weng et al., 2009). Functional over-connectivity between the left visual primary cortex and the right IFG and the pars orbitalis in autism has also been

associated with increased repetitive behaviours (Traynor et al., 2018). Increased severity of repetitive behaviours has been linked to stronger functional connectivity in the posterior cingulate cortex and the right parahippocampal gyrus in autism (Monk et al., 2009). However, the role of functional connectivity in the production of repetitive behaviours is complex and unclear. A recent demonstration of this is evidence of widespread neural site dysfunctional connectivity in the autistic brain between participants with high- and low-repetitive behaviours, whilst autistic participants with low-repetitive behaviours specifically demonstrate a variable pattern of both over- and under-connectivity (Noriega, 2019).

The amygdala is typically associated with the processing of social and emotional information. However, whilst Dziobek, Fleck, Rogers, Wolf and Conit (2006) reported a lack of a mediating effect of this neural area in autism, a negative correlation between amygdala volume and repetitive traits was reported, indicating its significance to these repetitive behaviours in the disorder. It is plausible therefore, some repetitive behaviours may be a self-stimulating response to influence of negative emotion or difficulties in social processing.

Measures of the autonomic nervous system have identified heart functionality – proposed to be more impaired functioning under stressful situations – appears to be correlated to repetitive behaviours in children with autism (Condy, Scarpa & Friedman, 2017). Whilst this link is correlational, it provides insight into the role of stress and the onset of repetitive behaviours in autism. The possibility some repetitive behaviours may be a consequence of poor self-monitoring in autism is backed up by evidence of a

relationship between such traits and dysfunction in the anterior cingulate cortex – a neural site implicated in response monitoring (Thakkar et al., 2008).

In summary, there is widespread evidence of associations between disordered neuroanatomical sites and repetitive behaviours in autism. However, the specificity in neural region is perhaps a consequence of plasticity (Traynor et al., 2018) and may provide limited insight alone into the nature of repetitive behaviours in the disorder. It is important to acknowledge the widespread neurological dysfunction in autism (e.g. Minshew & Keller, 2010). A lack of clarity over the relationship between neural functioning and repetitive behaviours is compounded by the heterogeneous nature of repetitive behaviours.

4.3.4. Summary.

There is some evidence of a specific pattern related to the function and nature of repetitive behaviours in autism. Whilst it appears the effect of variables such as sensory issues and anxiety is yet to be fully understood, they appear significant to the onset of repetitive behaviours. There is consistent evidence of a lower-level and higher-level two-factor solution. It would appear only higher-level repetitive behaviours, such as insistence on sameness, appear to be distinct to autism. It would be crucial, therefore, to understand the neuropsychological evidence related to repetitive behaviours in autism to better understand the phenomenology.

4.4. Causes of Repetitive Traits in Autism

There is a wealth of evidence reporting unique and general deficits as well as processing styles, which together appear to result in a pattern of neuropsychological

functioning resulting in autistic symptoms. As repetitive behaviours are so significant (diagnostically) in autism (APA, 2013), understanding the neuropsychological functioning me be critical. Two major lines of enquiry to this extent are context processing abilities, and executive functioning.

4.4.1. Context processing.

The Weak Central Coherence theory has also been associated with certain repetitive behaviours in autism. This theory indicates a preference for segmental over holistic information processing in autism appears to create a tendency towards certain higher-order repetitive traits, such as narrow/restricted interests (Frith, 1997; Gomot &Wicker, 2012).

Weak central coherence has been proposed to explain the unique cognitive-perceptual processing style in autism. Central cohesion is the typical processing style whereby individuals have a 'tendency to process incoming information in its context' (Hill, 2004a). Drawing on a body of evidence, researchers have noted that the processing abilities of individuals with autism appear to have a 'bias' for processing local, surface information over the more global and semantic form (Happé & Frith, 2006). Laboratory studies such as Block Design (e.g. Shah & Frith, 1993) and Embedded Figure tests (e.g. Ropar & Mitchell, 2001) strongly support this account. Local processing bias (also regarded as stimulus over-selectivity) may result from either a problem in sensory integration or from attentional difficulties in selecting relevant cue. Whilst it is not unique to autism, or even present in all individuals with autism, it is highly prevalent in the disorder and central to other cognitive and behavioural abnormalities (Ploog, 2010).

Although some experiments have failed to find evidence to support the weak central coherence hypothesis in high-functioning autism (e.g. Burnette et al., 2005; López, Leekam & Arts, 2008), a more fundamental issue with the theory is its inability to explain many of features of low-level repetitive behaviours, such as stereotyped movements and echolalia (Turner, 1999). This would seem to suggest that the aetiology of repetitive behaviours is multi-faceted.

4.4.2. Executive functions.

Executive functions are specific psychological mechanisms used to guide actions. These mechanisms are rooted in the prefrontal structures of the brain, particularly the prefrontal cortex, and they typically control actions through functions such as planning, working memory and attention (Hill, 2004b). The neuropsychological profile has been shown in autism to be complex and varied. Certain functions are impaired and others are intact, as can be seen in the "spiky profile" shown in tests of intelligence (e.g. Happé, 1994).

Evidence from the study of executive dysfunction has identified a number of mechanisms which may contribute to the phenomenology of repetitive behaviours in autism (Frith, 1997). Turner (1999), for example, explains how problems in self-regulation can leave individuals with "little option but to carry out the same behaviour over and over again." It is possible the specific profile of executive dysfunction in autism may influence repetitive behaviours.

4.4.3. Mental flexibility.

Inflexibility, characteristic of autism through insistence on sameness and resistance to change, are of great significance to compulsive behaviours. Compulsive behaviours have been described to become pathological based on factors such as interference, resistance and control (see Scahill et al., 1997). Cognitive flexibility has been reported to be significant in autism. Inhibitory control and attentional flexibility, impaired in autism, have been reported to be associated to repetitive behaviours in high-functioning children with autism (Mostert-Kerckhoffs, Staal, Houben, & de Jonge, 2015).

Research using set shifting tasks has indicated a specific cognitive dysfunction in autism. Hughes et al. (1994) reported errors made by the individuals with autism were distinctly distributed in early trials on the rule-shift, suggesting a difficulty in transferring knowledge to a new task. It was identified what was unique to autism was not perseveration generally, but the fact that individuals with autism became 'stuck-in-set'. Although some researchers have failed to confirm the above deficits in set-shifting tasks in children with autism (e.g. Goldberg et al., 2005), Yerys et al. (2009) believed that their study addressed this issue, showing the effect in younger participants than with high-functioning autism. Intriguingly, some research has reported the deficit to be present in high-functioning autism but not in Asperger syndrome (Rinehart et al., 2001). Although the etiological significance of this information is still little understood, results from these studies are still intriguing: Yerys et al. (2009) report a positive correlation between set-shifting deficits and frequency of repetitive behaviours. Furthermore, evidence of a relationship between difficulty inhibiting interfering information and cognitive shifting abilities, with increased higher-order repetitive

behaviours in autistic children (Faja & Darling, 2018) indicates the importance of mental flexibility in protection against higher-level repetitive behaviours in the disorder.

It is perhaps crucial to note metal inflexibility – as per the failure to maintain set – in autism have been demonstrated to be associated to difficulties in social interaction, which Varanda and Fernando (2015) report is not a failure to shift attention, but difficulties arising because of constantly shifting attention in social interactions. Similarly, mentalizing tasks, whereby an individual requires the ability to understand the mental state of others, have been argued to be central to social communication deficits in autism (Hill & Frith, 2003). However, Jones et al. (2018) recently reported Theory of Mind – a commonly reported mentalizing ability – to be associated to repetitive behaviours in a sample of adolescents with autism. Whilst the correlational relationship is unclear, it is possible some repetitive traits may be a consequence of the effect the confusing social world has on the individual (Carruthers, 1996). Other theorists have not been convinced of the link between mentalizing and repetitive traits, suggesting not only that they do not adequately explain either many low-level repetitive behaviours, restricted interests or sameness, but they also do not appear to occur more often in social situations as the theory would predict (Levy, 2007).

4.4.4. Perseveration.

Perseveration, the "inability to release attention from a perceptual dimension" (Stahl & Pry, 2005), appears to be central to the nature of repetitive behaviours in autism. Perseveration has been reported to be as significant in repetitive speech as repetitive behaviours in autism (de Villiers, Fine, Ginsberg, Vaccarella & Szatmari, 2007),

although it may be a feature more specifically in early development in autism, as it is also in typical development (Elison et al., 2012; Sasson et al., 2008).

Research in mental inflexibility in autism has highlighted the nature of perseverative thought and behaviour. Individuals with autism have been reported to have difficulty in flexibility shifting attention during daily activities (Gioia, Isquith, Kenworthy, & Barton, 2002). This has been studied empirically through set-shifting i.e. the ability to move from one cognitive set to another such adapting to a changing rule. Stahl and Pry (2005) showed that a strong correlation between set-shifting and joint attention is present in typical two year olds, but absent in their autistic counterparts.

Perseverative errors (the "inability to modify an ineffective behaviour"; Adrien et al., 1995) have been reported as a feature in autism when undertaking executive function tests. Children with autism have been shown to make perseverative errors even when they are aware that they are following an incorrect strategy (Shu, Lung, Tien & Chen, 2001). Adrien et al. (1995) specifically found perseverative errors in children with autism to occur when tasks become more abstract on object permanence task. Ciesielski and Harris (1997) identified perseverative errors in autistic participants undertaking five executive functioning tests of selective inhibition/switching abilities appeared to be a result of the failure to either "selectively inhibit or disengage from a previous mental set". The researchers reported participants with autism would become stuck-in-set, particularly when it is not explicit what to attend to or what to disengage from.

Broadbent and Stokes (2013) found autistic participants to perform significantly better (fewer perseverative errors) when negative feedback was absent during a Wisconsin Cart Sorting Task (WCST) than when it was present (compared to control participants), suggesting they do not use negative feedback in the same way as typically developing individuals. Memari et al. (2013) found no effect of age on perseverative errors (on a WCST task) but gender was associated with performance: girls demonstrated worse performance than boys (although there was a relatively small sample size of girls). Furthermore, perseveration in the children with autism was negatively correlated with "appropriate daily social play" and the amount of sleep children reported. Perseverative errors in autism, under WCST tasks, appear to be specifically a consequence of difficulties in task-switching, as opposed to other cognitive difficulties (Van Eylen et al., 2011).

4.4.5. Inhibition.

Tests of inhibitory control have often produced mixed results in individuals with autism. A number of researchers have found individuals with autism to show comparable or even increased performance compared to typically developing individuals (Adams & Jarrold, 2009; Ozonoff & Strayer, 1997). Russo et al. (2007) suggest "when inhibition is strictly defined and when participants have developmental levels greater than 6 years" inhibition seems to be intact in autism. However, using a range of different measures and controls other researchers have reported evidence of deficits in inhibitory control in autism (e.g. Christ, Holt, White & Green, 2007; Mosconi et al., 2009), whilst other research has found impaired tests of inhibition that were not specific to autism but matched to other language impaired groups (Bishop & Norbury, 2005). However, a study by Zandt, Prior & Kyrios (2009) reported the only significant

association between repetitive behaviours and executive dysfunctions in autism were between inhibition and obsessions.

Overall, evidence shows a pattern of autism specific inhibitory control to be generally intact, but under certain conditions it appears to be increased. Following a finding by Mosconi et al. (2009) that inhibitory errors appear to be related to higher-level but not lower-level repetitive behaviours, it is has been indicated to be important to continue to ask "when and how" rather than "if" inhibition affects individuals with autism differentially.

4.4.6. Summary.

Evidence of links between executive functioning and repetitive behaviours is mixed (see Brunsdon & Happé, 2004, for review). Whilst Zandt, Prior and Kyrios (2009) found only weak correlations between executive functioning variables and repetitive behaviours, Lopez et al. (2005) reported repetitive behaviours in autism may be linked to cognitive flexibility, working memory and response inhibition deficits. Evidence such as South, Ozonoff & McMahon's (2007) emphasise the significance of the link between repetitive behaviours and set-shifting deficits in autism. Jones et al. (2017) has even reported no relationship between measures of repetitive behaviour and executive functioning, whilst surprisingly finding Theory of Mind to be associated with repetitive behaviours in autism. Theory of Mind is a social-processing construct, whereby an individual is to infer the knowledge of another person. If this relationship is accurate, certain repetitive behaviours may be a response to efforts to process difficult information in the social world, and social processing may be a critical moderator.

Crucially, Brunson and Happé (2004) highlight there is no single task which definitively measures executive functioning, which may explain the mixed evidence. This is just as important to note as the heterogeneity (and likely multiple pathway) of repetitive traits in autism. It is likely, therefore, distinct neuropsychological functions describe a small part of a wider cognitive picture. Future work requires the interactions between executive mechanisms in autism to be realised within a pathway model. This model will likely need to incorporate evidence of sensory abnormalities, social deficits, anxiety and mood in autism in parallel with cognitive dysfunctions.

4.5. Mood and Repetitive Traits: Ego-dystonic and Ego-syntonic Origins of Repetitive Traits.

An important feature of repetitive traits appears to be whether they are ego-dystonic or ego-syntonic. A trait is considered ego-dystonic if it is inconsistent with an individual's sense of self; it opposes how an individual defines themselves. As Purdon, Cripps, Faull, Joseph and Rowa (2007, p.200) defines, ego-dystonic thoughts are those "occurring outside of the context of one's morals, attitudes, beliefs, preferences, past behaviour and/or one's expectations". OCD is defined by its ego-dystonic nature: obsessive thoughts are incompatible with the individual's sense of self, being unwanted, undesirable or even repulsive to them. For example, an individual with a checking compulsion does not want to complete the act, they are compelled to do so out of fear of some negative consequence. Accordingly, these traits are likely to be associated with negative mood, such as distress and anxiety.

Contrastingly, traits are ego-syntonic when they are consistent with the individual's sense of self; or at least they are not-inconsistent with their sense of self. As Belloch,

Roncero and Perpiñá (2012, p. 95) describe, ego-syntonic traits as being "consistent with the person's values, personality structure, explicit feelings and/or desired self-view". Consequently, these traits are likely to be thought of as important, necessary or desirable to the individual. Such traits evoke feelings of pleasure, arousal and/or desire. Accordingly, these traits are likely to be associated with measures of positive (or at least neutral) mood. A clinical example is impulsive behaviour, such as pathological gambling. In this example, the individual seeks out the behaviour through some sense of enjoyment. Historically, repetitive traits within autism have been typified by an ego-syntonic nature (e.g. Baron-Cohen, 1989). "Repetitive Behaviours" demonstrated in individuals with autism (e.g. Bodfish, Symons, Parker, & Lewis, 2000; Leekam et al., 2007; Moss, Oliver, Arron, Burbidge, & Berg, 2009) are so relevant to the disorder – arguably important, necessary or desirable – a diagnosis is dependent on their presence (APA, 2013). Despite these implications, recent evidence indicates Repetitive Behaviours in autism may be more ego-dystonic than originally thought (see review by Barber, 2015).

Phenomenologically, distinguishing between ego-dystonic and ego-syntonic repetitive traits may be fundamental to understand overlap (or distinction) between OCD and autistic symptomology. As negative consequences and/or mood may be indirect (such as the shame or disappointment of losing money), care must be taken to interpret mood at the origin (not the consequence) of the trait, otherwise we may falsely attribute an ego-dystonic function.

For many decades, repetitive traits in autism have been generally assumed to be of an ego-syntonic origin, being thought of as pleasurable and self-stimulating (e.g.

Carruthers, 1996; Colman, Frankel, Ritvo, & Freeman, 1976; Hutt, Hutt, Lee, & Ounsted, 1964; Frankel, Freeman, Ritvo, & Pardo, 1978; Freeman Frankel, & Ritvo, 1976). Repetitive traits are so fundamental to autism, as a heterogeneous array of symptoms they are one of two diagnostic criteria, with no indication of associated mood. To emphasise this point, when Scahill et al. (2006, p. 1116) modified an Obsessive-Compulsive measurement tool for use in autistic samples, to expand the compulsions checklist they added "behaviours commonly seen in children with [autism]" such as "repetitive water play". This traditional view of solely ego-syntonic autistic repetitive traits is being challenged. For example, Barber's (2015, p. 9) systematic review claims "Repetitive Behaviours in [autism] may not be ego-syntonic and may cause as much distress as those similar Repetitive Behaviours seen in OCD". Identifying ego-dystonic traits in autism would challenge the traditional view that repetitive traits in autism are an inherent part of the self (e.g. Baron-Cohen, 1989).

As there is very little direct evidence to distinguish traits across OCD and autism (Cath et al., 2008; Fischer-Terworth & Probst, 2009), comparing these disorders within a shared framework may provide insight into similarities and differences between them. Whilst evidence of exclusively ego-syntonic repetitive traits in autism would render a comparison between the disorders clinically meaningless, the presence of some ego-dystonic traits in autism and/or ego-syntonic traits in OCD would demonstrate a validity in comparing autism and OCD. In line with the above definitions of each of these traits, ego-dystonic traits would be expected to be associated with (direct) measures of negative affect, whereas ego-syntonic traits would be related to either positive or neutral affect.

4.5.1. Ego-dystonic and ego-syntonic repetitive traits in autism.

Whilst Repetitive Behaviours have typically been viewed as part of the autistic phenomenology, more recent investigations have suggested both ego-syntonic and ego-dystonic factors may be associated with repetitive traits in the disorder (Barber, 2015). Rice (2009) suggests distress and pleasure-seeking may be differential motivators for these traits in autism.

Below is a comparison within empirical literature of ego-dystonic and ego-syntonic features of repetitive traits in autism. There are, however, very few studies which directly investigate whether an individual believes specific repetitive traits to be either consistent or inconsistent with their sense of self (Cath et al., 2008; Rice, 2009). Therefore, the available evidence of the functions of Repetitive Behaviours can only be placed within an ego-dystonic versus ego-syntonic framework based on theoretical scrutiny: this categorising does not definitively or exclusively place these traits as ego-syntonic or ego-dystonic, but instead provides evidence for a Compulsive and Repetitive Traits framework.

4.5.1.1. Evidence of ego-dystonic repetitive traits in autism.

Ego-dystonic traits are much easier to identify than ego-syntonic traits. If a measure of distress is present, then it is possible the trait is ego-dystonic (not consistent with the sense of self; unwanted). As previously described, this distress must be directly related to the origins of the trait. Anxiety has been demonstrated to increase repetitive traits in neurotypical individuals (e.g. Lang, Krátký, Shaver, Jerotijević, & Xygalatas, 2015) and a similar association has been described in autism (Rodgers, Riby, Janes, Connolly, & McConachie, 2012). The relationship between Repetitive Behaviours and autism is still

unclear (e.g. Deramus, 2009; Factor, Condy, Farley, & Scarpa, 2016), with Cadman et al. (2015) suggesting "little is known about the symptom profile of OCD in individuals who have autism". However, there is an emerging picture of the role of anxiety as a mediating variable to Repetitive Behaviours in autism, potentially in the same manner they are in OCD.

Statistical modelling and mediating analyses have provided some clarity, whilst also highlighting this complex relationship. Deramus's (2009) mediation analysis suggests anxiety is fully mediated through Repetitive Behaviours, leading to social problems in children with autism. This indicates Repetitive Behaviours are primary to social problems, with a pathway leading from anxiety to social difficulties through Repetitive Behaviours. It also appears lower- and higher-order repetitive traits may have a differential relationship with anxiety in autism (Bourreau et al., 2009; Rodgers et al., 2012) and Insistence on Sameness (a higher-order, i.e. more cognitively complex) Repetitive Behaviours have been specifically implicated (Factor et al., 2016; Lidstone et, 2014). However, this association may only be in high anxiety individuals, whilst sensory motor (a measure of lower-order, i.e. less cognitively complex) Repetitive Behaviours appear to be associated to anxiety in low-anxiety individuals with autism (Rodgers et al., 2012).

The relationship between anxiety and insistence on sameness is theoretically significant and there are many theories to support this association (e.g. Carruthers, 1996; Hutt et al., 1964). Baron-Cohen's (2008) hypersystemizing theory of autism proposes autistic individuals are driven by a desire to keep the world around them predictable (possibly because to them it is not). This may produce a sense of anxiety,

which the individual attempts to control by engaging in Repetitive Behaviours. Similarly, it may reflect why the need for consistent routines (Marquenie, Rodger, Mangohig, & Cronin, 2011; Turner, 1999), as well as clear and consistent environments (Tutt, Powell, & Thornton, 2006), is often central to autism.

To further complicate the model, abnormal sensory processing in autism (Tomchek & Dunn, 2007) has been demonstrated to be correlated with increased Repetitive Behaviours (e.g. Boyd, McBee, Holtzdaw, Baranek, & Bodfish, 2009; Chen, Rodgers, & McConachie, 2009; Colman et al., 1976; Freeman et al., 1976). Pathway models have begun to clarify this relationship. Wigham et al. (2015) claim sensory processing appears to have a differential role in mediating between anxiety and Repetitive Behaviours in children with autism: there appears to be a direct path from sensory under- and over-responsiveness to Insistence on Sameness, whilst there may be only a direct path to repetitive motor behaviours through sensory under-responsiveness. Furthermore, Wigham et al. (2015) suggest Intolerance of Uncertainty appears to mediate a link from sensory responsiveness, through anxiety, to Insistence on Sameness.

In summary, there is strong evidence of a relationship between measures of distress (specifically anxiety) and Repetitive Behaviours in autism. This pathway, however, is complex and it is uncertain whether this causal relationship is truly ego-dystonic (unwanted and unpleasant), particularly as negative affect may be secondary to factors such as social and sensory difficulties. The evidence of ego-syntonic traits would provide an indication of the structure of the CaRT framework.

4.5.1.2. Evidence of ego-syntonic repetitive traits in autism.

It is difficult to determine ego-syntonic Repetitive Behaviours in autism. Ego-syntonic means the individual regards the symptom as being consistent with their sense of self i.e. it is not unwanted. However, for the same reason in which Baron-Cohen (1989) suggests ego-dystonic traits may be irrelevant to at least some individuals with autism, ego-syntonic traits may be equally difficult to measure as they require a degree of introspection, which may be lacking in the disorder (Marris, 1999).

There is some evidence of ego-syntonic functions of Repetitive Behaviours in autism. For example, Lehnhardt et al. (2013) claim individuals with autism generally perceive their Repetitive Behaviours to be reasonable and appropriate. Absence of anxiety or distress alongside a repetitive trait may be a reasonable marker for ego-syntonic origins, and Ruta, Mugno, D'Arrigo, Vitiello and Mazzone (2010) propose individuals with autism do not demonstrate distress in response to repetitive traits. However, as Repetitive Behaviours in autism are heterogeneous (DSM-5, APA, 2013; Turner, 1999), an understanding of the functions across these diverse traits is necessary.

The clearest case for an ego-syntonic repetitive trait in autism is circumscribed interests, which are strong interests in a very specific topic. Interests range from intense and focussed hobbies or topics (as narrow ranging as interest in washing machine models or bottle tops) to strong attachments to objects. Whilst other Repetitive Behaviours in autism tend to decrease in severity after a peak earlier in life, circumscribed interests appear to be relatively stable throughout life (South, Ozonoff, & McMahon, 2005). They appear to be phenomenologically distinct from other Repetitive Behaviours (Caldwell-Harris & Jordan, 2014), and evidence of their

significance to autism (Bartak & Rutter, 1976; Lam, Bodfish & Piven 2008) is perhaps reflective of the cognitive processing style in autism (e.g. Baron-Cohen, 2008). This suggestion is supported by the evidence that special interests occur regardless of stimulation and/or anxiety (Turner, 1999).

It is much more difficult to conclude whether a trait is ego-syntonic and there are very few studies which attempt to directly assess whether repetitive traits in autism are wanted (e.g. Cath et al., 2008; Rice, 2009). Although ego-syntonic traits are likely to not be associated directly with distress, two main difficulties confound identification. Firstly, absence of evidence is not evidence of absence: these traits may be associated with measures of mood, but the investigations may have failed to measure it at the appropriate level. Secondly, as emphasised in the above section, evidence of distress does not preclude the trait as being ego-syntonic, particularly if the distress is associated indirectly.

4.6. Summary: Repetitive Traits between Obsessive Compulsive Disorder and Autism

Two key issues have been identified in the above literature. Firstly, it seems little is understood about the symptom profile of OCD for individuals with autism (Cadman et al., 2015). Whilst there seems to be overlapping symptomology between OCD and autism (Fischer-Terworth & Probst, 2009), it appears further investigation is needed to delineate Repetitive Behaviours, a core feature of autism, from OCD traits. Secondly, whilst ego-dystonic and ego-syntonic principles appear fundamental to distinguishing between repetitive traits in OCD and other disorders, direct measures of ego-dystonic and ego-syntonic properties of Repetitive Behaviours are, presently, not validated for clinical groups (e.g. Cath et al., 2008; Rice, 2009).

Traditionally, Repetitive Behaviours in autism have been viewed through an ego-syntonic approach (consistent with the individual's sense of self). They have been recognised as a fundamental part of the disorder (Baron-Cohen, 1989) to such an extent they are recognised as one of two major diagnostic criteria (DSM-5, APA 2013). However, the evidence presented in this chapter challenges this original (and simplistic) view, indicating it is relevant to continue to investigate the link between ego-syntonic and ego-dystonic repetitive traits in autism and OCD. The distinction between these appears to be linked to mood correlates, with ego-dystonic traits related to negative mood (distress) and ego-syntonic traits related to non-negative (positive or neutral) mood. In line with this line of enquiry, the following section will systematically review empirical evidence of mood and repetitive traits in adults with autism (the population of interest to the overall thesis aims).

Chapter 5. Systematic Literature Review: Repetitive Traits and Mood in Adults with Autism

5.1. Introduction

The previous sections provide the theoretical basis for a shared symptomological framework between OCD and autism. The relevance of such a framework is based on the increasing evidence of an aetiological overlap between OCD and autism (e.g. Anholt et al., 2010; Bejerot et al., 2001; Deramus, 2009; Hollander et al., 2003; Hutton et al., 2008; Ivarsson & Melin, 2008; Lehnhardt, 2013; Russell et al., 2005) and is likely to give further insight into how comparable (and how different) OCD and autism are. The CaRT framework combines the knowledge of two different types of repetitive traits. The first type of repetitive traits are obsessions and compulsions, clinically defined as OCD symptoms. The second type of repetitive traits are those referred to simply as "Repetitive Behaviours", originating from studies of autism and intellectual disabilities (e.g. Bodfish et al., 2000; Leekam et al., 2007; Moss et al., 2009).

As described in Chapter 4, a major distinction between these two types of traits may lie in their ego-dystonic and ego-syntonic origins. OCD symptoms are ego-dystonic, meaning they are inconsistent with the individual's sense of self, unwanted and are therefore associated with distress. In OCD, for example, an individual would not want to complete activities such as repetitive hand-washing, but is compelled to alleviate distress caused by obsessive thoughts (e.g. "if I don't do this then something bad will happen"). However, ego-syntonic traits are those which the individual doesn't oppose to undertaking, because to some extent they derive (directly) pleasure from them. Impulsive traits are often clinically regarded as ego-syntonic. For example, a pyromaniac enjoys starting fires (even if later – indirect – consequences are negative).

If these premises are correct, then mood measured directly at the onset of the traits may reflect whether different repetitive traits have ego-dystonic or ego-syntonic origins.

5.2. Rationale

Most of the evidence presented so far relates to studies involving autistic children and adolescent participant groups. As there is a distinct lack of knowledge of how autism is presented in adulthood (Happé & Charlton, 2012), evidence from studies of autistic children can inform our hypotheses about what is likely in an adult autistic population. However, these hypotheses must be tested.

For repetitive traits, the DMS-5 (APA, 2013) does not regard them as diagnostically necessary in autistic adults. However, the implication repetitive traits are not valid in adulthood in the disorder is upheld through various studies (e.g. Chowdhury et al., 2010; Esbensen et al., 2009; Fecteau et al., 2003; Howlin et al., 2000; Lord et al., 2015; Seltzer et al., 2003). It is plausible repetitive traits may be phenomenologically relevant in this population, but in later life adults may conform to social norms and mask such behaviours. As it has been reported the function and phenomenology of repetitive traits in adults with autism is unclear (Deramus, 2009; Factor et al., 2016), and evidence from childhood studies can only inform our hypotheses, this systematic review attempts to address this issue by using adults with autism as the target population.

In line with the proposed shared OCD and autism symptomological framework, a major distinction which needs to be made is between ego-dystonic and ego-syntonic traits.

Ego-dystonic traits are likely to be associated with measures of negative affect (including distress, anxiety or depression), and ego-syntonic traits are likely to be associated with positive affect, or at the very least the absence of negative mood. Succinctly, the following research question was generated to systematically review existing evidence to attempt to identify and understand what the relationship is between affect and repetitive traits in adults with autism. A systematic search was designed in line with this, to combine scientific evidence of repetitive traits in adults with autism when reported with measures of mood.

5.3. Method

5.3.1. Research aim and questions.

The general aim of the systematic literature review was to identify the type and function of the range of repetitive traits in adults with autism. Specifically, the aim was to understand whether repetitive traits in autistic adults can be differentiated by ego-dystonic and ego-syntonic origins. If there is a lack of directly studying these ego-origins, the presence of a measure of mood may be most relevant to identify a large enough evidence based for this research.

The research questions related to this study were:

1. What are the repetitive traits displayed by adults with autism, and how are they related to measure of mood?

5.3.2. Search strategy.

To yield as much potential evidence within the field of clinical psychology as possible, articles were retrieved using EBSCO (PsycINFO, PsycARTICLES, MEDLINE and CINAHL

Complete) for published articles, as described in section 5.3.6. To include as much relevant evidence as possible, there were no year limits in place. Due to differences in the set-up of each database, the limiters for each of the separate search engines are described in Table 5.1. Grey literature (such as unpublished articles, research reports and doctoral dissertations) was searched through Open Grey (Europe) and ProQuest Dissertations and Theses A&I. The PRISMA (Moher et al., 2009) study selection diagrams are shown in separate figures for the main search and the grey literature search, as significant numbers of articles were identified in the grey literature (see Figures 5.1 & 5.2). Open Grey was searched using this method. For ProQuest, as the yield was very large, the three search terms described in Table 5.1 were limited to "abstract".

5.3.3. Selection strategy.

The abstracts of all yielded articles identified were screened for relevance by the lead researcher against the inclusion and exclusion criteria in 5.3.4. and 5.3.5. Ten articles were included in the final systematic review, as outlined in the study selection flow chart (Figures 5.1 and 5.2).

5.3.4. Inclusion criteria.

Articles were included in which the study made direct reference to: 1) assessing a Repetitive Behaviour or OCD trait; 2) some measure or indication of related mood; and 3) in adult participants with a diagnosis of autism. The aim of the current investigation is to understand Compulsive and Repetitive Traits in adults with only a diagnosis of autism; comorbidity with any other disorder may otherwise increase the chance of false positives.

5.3.5. Exclusion criteria.

Following the rationale outlined in the inclusion criteria, studies were excluded if they fell into the following criteria:

- Participants were below the age of 18 years (as they are not within the adult age range of the final empirical investigation);
- Animal studies;
- Participants without a sole diagnosis of autism, due to comorbidity issues (i.e. difficulty in attributing the repetitive traits specifically to autism);
- Participants with intellectual disabilities/learning disabilities, also due to comorbidity issues;
- Studies which were not relevant to the aims of the search i.e. did not directly or indirectly measure both Repetitive Behaviours and mood; or
- Single case studies, or studies involving fewer than 10 participants, due to reduced reliability of such evidence when generalising to the wider population.

5.3.6. Search terms.

Search terms in Table 5.1 were used, with the final search being completed on 7th August 2018.

Table 5.1. *Search terms used for main study systematic search using PsycINFO, PsycARTICLES, MEDLINE and CINAHL Complete.*

Any of the following terms for autism as "select a field"	Combined with "AND"	The following term for Repetitive Behaviour as "abstract"	Combined with "AND"	The following term for mood as "select a field"
autis* or Asperger* or "pervasive developmental disorder" or pdd		"repetitive behavi*" or obsess* or compuls* ritual* or stereotyp* or sameness or rigid* or persev* or want* or unwant*		ego* or pleasur* or mood* or anxi* or stress* or happ* or distress* or self* or arous* or sad* or like* or dislike* or enjoy* or depress* or feeling* or emotion* or attitude* or tolera* or gratif* or arous*

* indicates the word was truncated to allow for multiple word endings
Nb. Special limiters placed for PsycARTICLES: all articles for participants aged 18 years and over; human; exclude book reviews; exclude non-article content. Special limiters placed for CINAHL Complete: human; English language; all adult. Special limiters placed for MEDLINE: human; English language; all adult. Special limiters placed on PsycINFO: all articles for participants aged 18 years and over; English language; human.

5.4. Data Analysis and Synthesis

A narrative synthesis was used to report the results as this is argued to be the most appropriate to compare across the range of studies with a mix of methods, participants and different outcome measures (Popay et al., 2006). This approach is outlined in section 3.3.

Initially, six articles were yielded, four of which were from the grey literature search. To broaden the search criteria, all studies were included which involved some measure of a repetitive trait (including repetitive OCD symptoms) in addition to some measure of mood (even indirectly, such as irritability) in adults with autism. A lack of directly relevant studies required a few indirectly relevant investigations to be included. This involved the inclusion of seven pharmacological trials, which otherwise met the inclusion criteria. The final yield consisted of ten articles. As the range of empirical

evidence was varied, rather than adopting quantitative analysis, this led to the necessity of a narrative synthesis approach. This approach is guided by the recommendations by Aveyard (2010); the evidence is presented within themes.

Themes emerged through the following process: each study was read and reviewed; summaries were made of the relevant findings from each study; the key finding was summarised for each study, generating themes; closely associated themes were combined, so similar evidence could be compared in the narrative synthesis. The themes which emerged were: ego-syntonic assessments; ego-dystonic assessments; insistence on sameness; and abnormal sensory processing. One final theme was generated to combine the indirect evidence from pharmacological studies. Evidence from clinical trials, including open-label studies, has been included, even though the relevance to the comparison between mood and repetitive traits in autism is indirect; the limitations this is outlined below.

5.5. Methodological Quality Assessment

Various quality assessment tools were utilised, appropriate to the methodology of each study. Summaries of the critical analyses are presented in Tables 5.2 to 5.5 below, followed by the narrative synthesis of the systematic review. The quality of the non-pharmacological studies were appraised according to the criteria suggested by Aveyard (2010, p103), with the summaries presented in Table 5.2. The medical trials were appraised using different tools, again each specifically designed to assess the different methods used. For the qualitative studies, the Quality Appraisal Tool (Effective Public Health Practice Project, 2010) was used to assess the quality of the methods, as presented in Table 5.3. Randomised control trial studies were appraised using the

Critical Appraisal Skills Programme (2015) randomised control trials checklist; findings are presented in Table 5.4. For quantitative studies, the Quality Appraisal Tool (Effective Public Health Practice Project, 2010) was used to assess the quality of the methods, as presented in Table 5.5.

Table 5.2. *Critical appraisal questions by Aveyard (2010, p103) used for appraisal of studies in the main systematic review.*

Study	**Journal quality**	**Clear research question, appropriate for research?**	**Valid research method?**	**Sufficiently large sample?**	**Appropriate sample?**	**Data collection appropriate to the research method?**	**Appropriate statistical test?**	**Quality – overall appraisal**
Benford (2008)	Unpublished dissertation. Peer-reviewed via PhD.	Yes.	Yes.	Yes.	Partially – no screening for comorbid disorders. Only 60% completed the Autism Quotient, and 19% of those which did failed to meet the clinical cut-off for autism.	Yes.	Not applicable – no statistical test in thematic analysis.	Medium.
Gotham and Brunwasser (2014)	Good. Peer-review with impact factor 4.532	Yes.	Yes.	Yes.	Yes, but no control group so not sure if autism-specific.	Yes.	Yes.	Medium to high.
Rice (2009)	Unpublished thesis.	Yes.	Yes.	Yes, but not for factor analysis.	Possibly not as intellectual disability may, to some extent, be a confounding variable in this study, with mean Global Adaptive Composite scores across all participants in the low range (*M* = 75.25). Furthermore, participant characteristics are not broken down for clinical groups so unclear about whether these are representative of their wider population.	Yes.	No. The sample size is not sufficiently large (*N* = 82) for adequate power for factor analysis.	Low to medium.
Russell et al. (2005)	High. Peer-review with impact factor 7.06	Yes.	Yes.	Yes.	Yes.	Yes.	Yes.	High.

Table 5.3. *Appraisal of the qualitative studies using the Critical Appraisal Skills Programme (2015) qualitative checklist in the main systematic review.*

Qualitative Checklist	Maloret and Scott (2018)		
	Yes	Can't tell	No
1. Was there a clear statement of the aims of the research?	✓		
2. Is a qualitative method appropriate?	✓		
3. Was the research design appropriate to the address the aims of the research?	✓		
4. Was the recruitment strategy appropriate to the aims of the research?	✓		
5. Was the data collected in a way that addressed the research issue?	✓		
6. Has the relationship between researcher and participants been adequately considered?	✓		
7. Have ethical issues been taken into consideration?	✓		
8. Was the data analysis sufficiently rigorous?			X
9. Is there a clear statement of findings?			X
10. How valuable is the research? (Comments)	Quite valuable, looking at specific causes of anxiety (mental health unit admission) in a relatively understudied population (autistic mental health patients), using a relatively unstudied measure of repetitive behaviour (Intolerance of Uncertainty) which may be a key feature of autism.		
Any comments from any of the items:	Question 4: Snowballing rather than random sampling. Question 9: Lack of any quantitative data and very unspecific findings (e.g. "a number of participants…") confounds a general unclarity to the findings.		
Quality appraisal:	**Low to medium** (due to lack of clear findings).		

Table 5.4. *Appraisal of the randomised control trial studies using the Critical Appraisal Skills Programme (2015) randomised control trials checklist in the main systematic review.*

Randomised Control Trial Checklist	Hollander et al. (2012)		
	Yes	Can't tell	No
1 Did the trial address a clearly focussed issue?	✓		
2 Was the assignment of clients to interventions randomised?	✓		
3 Were all of the clients who entered the trial properly accounted for at its conclusion?	✓		
4 Were clients, health workers, and study personnel 'blind' to intervention?	✓		
5 Were the groups similar at the start of the trial?	✓		
6 Aside from the experimental intervention, were the groups treated equally?	✓		
7 Was the intervention effect significant?	✓		
8 Was the estimate of the intervention effect precise?	✓		
9 Can the results be applied locally?	✓		
10 Were all important outcomes considered?	✓		
11 Are the benefits worth the harms and costs?	✓		
Quality appraisal:	High		

Table 5.5. *Quality Appraisal Tool (Effective Public Health Practice Project, 2010) for Quantitative Studies in the main systematic review.*

Quality Assessment Tool for Quantitative Studies	Brodkin et al. (1997)	Buckby (1999)	McDougle et al. (1998a)	Miyaoka et al. (2012)
Are the individuals selected to participate in the study likely to be representative of the target population?	Somewhat likely – high proportion of Intellectual Disability	Yes (clinical group). Control group may be unrespresentative of general population.	Somewhat likely – high proportion of Intellectual Disability	Very likely
What percentage of selected individuals agreed to participate?	Can't tell	41%	Can't tell	80% - 100% agreement
Indicate the study design	Cohort	Cross-sectional questionnaire	Cohort	Cohort
Was the study described as randomized?	No	No	No	No
Were there important differences between groups prior to the intervention?	No	Yes - significany different age and ability levels.	No	No
Was (were) the outcome assessor(s) aware of the intervention or exposure status of participants?	Yes	Yes	Yes	Yes
Were the study participants aware of the research question?	Yes	No	Yes	Yes
Were data collection tools shown to be valid?	Yes	Yes	Yes	Yes
Were data collection tools shown to be reliable?	Yes	Yes	Yes	Yes
Were withdrawals and drop-outs reported in terms of numbers and/or reasons per group?	Yes	Yes	Yes	Yes
Indicate the percentage of participants completing the study.	80% - 100%	75% (clinical) and 66.7% (control group)	80% - 100%	80% - 100%
What percentage of participants received the allocated intervention or exposure of interest?	80% - 100%	75% (clinical) and 66.7% (control group)	80% - 100%	80% - 100%
Was the consistency of the intervention measured?	Yes	No	Yes	Yes
Is it likely that subjects received an unintended intervention (contamination or co-intervention) that may influence the results?	No	No	No	No
Indicate the unit of allocation	Community	Community	Community	Community
Indicate the unit of analysis	Practice/office	Practice/office	Practice/office	Practice/office
Are the statistical methods appropriate for the study design?	Yes	Yes	Yes	Yes
Is the analysis performed by intervention allocation status (i.e. intention to treat) rather than the actual intervention received?	Yes	N/a	Yes	Yes

Quality Assessment Tool for Quantitative Studies: Component Ratings	Brodkin et al. (1997)	Buckby (1999)	McDougle et al. (1998a)	Miyaoka et al. (2012)
Selection Bias	Moderate	Moderate	Moderate	Strong
Design	Moderate	Moderate	Moderate	Moderate
Confounders	Strong	Moderate	Strong	Strong
Blinding	Weak	Weak	Weak	Weak
Data Collection Methods	Strong	Strong	Strong	Strong
Withdrawals and drop-outs	Strong	Moderate	Strong	Strong
Global rating for the paper	**Moderate**	**Moderate**	**Moderate**	**Moderate**

5.6. Results

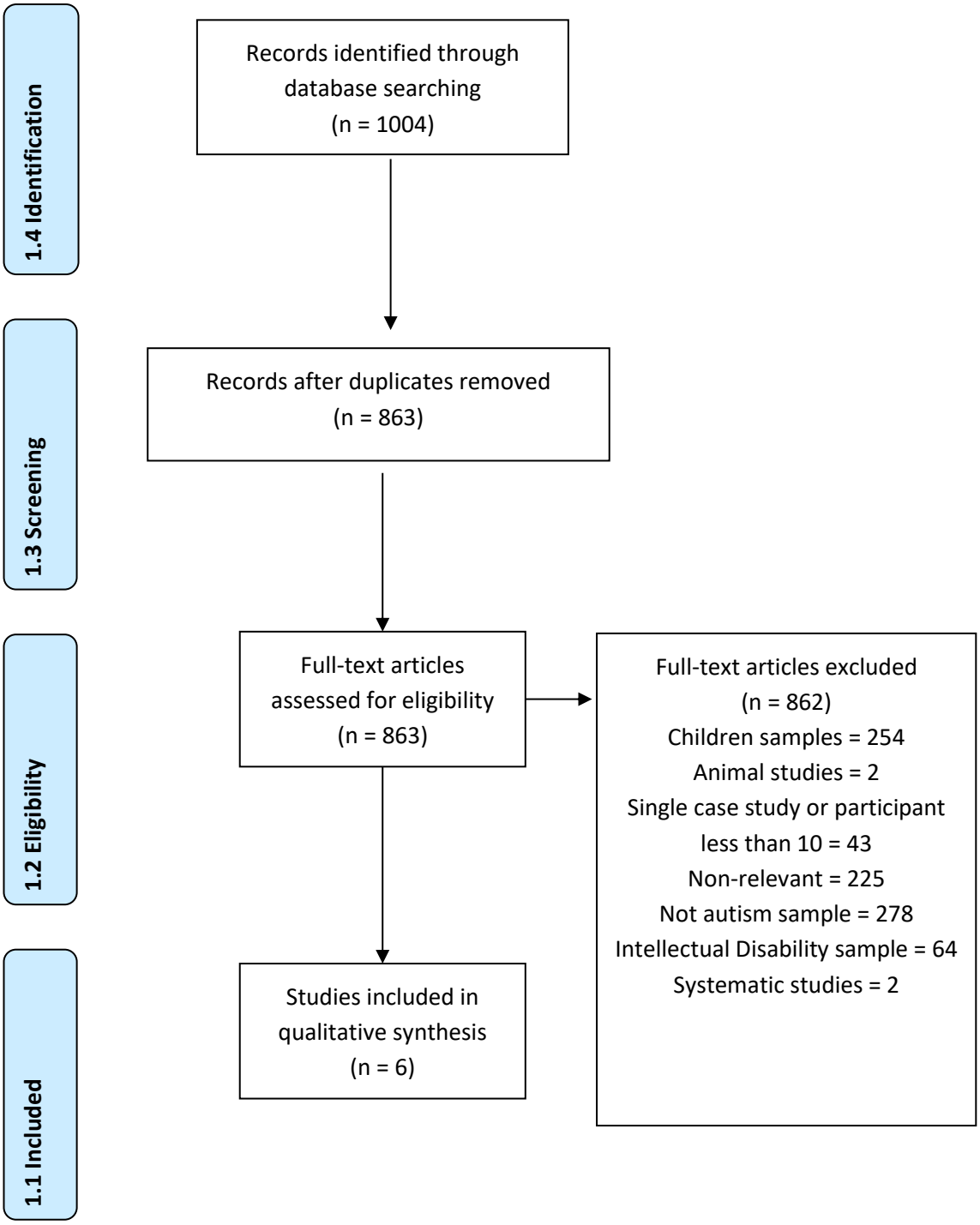

Figure 5.1. PRISMA (Moher et al. 2009) study selection flow chart for main study systematic literature review.

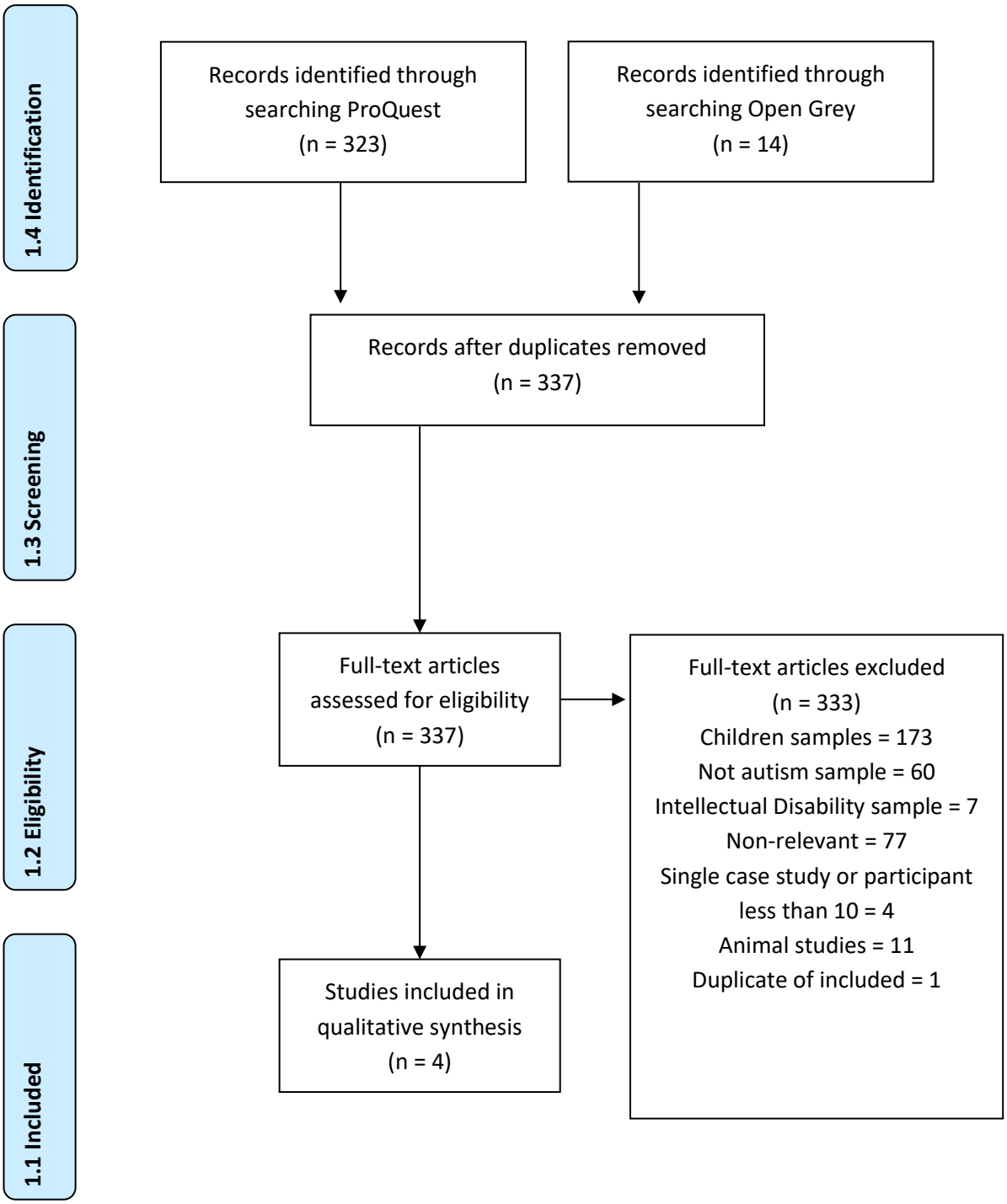

Figure 5.2. *PRISMA (Moher et al. 2009) study selection flow chart of grey literature for main study systematic literature review.*

5.6.1. Overview of literature.

A summary of the main findings of the included studies are described in Table 5.6. A narrative overview of the critical appraisals is presented below, before the main thematic findings. Included papers were published in 1996 onwards. There were three main methodologies across the studies. The two most relevant methodologies provided some direct comparison of affect and repetitive traits: four of the papers involved a cross-sectional questionnaire method (Benford, 2008; Gotham & Brunwasser, 2014; Russell et al., 2005; Rice, 2009); whilst a further two studies made a comparison through a semi-structured interview method (Buckby, 1999; Maloret & Scott, 2018). Two of the studies attempted novel ego-dystonic/ego-syntonic measures of repetitive traits (Buckby, 1999; Rice, 2009). The final yield included pharmacological trials, despite the indirect comparison between affect and Repetitive Behaviours (Brodkin, McDougle, Naylor, Cohen, & Price, 1997; Hollander et al., 2012; McDougle et al., 1998a; Miyaoka et al., 2012). This indirect evidence was retained for the systematic review after appraisals revealed variable relevance of the already limited evidence from the cross-sectional and interview designs.

Table 5.6. *Summary of the main findings of each article for the main systematic review.*

Study	Design	Questionnaires	Participants	Statistical tests	Outcome measure	Results	Appraisal
Benford (2008)	Mixed methods: cross-sectional questionnaires and semi-structured interview.	1) Internet questionnaire. 2) Autism-spectrum quotient. .	133 adults (16 years plus) with a self-reported diagnosis of autism (5 additional reported no formal diagnosis). Mode age 21-30 years. 72% male. 85% Caucasian.	Although Chi Squares and correlations were run for some of the data, all the data of relevance for the present research question were analysed using thematic analysis.	Investigation of the relationship between internet use and communication preferences (including online satisfaction).	Primary: Satisfaction (affect) inversely related to (repetitive) use of the internet. Secondary: Higher proportion use of internet for communication compared to previously found in non-clinical groups (though relatively young age in study). Communication via email more popular than face-to-face conversations with friends. Despite reports of the benefits of internet as less socially demanding form of communication, relatively few used for online live chat.	No clear statistical evidence in relation to the present research question due to the thematic analysis used for the mood aspect of communication. Despite meeting 6 of the 7 appraisal principles, low relevance to research question – very narrow measure of both repetitive trait (use of internet) and affect (satisfaction).

Study	Design	Questionnaires	Participants	Statistical tests	Outcome measure	Results	Appraisal
Brodkin et al. (1997)	Open-label pharmacological trial.	Yale-Brown Obsessive Compulsive Scale for measures of OCD-related repetitive traits. CGI scale used as a measure of clinical improvement and Brown Aggression Scale as measure of aggressive symptoms. Ritvo-Freeman Real-Life scale used to assess improvements to "autistic symptoms".	35 patients (mean age 30.7 years) with DSM-IV confirmed diagnoses of autism (and no other Axis I or II disorder) referred by homes, psychiatrists and families. 69% males. Full-scale IQ of 64.6 (*SD* = 27.2).	One-way ANOVA to assess main effect of treatment over time. Chi square - relationship between treatment response and autistic behaviour.	To measure the clinical efficacy of sertraline on repetitive behaviours and global severity.	18 (55%) were categorised as treatment responders, based on minimum of "much improved" on CGI scale. Treatment significantly reduced aggression over time in all participants. Significant decrease of total Y-BOCS scores over time, whilst no significant decrease of repetitive thoughts over time for 18 verbal participants. Effect sizes are not reported due to the open-label methodology (no control group).	Moderate global rating for the paper – see Critical Appraisal Tool (Table 5.5). Low-IQ as a confounding factor.

Study	Design	Questionnaires	Participants	Statistical tests	Outcome measure	Results	Appraisal
Buckby (1999)	Semi-structured interview	1) Obsessive-Compulsive Inventory. 2) Novel semi-structured interview of functional assessment of repetitive interests based on the severity subscale of the Yale-Brown Obsessive Compulsive Scale. A measure of ego-dystonic and ego-syntonic origins (self-reported: distress; interference; sense of compulsion; resistance; avoidance; excessive sense of responsibility; indecisiveness; and positivity or negativity in relation to how the trait affects them personally).	1)16 adults with autism (mean age 27.25 years). 2) 11 informants of autism participants. 3) 21 hobbyists (mean age 54.67 years) 4) 14 informants of hobbyists.	1) Kappa coefficients 2) T-tests. 3) Fisher's Exact Test. In addition, observations were made by the researchers based on the qualitative data.	1) To compare difference between groups on motivational reasons for repetitive interests (hobbies).	Few overall differences between the two groups on measures of ego-dystonic and ego-syntonic (affective) functions of hobby (repetitive trait). No significant difference in motivation for hobby ($t(35) = 1.98$; $p = .06$; $d = 0.66$). Higher OCD trait scores via the Y-BOCS for the autism group: obsessions subscale (medium effect size: $r = -.40$; $p = .02$); total score (medium effect size: $r = -.39$; $p = .02$). No significant difference for compulsions subscale ($t(34) = -1.81$; $p > .05$; $d = -0.60$).	Not high relevance to research question – narrow repetitive measure. Moderate global rating for the paper – see Critical Appraisal Tool (Table 5.5).

Study	Design	Questionnaires	Participants	Statistical tests	Outcome measure	Results	Appraisal
Gotham and Brunwasser (2014)	Cross-sectional questionnaires	1) Autism-Diagnostic Interview-Revised. 2) Vineland Adaptive Behavior Scale 3) Battery of self-reported demographics and psychological health. 4) Behavioral Perception Inventory 5) Social Interests and Habits Questionnaire	50 adolescents and adults (16-36 years) with a clinical diagnosis of an autism spectrum disorder. Screened to exclude: verbal IA below 70; low reading comprehension; significant sensory impairment; and psychiatric disorder.	1) Multiple linear regression modelling. 2) Exploratory methods based on Principal Component Analysis.	The relationship between depressive symptoms and psychosocial constructs, including rumination.	Primary: Association between rumination (affect) and Insistence on Sameness (repetitive trait) approached significance with a small effect size (sr^2 = .22; b = 0.10). Increased depressive symptoms associated with increased rumination with a medium effect size (sr^2 = .40; b = 0.40).	High relevance to research question. Meets at least 6 quantitative study appraisal principles.
Hollander et al. (2012)	Randomised control pharmacological trial.	The compulsion subscale of the Yale-Brown Obsessive Compulsive Scale was used as measures of repetitive traits. Aberrant Behavior Checklist used to measure an element of mood.	37 adults (mean age 31.4 years) who were assessed to meet DSM-IV criteria for Autism. 69% male, 73% white. Mean Full-Scale IQ 103.2.	Fisher's exact test for clinical improvements on the CGI scale (Clinical Global Impression).	To measure the clinical efficacy of fluoxetine (compared to placebo) on repetitive behaviours and global severity.	Compulsions significantly reduced between the treatment and placebo group at the end of the 12 week trial with a medium effect size (F=9.24, df=1, 30.7, p=0.005, d=0.53). Whilst irritability was reduced in the treatment group also, this did not reach statistical significance.	Strong methodology, although only indirect evidence for the current research question.

Study	Design	Questionnaires	Participants	Statistical tests	Outcome measure	Results	Appraisal
McDougle et al. (1998a)	Open-label pharmacological trial.	Vineland Adaptive Behaviour Scale and Y-BOCS as measures of clinical and repetitive traits. CGI scale used as a measure of clinical improvement.	42 adults (mean age 26.1 years) of which 2 were inpatients and 40 outpatients, referred by homes, psychiatrists and families. 64% male. Minimum Y-BOCS score of 15 required by excluded participants with other DSM-IV axis I or II disorder.	ANOVAs were used to analyse the effect of time and subtype (of autism) on behavioural ratings. Chi-square was used to analyse the relationship between IQ and treatment response.	To measure the clinical efficacy of sertraline on repetitive behaviours and global severity.	24 participants (57%) were reported to be treatment responders, mostly with improvements in repetitive and aggressive symptoms – none to social symptoms.	Moderate global rating for the paper – see Critical Appraisal Tool (Table 5.5). Open-label trial methodology (with lack of control) limits the validity.
Maloret and Scott (2017)	Semi-structured interview.	None.	21 mental health clinical patients - adults with confirmed autism diagnosis, recruited by purposive then snowball sampling. 60% male, average age 35.5 years	1) Interpretative phenomenological analysis.	Exploration of emotional and psychological experiences of being a mental health inpatient.	Two superordinate themes of anxiety and coping strategies – lack of routine and structure (restricted and Repetitive Behaviour) a subtheme under anxiety (affect).	Not high relevance due to lack of direct repetitive measure or mood. Only a low to medium quality appraisal (see Table 5.3).

Study	Design	Questionnaires	Participants	Statistical tests	Outcome measure	Results	Appraisal
Miyaoka et al. (2012)	Open-label pharmacological trial.	CGI scale used as a measure of clinical improvement. Aberrant Behaviour Checklist-Irritability Scale.	40 participants (children, adolescents and adults: mean age 22.7 years ± 7) referred through mental health clinic, with DSM-IV diagnosed autistic disorder. 55% male. IQ = 88.9± 7.3.	T-tests calculated to analyse the change in behaviour at start-point and at the end of the 12 week trial.	To measure the clinical efficacy of herbal Yokukansan (TJ-54).	90% of participants were reported as respondents, with a minimum CGI-S of 1 and 25% improvement on ABC-1. These participants improved in irritability measures (aggression, self-injury and tantrums ($p < 0.0001$).	Moderate global rating for the paper – see Critical Appraisal Tool (Table 5.5). Open-label trial methodology (with lack of control) limits the validity.

Study	Design	Questionnaires	Participants	Statistical tests	Outcome measure	Results	Appraisal
Rice (2009)	Cross-sectional questionnaires.	1) Novel functional assessment of Repetitive Behaviours. 59 ego-related functional characteristics consisting of: risk-avoidance; pleasure-seeking; soothing; distress; adaptive; and disruptive. 2) Yale-Brown Obsessive Compulsive Scale. 3) Adaptive Behavior Assessment System-Second Edition. 4) Gilliam Autism Rating Scale-Second Edition.	82 children and adults (43 adults; 70 male; 61 Caucasian) within 3 groups: OCD-only (25); autism-only (23); and comorbid OCD and autism (34). Unclear characteristics within each disorder.	1) Exploratory factor analysis and validity analysis. 2) Correlational analysis. 3) Analysis of means (MANOVA/ ANOVA).	To assess the functional characteristics of Repetitive Behaviours in autism.	Positive affect (both self-reported soothing and pleasure-seeking) higher in the autism group: soothing (autism only compared to the OCD-only and comorbid groups) with a small effect size (*d* = -.21; *F* = 12.50); pleasure-seeking (autism only compared to the OCD-only and comorbid groups) with a small effect size (*d* = .21; *F* = 17.10). Parent-reported (but not self-reported) levels of distress self-reported intrusiveness was reported to be significantly increased in the OCD-only group; distress with a medium effect size (*d* = -.40; *F* = 7.79); and intrusiveness with a large effect size (*d* = -.82; *F* = 15.72).	Fairly high relevance to research question, although measure of Repetitive Behaviour not comprehensive. Meets only 4 of the quantitative study appraisal principles.

Study	Design	Questionnaires	Participants	Statistical tests	Outcome measure	Results	Appraisal
Russell et al. (2005)	Cross-sectional questionnaire	1) Yale-Brown Obsessive Compulsive Scale, including symptom checklist	Consecutive referrals to clinic. Screened to exclude: intellectual disability; comorbid psychosis; substance misuse. Two groups: 1) 40 adults with ICD-10 confirmed autism, 2) 45 gender matched adults diagnosed with DSM-IV or ICD-10 OCD.	1) Discriminant functional analysis 2) Chi-square 3) T-test	Comparison of OCD measures between the two clinical groups.	Primary: High distress (affect) and obsession (repetitive thought) in autism group: 39% ≥ moderate duration; 47% ≥ moderate interference; 60% ≥ moderate distress. High distress (affect) and compulsions (repetitive trait) in autism group: 26% ≥ moderate duration; 42% ≥ moderate interference; 42% ≥ moderate distress. Secondary: Similar frequency of OCD traits between the groups, except increased somatic obsessions and repeating and checking compulsions in OCD. Higher severity of obsessions and compulsions in the OCD group.	Fairly high relevance to research question, although limited measure of repetitive trait (OCD trait). Meets all 7 quantitative study appraisal principles.

5.6.2. Critical appraisal summary: cross-sectional and interview methodologies.

The critical appraisal methodology used was guided by Aveyard (2010, p.103) and presented in Table 5.2. Two of the studies recruited participants within mental health clinics (Maloret & Scott, 2018; Russell et al., 2005) and therefore may not be representative of the wider autistic population, due to acute psychiatric symptoms. Additionally, comorbid diagnoses and/or symptoms were an issue in of the studies (Maloret & Scott, 2018; Rice, 2009) including: a large proportion of acute psychiatric symptoms (Maloret & Scott, 2018); and intellectual disability as a potential confound (Rice, 2009). Regardless, these articles were retained as otherwise they were relevant to the research question.

Of the remaining cross-sectional questionnaire methods, only one was highly relevant to the present research question, with both strong measures of repetitive traits and affect (Gotham & Brunwasser, 2014). Of all the cross-sectional questionnaire studies, there were some minor concerns with the strength of the methods. Gotham and Brunwasser (2014) met with good appraisal characteristics (see Table 5.2), although the lack of a control sample means it is unclear whether the effects are autism-specific. Of the other four cross-sectional studies, two were relevant to the research question, although each involved limited measures of repetitive traits (Russell, Mataix-Cols, Anson, & Murphy, 2005; Rice, 2009): one of these relied on OCD traits as the sole measurement of repetitive traits (Russell, Mataix-Cols, Anson, & Murphy, 2005). The final study (Benford, 2008) had a low relevance to the research question with both a narrow measure of repetitive traits (i.e. internet usage) and affect (satisfaction). Of the two semi-structured interview methods, both had a low relevance to the research

question: Buckby (1999) used a narrow repetitive measure based on OCD traits, whilst Maloret and Scott (2018), lacked a direct measure of both repetitive traits and affect (in addition to receiving a low to moderate critical appraisal: Table 5.3).

5.6.3. Critical appraisal summary: pharmacological studies.

The quality of the single randomised control trial (Hollander et al., 2012) was also strong (see Table 5.4). The remaining three studies used quantitative methods (Brodkin et al., 1997; McDougle et al., 1998a; Miyaoka et al., 2012) and were appraised using the Critical Appraisal Tool (Effective Public Health Practice Project, 2010), as outlined in Table 5.5. Global ratings for all three papers were moderate. A notable weakness in all was a lack of blinding procedures. Relative strengths for all studies were in control of confounding variables, good data collection methods and control of withdrawals and drop-outs. The only difference between all three quantitative methods was the relative strength of controlling for selection bias in one of the studies (Miyaoka et al., 2012).

5.7. Main findings

The following section provides a discussion within the themes, following the recommendation of Aveyard (2010).

5.7.1. Association between positive mood and repetitive traits.

Ego-syntonic is defined as being consistent with the individual's view of their self. Accordingly, these traits are likely, to some extent, to be wanted, desirable and related to positive affect. As OCD traits are clinically ego-dystonic (i.e. unwanted and

undesirable), identifying ego-syntonic traits in autism would indicate a distinction between repetitive traits in autism and OCD. This would provide an initial starting point to establishing the theoretical validity of a Compulsive and Repetitive Trait framework, to compare disorders such as autism and OCD.

Although repetitive traits in autism have been typically assumed to be ego-syntonic (see Chapter 4), only three of the studies reported generally ego-syntonic qualities of repetitive traits in adults with autism (Benford, 2008; Buckby, 1999; Rice, 2009). The stronger of the three methodologies, (Rice, 2009), reported higher pleasure-seeking and soothing qualities of OCD traits and Repetitive Behaviours in adults with autism, compared to OCD participants. Similarly, internet usage appears to be related to positive mood (satisfaction) in adults with autism; although the validity of this study by Benford (2006) is compromised by the lack of autistic participants identified by Autism Quotient screening (see Table 5.2). Importantly, there is evidence adults with autism appear to be able to distinguish between repetitive interests and OCD traits (Buckby, 1999), indicating the necessary levels of introspection needed for ego-dystonia may be present in autism. This should not be surprising, however, as the Bucky's (1999) recruited sample was specifically recruited with high verbal ability.

Buckby (1999) indicated hobbies and interests may be related to positive mood through the function of self-esteem, although this was not found to be autism specific, being a feature in non-autistic "hobbyists" also. This result, from a study with a moderate global rating (see Table 5.5), measures only a narrow facet of repetitive traits (i.e. hobbies and interests). Rice (2009), however, also reported repetitive traits

may be driven more by ego-syntonic function in adults with autism, compared to matched peers with OCD (Rice, 2009). Buckby (1999) suggests interests seem to be driven as a compensatory mechanism for difficulties with social relationships in autistic adults, compared to more neurotypical peers, who may be likely to be motivated by factors related to intellect, skill or inherent qualities of an interest. Few differences in ego-dystonic measures (e.g. interference, distress, feelings of compulsion) appear to differentiate autistic from more neurotypical participants (Buckby, 1999). High levels of intrusiveness may be a feature of repetitive traits in peers with OCD (Rice, 2009), whilst measures of OCD traits (specifically obsessions, rather than compulsions) have been reported to be higher in autistic adults (Buckby, 1999).

The functional assessment method (Buckby, 1999; Rice, 2009) are highly relevant to understanding ego-syntonic and ego-dystonic origins of repetitive traits. Understanding motivational factors may be key to understanding whether these repetitive traits are ego-dystonic (undesirable and unwanted) or ego-syntonic (serving a purpose for the individual and positive in nature). However, both these studies had methodological limitations. Despite a more thorough methodology – employing Repetitive Behaviour and OCD assessment tools, for example – Rice (2009) recruited an insufficient sample size, meaning the novel set of 59 ego-related functional characteristics could not be adequately validated.

Further methodological issues confound the studies employing semi-structured interview methods (Benford, 2008; Buckby, 1999). Firstly, this method is probably not the most reliable method for autistic participants with inherent communication and

social difficulties. Additionally, measures of hobbies/interests (Buckby, 1999) as well as internet usage (Benford, 2008) involves a very narrow subset of a larger set of Repetitive Behaviours. Furthermore, Benford's (2008) correlational methodology limited the ability to identify whether any of these findings were autism-specific.

In summary, whilst there is evidence some repetitive traits demonstrated by autistic adults may have ego-syntonic (pleasure-seeking and soothing) qualities (Rice, 2009), as well as repetitive interests being important for self-esteem (Buckby, 1999), the evidence of ego-syntonic repetitive traits is both limited and based on methodologies which lack robustness: as described above, the results from the studies reported need replicating (Rice, 2009) or refining (Benford, 2008; Buckby, 1999). However, if the findings from these studies are replicated, it would indicate at least some repetitive traits in autism can be distinguished from the ego-dystonic characteristics of repetitive OCD traits. The lack of evidence of ego-syntonic traits in autistic adult samples may reflect that repetitive traits is not regarded necessary for a diagnosis in adults with autism, which may have resulted in few researchers finding it relevant to study. Demonstrating, ego-dystonic repetitive traits in autism would increase the theoretical validity in establishing a symptomological model to compare these autism and OCD.

5.7.2. Association between negative mood and repetitive traits.

5.7.2.1. Obsessive-compulsive disorder traits in autism.

Ego-dystonic is defined as being inconsistent with the individual's sense of self. Being unwanted and undesirable, these traits should be accompanied by a measure of negative affect, such as distress or anxiety. As OCD is diagnostically ego-dystonic, presence of OCD symptoms in autism would further indicate the relevance of a shared

framework. Evidence linking autism and OCD suggests ego-dystonic traits may be present in autism.

One study (Russell et al., 2005) measured repetitive traits exclusively as OCD traits. Comparable frequencies of OCD traits between adults with OCD and autism have been reported (Russell et al., 2005). There is evidence of obsessions being at least moderately distressing in 47% of a group of adults with autism, with compulsions being at least moderately distressing in 42% of the sample (Russell et al., 2005). This study is high in quality (see appraisal Table 5.2), although it slightly lacks relevance to the present research question through measuring a single narrow construct of repetitive traits (i.e. OCD symptoms).

5.7.2.2. Insistence on Sameness.

From the two studies which indirectly measured ego-dystonic origins of repetitive traits in autism (Gotham & Brunwasser, 2014; Maloret & Scott, 2018), two implicated the significance of Insistence on Sameness (Gotham & Brunwasser, 2014; Maloret & Scott, 2018), a higher-order (more cognitively complex) component of Repetitive Behaviours (a repetitive trait measure originating from autism and intellectual disability research). The reported significant correlation of Repetitive Behaviour traits and depressive symptoms (Gotham & Brunwasser, 2014) appear to indicate Insistence on Sameness, specifically, moderating the link between rumination (perseverative and passive thinking as a reaction to a stressor) and depression in adults with autism. This may be significant as higher-order repetitive traits have been suggested to be specific to autism (see Chapter 4).

Whilst this potential link between Insistence on Sameness implies a differential pathway between lower- and higher-order repetitive traits and mood in autism, again these finding needs replicating, particularly as both studies failed to recruit a control group.

5.7.3. Pharmacological trials: overview.

Only two studies attempted to measure ego-dystonic and ego-syntonic properties directly (Buckby, 1999; Rice, 2009), whilst all other studies indirectly measured these properties using an assessment of mood/affect alongside a repetitive trait measure. As detailed above, all these studies lack either relevance or robustness, particularly through methodological issues which necessitate replication of the key findings. Therefore, to increase the evidence base, four pharmacological articles (Brodkin et al., 1997; Hollander et al., 2012; McDougle et al., 1998a; Miyaoka et al., 2012) were identified which met all the inclusion criteria as outlined in section 5.3.4. These were the only pharmacological trials which reported some measure of both mood and repetitive traits as outcomes. Although the link between mood and repetitive traits were only indirectly reported (i.e. as separate outcomes, rather than as a correlation between the two variables), the evidence was retained for this narrative synthesis due to the lack of studies otherwise yielded.

A small number of pharmacological studies have measured both repetitive traits (OCD traits) and an assessment of mood as outcomes to intervention in samples of adults with autism. Reductions across both domains have provided some evidence to indicate mood and repetitive traits in autism may be linked. However, this evidence is weak,

with relatively few trials employing randomised control trial methodologies. Overall, OCD trait scores of participants recruited in these studies were typically within the moderate range, potentially comparable to the wider autistic population. However, eligibility to be enrolled on these medical trials requires a high level of general maladaptive behaviour dysfunction. The evidence provides some indication of a relationship between mood and repetitive traits in autism, with reports of improvements in both symptoms. However, this association is only indirect – there may be other more important factors which moderate the pathway between mood and repetitive traits and the overall evidence from these clinical trials is not sufficiently robust. Whilst it adds some weight to the link between negative mood and repetitive traits (specifically OCD traits) in adults with autism, a potential link between repetitive traits and mood can only be very tentatively made from these findings.

5.7.3.1. Randomised controlled trials.

Randomised controlled trials (RCTs) are the strongest methods for analysing pharmacological effects. One study employed this methodology (Hollander et al., 2012). Hollander et al. (2012) failed to find a link between repetitive traits and negative mood in another RCT. Employing a double-blind placebo-controlled methodology to investigate the clinical effects of fluoxetine across 12 weeks, repetitive traits were measured exclusively with an OCD measurement tool (the Yale-Brown Obsessive Compulsive Scale), with clinical improvements measured using the Clinical Global Impression scale. Across 37 adult participants (with unclear recruitment techniques, presumably through access to psychiatric clinics), compulsions (the sole measure of repetitive trait) was significantly reduced in the treatment group,

compared to the placebo-administered group, with a medium effect size (*d* = 0.53). However, whilst irritability (a measure of mood taken) was reported to also reduce in the treatment group, this result did not reach clinical significance.

Future investigations may provide more evidence if they expand measures of repetitive traits, as well as more accurate measure of mood (which may consist of both ego-dystonic and ego-syntonic perceptions). However, whilst the quality of the Hollander et al. (2012) study is high (see Table 5.4), the relevance is low due to the indirect measure of mood and repetitive trait measure. There is currently no clear evidence of a link between any measure of mood and repetitive trait in the indirect evidence of pharmacological RCTs.

5.7.3.2. Open-label trials.

Open-label trials use a methodology where all individuals involved are aware of the interventions used. Lacking many of the controls used in randomised control trials (such as placebo, control group and double-blinding), firm conclusions cannot be made but may provide some further evidence of the relationship between repetitive traits and mood in autism.

There is more evidence to indirectly indicate a potential link between repetitive traits and mood in these less robust methodologies. Similar findings have been reported in response to reducing both repetitive and aggressive symptoms in adults with autism as a result of a 12-week intervention of sertraline (McDougle et al., 1998a) and clomipramine (Brodkin et al., 1997), with 57% of 42 participants and 55% of 33

participants demonstrated a reduction in these symptoms, respectively. McDougle et al. (1998a) reported improvements in repetitive and aggressive, but not social relatedness symptoms, potentially indicating a link between mood and repetitive traits. Brodkin et al. (1997) did, however, report improvements on all "autistic symptom" subscales across the treatment responders. Furthermore, general clinical improvements reported included compulsive symptoms, aggressive symptoms, agitation and self-injury, although intervention did not decrease repetitive thoughts (Brodkin et al., 1997). Despite not being placebo-controlled, comparable clinical improvements were also reported in an open-trial of a Japanese herbal remedy, yokukansan or TJ-54 (Miyaoka et al., 2012), with 90% of the 40 participants demonstrating improvements in clinical symptoms including irritability, aggression and stereotyped behaviours. Despite the lack of blinding, the global rating for the strength of the evidence of all these studies were moderate and comparable (see Table 5.5). Whilst comorbid intellectual disabilities were high in two of the studies (Brodkin et al., 1997; McDougle et al., 1998a), clinical response was not found to be associated with diagnosis of intellectual disability (McDougle et al., 1998a).

In summary, there is some evidence of links between (negative) mood and repetitive traits in pharmacological trials, but only from open-trial studies. This evidence can only be very tentatively taken currently, as the quality all three studies are moderate due to the lack of strict control in these open-label designs (see Table 5.5).

5.8. Discussion

Many repetitive traits in autism have been traditionally viewed as being ego-syntonic in nature. Simple repetitive motor behaviours (such as spinning, twirling and flapping) – overtly typified by individuals with comorbid intellectual disabilities – were given more prominence in the theory of functional origins of Repetitive Behaviours in autism. Increasing evidence of links between OCD and autism has generated research comparing OCD trait symptomology in autism. From this systematic search, only two studies were found which attempted to directly define and assess ego-syntonic and ego-dystonic measures of repetitive traits in adults with autism (Buckby, 1999; Rice, 2009). These studies provide some indication there may be both ego-dystonic and ego-syntonic (Buckby, 1999; Rice, 2009) functions of repetitive traits in this population. Overall, there is a general lack of investigation comparing mood and repetitive traits in adults with autism. The systematic search yielded only six studies which directly compared some measure of repetitive trait with some measure of mood (Benford, 2008; Buckby, 1999; Gotham & Brunwasser, 2014; Maloret & Scott, 2018; Russell et al., 2005; Rice, 2009). There were several key findings. Fundamentally, adults with autism appear to be able to show sufficient introspection to distinguish between repetitive interests and OCD traits (Buckby, 1999). Some repetitive traits demonstrated by this group appear to be associated with positive affect, such as pleasure-seeking (Rice, 2009), or repetitive interests improving self-esteem (Buckby, 1999). As there is a severe lack of robust measures directly assessing ego-syntonic properties of repetitive traits, these findings may reflect ego-syntonic properties of these traits. Similarly, there is also a lack of direct measures of ego-dystonic properties of repetitive traits in adults with autism. However, there is evidence of increased OCD traits in this

population and these repetitive traits are, by definition, ego-dystonic in origin. Specifically, OCD traits appear to be highly relevant in autism, comparable in frequency to those found in peers with OCD and associated with distress (Russell et al., 2005). The pathway between mood and repetitive traits in autism appears to be complex, with higher-order repetitive traits (specifically Insistence on Sameness) being implicated (Gotham & Brunwasser, 2014). Pharmacological trials provide some further evidence of ego-dystonic origins of repetitive traits in autism, although they consist of narrow measures (OCD traits), are not sufficiently robust and provide only indirect support. The overall evidence generally requires replicating. Many methodological issues confound the results including problems with comorbidity (Maloret & Scott, 2018; Rice, 2009) and a lack of validated measures of ego-dystonic and ego-syntonic functions of repetitive traits (Buckby, 1999; Rice, 2009).

This evidence at least indicates there is insufficient evidence to reject a framework to compare autism and OCD based on repetitive traits. Adults with autism appear to display both ego-syntonic repetitive traits (which differentiate them from peers with OCD), and ego-dystonic traits (which may be relatively comparable between the two disorders).

A major limitation of the evidence identified is the lack of the quality of the reported studies (demonstrated in the quality appraisal information – see Tables 5.2 to 5.5). Only two of the studies (Russell et al., 2005; Buckby et al., 1999) scored highly on appraisal assessment. Most of the other studies were confounded by the lack of appropriate sampling specific to the present research question (i.e. repetitive traits

and mood variables in autistic adults). Additionally, the pharmacological trials present only indirect information in relation to this research question. Overall, the lack of evidence only goes to emphasise the necessity for future research to further this investigation.

To address these various methodological issues, improvements can be made to increase our understanding of the relationship between mood and repetitive traits in adults with autism. Procedures should minimise the impact on communication and social difficulties, to improve ecological validity in a population with such limitations. Prevention of traits has been indicated to be relevant (Buckby, 1999). Therefore, avoidance behaviours should be assessed to decrease the possibility of false negatives. Comorbidity issues must be carefully addressed. Also, a comparison with both OCD and neurotypical participants is likely to be necessary. Finally, the measures used must either be validated, or large enough samples should be recruited to sufficiently power psychometric analyses.

Ego-dystonic and ego-sytonic origins of repetitive traits have been indicated in the preceding chapters to be key to understanding a potential symptomological overlap between autism and OCD. Evidence of ego-syntonic relationships may be identified if investigations employ non-OCD related measures of repetitive traits, whilst combining measures of ego-dystonic and ego-syntonic repetitive traits would provide a more balanced view of the phenomenology of repetitive traits in autism. This framework, therefore, should combine the evidence and assessment of both Obsessive-Compulsive Traits and Repetitive Behaviours, originating from OCD research and

autism and intellectual disabilities research, respectively. The following chapter presents an empirical investigation to support this theory.

Chapter 6. Psychometric properties of Obsessive Compulsive Disorder and Repetitive Behaviour Assessment Tools

Before empirically testing repetitive traits in OCD and autism, it is pertinent to decide on the most valid assessment tools. Research investigating the psychometric properties of these tools provide a good evidence base for this purpose. Examples of such measures are presented in Table 6.1, as provided by Streiner and Norman (2008). For the purposes of the overall aim of this thesis, the assessment tools should cover repetitive traits identified in both OCD and autism. Accordingly, a measure of OCD traits (from OCD research) and a measure of Repetitive Behaviours (from autism research) is relevant. These measures, along with evidence of their psychometric properties, is presented in this chapter.

Background

The Yale-Brown Obsessive Compulsive Scale (Y-BOCS; Goodman et al., 1989b) is generally considered the "gold standard" measurement tool for OCD (e.g. Deacon & Abramowitz, 2005; Wu et al., 2014). For this reason, the Y-BOCS was highlighted as a potential measurement tool for OCD traits in the current study. However, to determine the validity in using this tool, a systematic literature review was performed on evidence of psychometric properties in the literature on Y-BOCS studies. Additionally, a systematic literature review was undertaken on other OCD assessment tools, to provide a comparison. These OCD tools were identified by an appropriate review by Storch, Benito and Goodman (2011), which highlighted significant tools to the literature: Padua Inventory (Sanavio, 1988); Leyton Obsessional Inventory (Cooper, 1970); Florida Obsessive Compulsive Inventory (Storch et al., 2007); Obsessive

Compulsive Inventory-Revised (Foa et al., 2002); NIMH Global Obsessive Compulsive Scale (Guy, 1976); and the Vancouver Obsessional Compulsive Inventory (Thordarson et al., 2004).

Additionally, a systematic literature review was performed to investigate psychometric properties of Repetitive Behaviour tools in adults with autism, with a view to selecting appropriately for the present investigation. The tools selected were identified from the summative in-depth review (a thorough review of the literature on compulsive and repetitive traits as part of the first two years of the present thesis) of the literature on compulsive and repetitive traits in disorders including autism, Tourette's syndrome and OCD, and consisted of: Repetitive Behaviour Questionnaire (Leekam et al., 2007); Repetitive Behaviour Questionnaire (Moss et al., 2009); Childhood Routines Inventory (Evans, Leckman, Reznick, Hanshaw, King, & Pauls, 1997); and the Repetitive Behaviour Scale (Bodfish et al., 2001).

Table 6.1. *Psychometric measure definitions by Streiner and Norman (2008), identified for the review.*

Internal consistency	This is the expectation that scores across the scale will be correlated with other scores on the same scale if they are all measuring the same thing i.e. in this case OCD.
Test-rest reliability	This is a measure of stability, which is concerned with whether the scale would reproduce the same results across time.
Inter-rater reliability	Another measure of stability, this is the degree of agreement between different clinicians using the same test.
Convergent validity	A measure of how related the measure is to other tools/measures of the same thing to "which it should be related". For example, OCD should be related to insight into the disorder, since it is a key feature of OCD. Streiner and Norman (2008) state there may be different causes for a lack of correlation, such as the new scale or the theory.
Concurrent validity	This is the correlation between the measurement scale with some other measure of the same trait, ideally the "gold standard". For example, it would be expected the Y-BOCS would be correlated with the previous most widely accepted OCD measurement tool.
Divergent validity	Also known as discriminant validity, this is an assessment to test whether the measure differs (i.e. does not correlate) with unrelated measures. For example, whether the Y-BOCS measure, as a measure of OCD, does not correlate with measures of anxiety or depression.

6.1. Method

A narrative synthesis was undertaken of the data yielded following the search strategy outlined below. The approach to the synthesis was followed as recommended by Popay et al. (2006), as outlined in section 3.3. The search terms used to extract the data from the studies is detailed in Table 6.2. The critical appraisal questions, as recommended by Aveyard (2010, p. 103), used to assess the robustness of the studies is described in Tables 6.6 and 6.7.

6.1.1. Research aim and questions.

The aim of the systematic literature review was to identify the psychometric properties of OCD and Repetitive Behaviour measurement tools, in order to inform a decision regarding the most appropriate tools to use for the empirical investigation.

The research questions related to this study were:

1. Which is the most robust OCD measurement tool, based on available evidence of psychometric properties?
2. Which is the most robust Repetitive Behaviour measurement tool, based on available evidence of psychometric properties?

6.1.2. Search strategy

To include studies from a large pool across clinical psychology research, articles were retrieved using EBSCO (PsycINFO, PsycARTICLES, MEDLINE and CINAHL Complete) for published articles related to the above questions. Limiters were placed to ensure studies of adult participants were identified, except for Repetitive Behaviour studies, which are predominantly based on samples of child participants.

6.1.3. Inclusion criteria

Articles were included where the sample consisted of any of the three groups from the overall investigation in this thesis i.e. adults with autism, OCD or neurotypical controls. Included studies were those which investigated any of the psychometric properties as outlined in Table 6.1, in OCD or Repetitive Behaviour assessment tools.

6.1.4. Exclusion criteria

Studies were excluded if they fell into the following criteria:

- Participants were below the age of 18 years (as they are not within the adult age range of the final empirical investigation);
- Single case studies, or studies involving fewer than 10 participants, due to reduced reliability of such evidence when generalising to the wider population.
- Indirect reference to measuring psychometric properties of these measurement tools i.e. the study did not direct measure any of the psychometric terms as outlined in Table 6.1 using the measurement tools identified.
- Participants with intellectual disabilities/learning disabilities, due issues related to comorbidity (i.e. difficulty in attributing the repetitive traits specifically to a specific condition).

Studies of cross-cultural measures were not included for OCD or Repetitive Behaviour measures, with the exception of the Y-BOCS. This approach was taken on grounds of brevity and clarity in the narrative review, with such a high proportion of studies investigating cross-cultural assessment (of OCD specifically). For example, whilst only 10 studies were finally included for the Obsessive-Compulsive Inventory-Revised, a

further 21 cross-cultural studies were excluded. However, cross-cultural psychometric studies of the Y-BOCS were not excluded. This was based on the decision to be comprehensive for the Y-BOCS, as the identified "gold standard" tool.

6.1.5. Search terms

Search terms as described in Table 6.2 were used, with the final search being completed on 14th May 2019.

Table 6.2. *Search terms used for main systematic search using PsycINFO, PsycARTICLES, MEDLINE and CINAHL Complete.*

Search number	Any of the following terms for measurement tools as "abstract"	Combined with "AND"	The following term for focus as "abstract"
1	YBOC* or yale brown obsessive compulsive scale or padua inventory or Leyton Obsessional Inventory or Maudsley Obsessive Compulsive Inventory or Florida obsessive compulsive inventory or Obsessive Compulsive Inventory or Global Obsessive Compulsive Scale or Vancouver Obsessional Compulsive Inventory		psychometrics or validity or reliability
2	repetitive traits or repetitive behav* or RB or repetitive behaviour scale or repetitive behavior scale or repetitive behaviour questionnaire or repetitive behavior or child routines inventory or repetitive and restricted behaviour scale or repetitive and restricted behavior scale		Psychometric or validity or reliability or consistency

* indicates the word was truncated to allow for multiple word endings
Nb. Special limiters placed for PsycARTICLES: all articles for participants aged 18 years and over; human; exclude book reviews; exclude non-article content. Special limiters placed for CINAHL Complete: human; English language; all adult. Special limiters placed for MEDLINE: human; English language; all adult. Special limiters placed on PsycINFO: all articles for participants aged 18 years and over; English language; human.

6.1.6. Selection strategy

The abstracts of all yielded articles identified were screened for relevance against the inclusion and exclusion criteria outlined in sections 6.1.3 and 6.1.4.

6.2. Data Analysis and synthesis

Relevant psychometric properties, as defined by Streiner and Norman (2008), are outlined in Table 6.1 and reported from the yielded studies from the systematic search.

A summary of the OCD and Repetitive Behaviour instruments identified are presented in Tables 6.6 and 6.7. The instruments selected for review are summarised in Tables 6.3 and 6.4. The characteristics of the studies which examined the psychometric properties of the instruments are presented in Tables 6.8 and 6.9.

Twenty-six articles were included, consisting of 19 for OCD measurement tools and seven for Repetitive Behaviour measurement tools, as outlined in the study selection flow charts (Figures 6.1 and 6.2), in addition to one additional Repetitive Behaviour Tool study. The additional study (Moss, Oliver, Arron, Burbidge, & Berg, 2009) was not yielded through this search (as in 6.1.5), but was included as it was the study which the Repetitive Behaviour Questionnaire was originally reported; the Repetitive Behaviour Questionnaire (Moss et al., 2009) being the one used in the final empirical investigation in Chapter 7 (for reasons stated in 7.1.3). For the OCD studies, this consisted of 9467 reported participants: 7615 neurotypical participants, 1278 with OCD, 251 with other disorder (mostly anxiety disorder), 152 with comorbid OCD and autism and 171 with an autism diagnosis. Notably, following the exclusion criteria as described in section 6.1.4, no articles were yielded for the Global Obsessive Compulsive Scale or the Maudsley Obsessive Compulsive Inventory. For the Repetitive Behaviour studies, this consisted of 1797 participants: 1112 autistic (275 adults and

837 children); 685 neurotypical adults (36 as a control group and 649 as a separate validation in two undergraduate samples).

A narrative synthesis (as recommended by Popay et al., 2006) was used to report the results as this method has been recommended to be the most appropriate to compare across the range of different outcome meausres, with too few data from any subset for quantitative analysis (see Tables 6.10 and 6.11). This approach taken in the narrative synthesis is outlined in section 3.3. As recommended by Popay et al. (2006), grouping characteristics were identified to facilitate the narrative synthesis (see Table 6.5). These groupings indicated studies were best described within either “focus of report” or “measurement tools”. The decision was made to group studies according to measurement tools, as this would give the most theoretic clarity within the review. The following narrative synthesis of the systematic review evidence has been guided by Popay et al. (2006), as outlined in section 3.3.

Table 6.3. *Obsessive Compulsive Disorder assessment tools overview.*

Instrument (reference)	**Purpose**	**Type of measure**	**Subscales**	**Number of items**	**Response Options**
Florida Obsessive-Compulsive Inventory (Storch et al., 2007)	Tool to create a quick assessment of symptom type and severity in a self-report format.	Self-report checklist	Symptom checklist and symptom severity.	20 (checklist)	Checklist (yes/no), followed by 5-pont Likert scale for the subscales.
Leyton Obsessional Inventory (Cooper, 1970).	To assess anxiety symptoms and treatment outcomes in patients with OCD.	Clinician administered.	4 subscales: Symptom; Trait; Resistance; and Interference. Each scores for individual items.	69 (46 for obsessive symptoms and 23 for obsessive personality traits).	Yes/no response to items, followed by selection of one of 4 statements for affirmed items, relating to resistance and interference of the symptoms.
Obsessive Compulsive Inventory-Revised (Foa et al., 2002)	Adaptation of the original OCI, which was developed as a self-report measure of severity of OCD symptoms.	Self-report measure	7 subscales: washing; checking; doubting; ordering; obsessing; hoarding; and mental neutralising.	18	Each item rated on a 5-point Likert Scale of symptom distress.
Padua Inventory (Sanavio, 1988)	Created as a self-report measure of OCD.	Self-report measure	Various reported, including contamination, checking, mental control and impulses.	60	5-point Likert scale for degree of disturbance.
Vancouver Obsessional Compulsive Inventory (Thordarson et al., 2004)	Revision of Maudsley Obsessional Compulsive Inventory, created to assess a greater range of OCD symptoms and make administration, scoring and interpretation easier (no double scoring of frequency and distress, such as in the OCI-R)	Self-report measure	6 subscales: contamination; checking; obsessions; hoarding; just right; and indecisiveness.	55	Each item rated on a 5-point Likert Scale.

Instrument (reference)	Purpose	Type of measure	Subscales	Number of items	Response Options
Yale-Brown Obsessive Compulsive Scale: Y-BOCS (Goodman et al., 1989b)	To assess clinical change of OCD traits.	Clinician administered	Obsessions and Compulsions subscales. Items for each subscale: distress; time; functional interference; efforts to resist; and control.	58	Checklist (current/past), followed by 5-pont Likert scale for the subscales.
Y-BOCS Symptom Checklist (Goodman et al., 1989b)	The subscales from the original Y-BOCS have been dropped, with the symptom checklist retained for ease of administration.	Clinician-administered	None	58	Checklist (current/past).
Y-BOCS Self Report (Y-BOCS-SR: Baer et al., 1992)	An identical format of the Y-BOCS created procedurally for self-administration.	Self-administered	Obsessions and Compulsions subscales. Items for each subscale: distress; time; functional interference; and control.	58	Checklist (current/past). Participants circle the main there obsessions and three main compulsions then use 6-point Likert scale for the subscales.
Y-BOCS-II (Storch et al., 2010)	Improved version of the Y-BOCS.	Clinician administered	Obsessions and Compulsions subscales. Items for each subscale: distress; time; functional interference; and control.	58	Checklist (current/past), followed by 6-point Likert scale for the subscales.
Y-BOCS-II Self Report (Y-BOCS-II-SR: Hiranyatheb et al., 2015)	An identical format of the Y-BOCS-II created procedurally for self-administration.	Self-administered	Obsessions and Compulsions subscales. Items for each subscale: distress; time; functional interference; and control.	58	Checklist (current/past), followed by 6-point Likert scale for the subscales (for the main three obsessions and three main compulsions).
Y-BOCS-II Symptom Checklist (Storch et al., 2010a)	To create a symptom checklist version (no subscale) of the Y-BOCS-II.	Clinician-administered	None.	58	Checklist (current/past), followed by 6-point Likert scale for the subscales.

Table 6.4. Repetitive Behaviour assessment tools overview.

Instrument (published year)	**Purpose**	**Type of measure**	**Subscales**	**Number of items**	**Response Options**
Autism Behavior Inventory (Bangerter et al., 2017)	A web-based tool to measure change of autistic symptoms (repetitive behaviours just one scale of the overall assessment).	Informant (e.g. parent) reported.	To core autism domains (restricted and repetitive behaviours; social communication) and three associated domains (mental health; self-regulation; challenging behaviour).	73 full version and 35 short version.	7 point Likert scale for each item, ranging from 0 = absence of symptoms to 6 = maximum symptoms.
Repetitive Behaviour Scale-Revised (Bodfish et al, 2000)	An assessment tool aimed at examining the phenomenology of repetitive behaviours in autism.	Clinician administered.	Conceptually grouped into 6 subscales: stereotyped behaviour; self-injurious behaviour; compulsive behaviour (i.e. according to rule or "just so"); ritualistic behaviour (daily living activity in similar manner); Sameness behaviour; and Restricted Behaviour.	43	4-point Likert Scale, ranging from "behaviour does not occur" to "behaviour occurs and is a severe problem".
Repetitive Behaviour Questionnaire (Moss, Oliver, Arron, Burbidge & Berg, 2009)	An assessment tool aimed at examining the phenomenology of repetitive traits in neurological disorders.	Informant (e.g. parent) reported.	5 subscales: stereotyped behaviour; compulsive behaviour; insistence on sameness; restricted preferences; and repetitive speech.	19	5 point Likert scale, ranging from "never" to "more than once a day" in reference to frequency of each trait over the preceding month.
Repetitive Behaviour Questionnaire-2 Adults (Barrett, Uljarević, Baker, Richdale, Jones & Leekam, 2015)	To provide a self-report repetitive behaviour questionnaire suitable for adults with autism	Self-report.	2 factors identified: insistence on sameness; and repetitive motor behaviours. Possibility of sensory items as additional factor.	20	Both 4-point Likert Scale or 3-point Likert Scale used across items. Scales based on both frequency and severity e.g. 1) never or rarely; 2) mild or occasional/one or more times daily; 3) marked or notable/15 or more times daily 4) serious or severe/30 or more times daily.

Table 6.5. *Grouping characteristics example for main systematic literature review.*

Group according to:	
Measurement tools	Focus of report
Yale-Brown Obsessive Compulsive Scale (Arrindell, de Vlaming, Eisenhardt, van Berkum, & Kwee, 2000; Goodman et al., 1989b; Moritz, Quaquebeke, Hauschildlt, Jelinek, & Gönner, 2012; Richter, Cox, & Direnfeld, 1994; Storch et al., 2007)	Cross cultural (Arrindell et al., 2000; Moritz et al., 2012)
Yale-Brown Obsessive Compulsive Scale-Symptom Checklist (Mataix-Cols, Fullana, Alonso, Menchón, & Vallejo, 2004; Sulkowski et al., 2008)	Self-report vs. clinician (Moritz et al., 2012; Hiranyatheb et al. 2015; Moritz et al., 2012; Steketee et al., 1996)
Yale-Brown Obsessive Compulsive Scale-II (Storch et al., 2010a)	Taking out scale (Mataix-Cols et al., 2004; Sulkowski et al., 2008; Storch et al., 2010a)
Yale-Brown Obsessive Compulsive Scale-II Symptom Checklist (Storch et al., 2010a; Wu, McGuire, Horng, & Storch, 2016)	Tool comparison (Richter et al., 1994; Storch et al., 2007)
Yale-Brown Obsessive Compulsive Scale-II Self Report (Y-BOCS-II-SR: Hiranyatheb et al. 2015)	Y-BOCS-II (Storch et al., 2010a; Storch et al., 2010a; Wu et al., 2016)
Yale-Brown Obsessive Compulsive Scale-II Symptom Checklist (Y-BOCS-II-SC: Storch et al., 2010a)	Y-BOCS-II SR (Hiranyatheb et al. 2015)
Y-BOCS Self Report (Steketee, Frost, & Bogart, 1996)	Repetitive Behaviour Scale-Revised (Bodfish et al, 2000)
Repetitive Behaviour Scale-Revised (Bodfish et al., 2000)	Repetitive Behaviour Questionnaire-2 (Barrett et al., 2018)
Repetitive Behaviour Questionnaire-2 (Barrett, Uljarević, Jones, & Leekam, 2018)	Autism Behavior Inventory (Bangerter et al., 2017)

6.3. Results

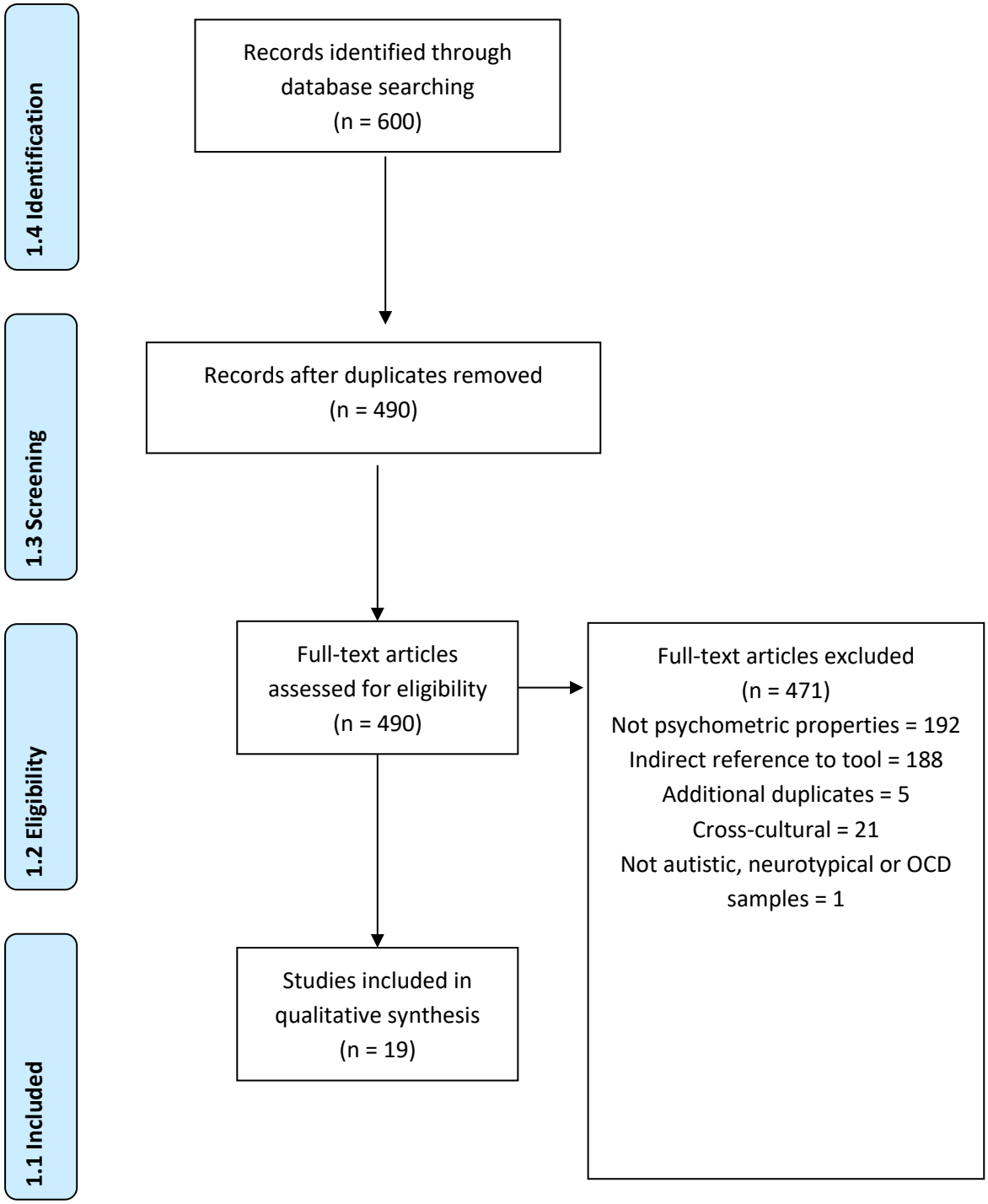

Figure 6.1. PRISMA (Moher et al. 2009) study selection flow chart for search 1: psychometric property studies of OCD measurement tools in adult participants.

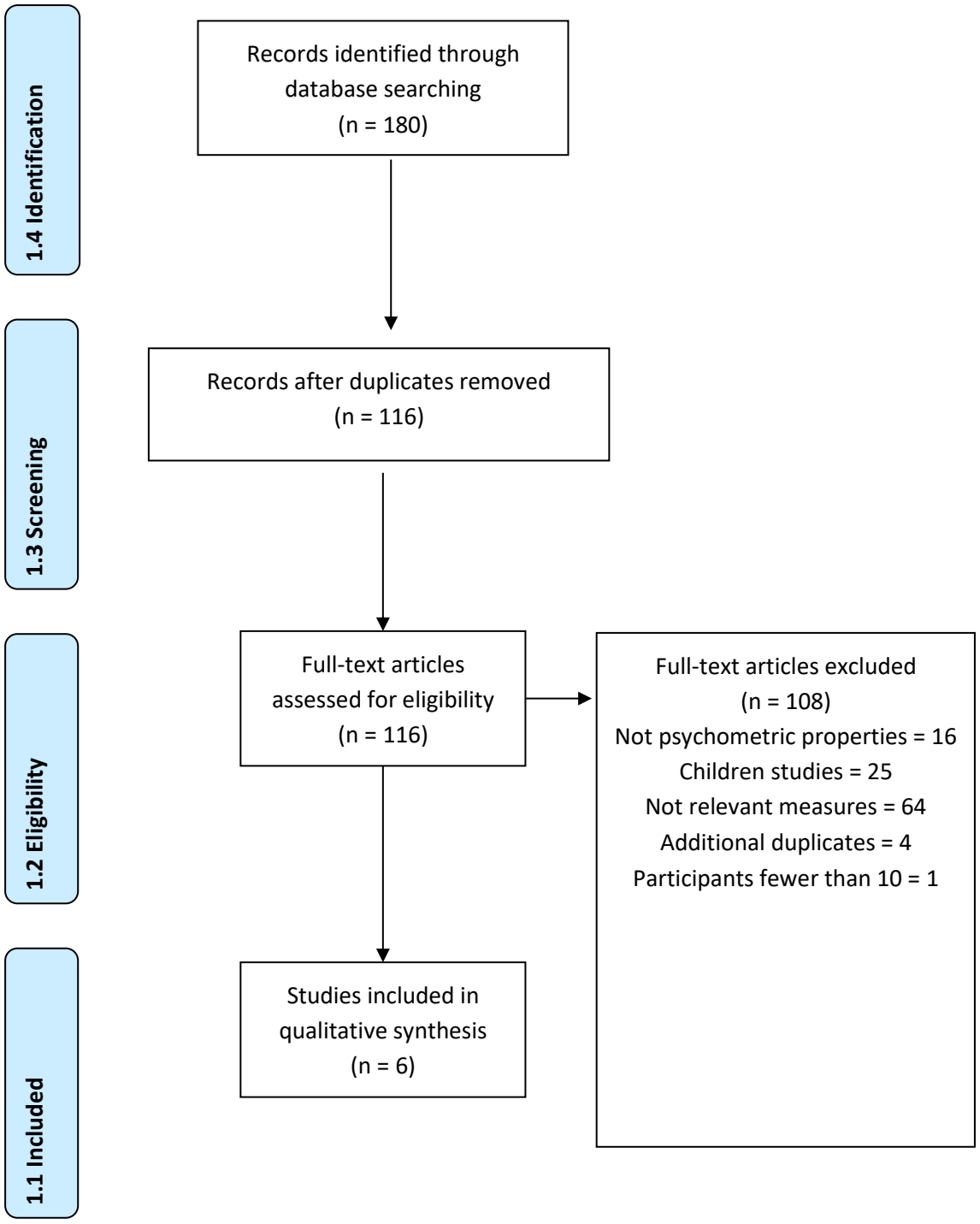

Figure 6.2. PRISMA (Moher et al. 2009) study selection flow chart for search 2: repetitive behaviour measurement psychometric property studies in adult participants.

Table 6.6. *Critical appraisal questions for OCD psychometric studies, using recommendations by Aveyard (2010, p103).*

Study	**Journal quality**	**Clear research question, appropriate for research?**	**Valid research method?**	**Sufficiently large sample?**	**Appropriate sample?**	**Data collection appropriate to the research method?**	**Appropriate statistical test?**	**Quality – overall appraisal**
Abramowitz and Deacon (2006)	Good. Peer-review with impact factor 3.105	Yes.	Yes.	Yes.	Yes, good inclusion and exclusion criteria and screening procedures.	Yes.	Yes.	High.
Arrindell et al. (2000)	Good. Peer-review with impact factor 2.397	Yes.	Yes.	Yes.	Probably. Non-exclusion of patients with other anxiety disorders increases generalisability. However, difficult to determine whether lack of subgroup membership (to OCD) affects validity in generalising to wider population. Also, recruitment procedures not stated, so difficult to discount any biases.	Yes.	Yes.	High.
Burns, et al. (1996)	Good. Peer-review with impact factor 4.134	Yes.	Yes.	Yes, huge number of participants. However, no clinical groups.	Not entirely. No clinical participants, but it was a psychometric validity study in a neurotypical sample. However, it was all self-report with no screening or exclusion criteria stated.	Yes.	Yes.	Medium to high.
Cadman et al. (2015)	Good. Peer-review with impact factor 4.532	Yes.	Yes.	Yes, across all clinical groups.	Yes. Good screening, confirmation of diagnoses and exclusion criteria.	Yes.	Yes.	High.

Study	Journal quality	Clear research question, appropriate for research?	Valid research method?	Sufficiently large sample?	Appropriate sample?	Data collection appropriate to the research method?	Appropriate statistical test?	Quality – overall appraisal
Goodman et al. (1989a)	Very good. Peer-review with impact factor 15.916	Yes.	Yes.	Yes.	Mostly, not a comprehensive list of exclusion criteria states and no screening for other disorders.	Yes.	Yes.	High.
Hajcak et al. (2004)	Good. Peer-review with impact factor 4.134	Yes.	Yes, but would benefit from recruitment of clinical (OCD) comparative sample.	Yes.	Not entirely. No clinical participants, but it was a psychometric validity study in a neurotypical sample. However, it was all self-report with no screening or exclusion criteria stated. Also not clear – no demographic information collected other than gender.	Yes.	Yes.	Medium.
Hiranyatheb et al. (2015)	Good. Peer-review with impact factor 1.741	Yes.	Yes.	Relatively small number for the psychometric analyses.	Yes. Good exclusion criteria, including intellectual disability, severe psychosis and mental disorder. However, recruitment procedures not stated, so difficult to discount any biases.	Yes.	Yes.	High.
MacDonald and de Silva (1999)	Good. Peer-review with impact factor 1.967	Yes.	Yes.	Yes, very large number.	Not entirely. No clinical participants, but it was a psychometric validity study in a neurotypical twin sample. However, it was all self-report with no screening or exclusion criteria stated.	Yes.	Yes.	Medium to high.

Study	Journal quality	Clear research question, appropriate for research?	Valid research method?	Sufficiently large sample?	Appropriate sample?	Data collection appropriate to the research method?	Appropriate statistical test?	Quality – overall appraisal
Mataix-Cols et al. (2004)	Good. Peer-review with impact factor 7.632	Yes.	Yes.	Yes.	Yes. Good inclusion and exclusion criteria. However, recruitment methods not stated.	Yes.	Yes.	High.
Moritz et al. (2012)	Good. Peer-review with impact factor 1.230	Yes.	Yes.	Relatively medium size groups for the subgroups.	Yes. Good characteristics across three groups. Good inclusion and exclusion criteria, plus screening. Recruitment in line with type of study (online) and good use of random allocation.	Yes.	Unclear statistical analyses for the correlations.	Medium to high.
Stanley et al. (1993)	Good. Peer-review with impact factor 3.371	Yes.	Yes.	Yes.	Potentially not. High proportion (77%) of comorbidity with other Axis 1 disorders. This may be representative of wider population, but does not portray "pure OCD". Good screening for anxiety caused by other origin and also medication use.	Yes.	Yes.	Medium.
Steketee et al. (1996)	Good. Peer-review with impact factor 4.134	Yes.	Yes.	Mostly, although the comparison clinical sample was small (*n* = 10).	Mostly: exclusion criteria not thorough in the clinical sample, neither was the recruitment procedures in the group.	Yes.	Yes.	Medium to high.

Study	Journal quality	Clear research question, appropriate for research?	Valid research method?	Sufficiently large sample?	Appropriate sample?	Data collection appropriate to the research method?	Appropriate statistical test?	Quality – overall appraisal
Storch et al. (2007)	Good. Peer-review with impact factor 2.116	Yes.	Yes.	Yes.	Yes. Good inclusion and exclusion criteria. However, recruitment methods not stated.	Yes.	Yes.	High.
Storch et al. (2010b)	Good. Peer-review with impact factor 3.105	Yes.	Yes.	Yes.	Yes, good screening/diagnosis and exclusion criteria, but recruitment methods not stated.	Yes.	Yes.	High
Sulkowski et al. (2008)	Good. Peer-review with impact factor 2.116	Yes.	Yes.	Yes.	Yes. Good inclusion and exclusion criteria. However, recruitment methods not stated.	Yes.	Yes.	High.
Thordarson et al. (2004)	Good. Peer-review with impact factor 4.134	Yes.	Yes.	Yes, although small for the factor analysis.	Mostly – 9% OCD wasn't primary diagnosis. 52% with comorbid Axis I disorder, therefore not "pure" OCD, but may be representative of wider population.	Yes.	Yes.	Medium to high.
Wu et al. (2016)	Good. Peer-review with impact factor 2.128	Yes.	Yes.	Yes.	Unclear – no inclusion/exclusion criteria, no screening or diagnosis, plus no recruitment methods stated.	Yes.	Yes.	Medium to high.

Table 6.7. *Critical appraisal questions for Repetitive Behaviour measures studies using recommendations by Aveyard (2010, p103).*

Study	**Journal quality**	**Clear research question, appropriate for research?**	**Valid research method?**	**Sufficiently large sample?**	**Appropriate sample?**	**Data collection appropriate to the research method?**	**Appropriate statistical test?**	**Quality – overall appraisal**
Bangerter et al. (2017)	Good. Peer-review with impact factor 2.901	Yes.	Yes.	Yes for phase 1, small sample (*n* = 23) for phase 2 (psychometric properties).	Unknown due to unclear recruitment, sampling and screening procedures. No exclusion criteria stated. Self-reporting (thought autism confirmed in phase 2) and lack of important demographic information such as IQ or comorbid disorders.	Yes.	Yes.	Low to medium.
Barrett et al. (2018)	Good. Peer-review with impact factor 5.712	Yes.	Yes.	Yes – large sample of 275 participants.	Mostly. Opportunity sample. Screened via the Autism Quotient, although otherwise self-reported and no exclusion criteria stated.	Yes.	Yes.	Medium to high.
Inada et al. (2015)	Good. Peer-review with impact factor 2.907	Yes.	Yes.	Yes, still suitably large when accounting for subgroup membership.	Mostly – randomly selected and diagnosed, but no exclusion criteria stated.	Yes.	Yes.	High.
Jia, et al. (2019)	Good. Peer-review with impact factor 3.341.	Yes.	Yes.	Yes.	Unclear – all non-clinical undergraduate students, however all self-reported (no screening) and no exclusion criteria stated.	Yes.	Yes.	Medium.

Study	Journal quality	Clear research question, appropriate for research?	Valid research method?	Sufficiently large sample?	Appropriate sample?	Data collection appropriate to the research method?	Appropriate statistical test?	Quality – overall appraisal
Lam and Aman (2007)	Good. Peer-review with impact factor 3.341.	Yes.	Yes.	Unclear - the demographic information isn't clear enough to ascertain whether non ID enough large for this research question (i.e. 54.4% of overall total intellectual disability)	Possibly, intellectual disability confound may be higher than data appears i.e. whilst 54.4% of the overall total, only 16.6% of sample attended "regular class in regular school". Also, there doesn't appear to be any exclusion criteria for the sample, which may further confound results.	Yes.	Mostly, although it would benefit from more partialing out of children and adult samples, as well as intellectual disorder.	Medium.
Martínez-González and Piqueras (2018)	Good. Peer-review with impact factor 3.341.	Yes.	Yes.	Yes.	Unclear – intellectual disability likely to be a confounding variable. No exclusion or screening stated.	Yes.	Mostly, although it would benefit from more partialling out of children and adult samples, as well as intellectual disorder.	Medium.
Moss et al. (2009)	Good. Peer-review with impact factor 3.341.	Yes.	Yes.	Yes.	No. Issues regarding likely comorbidity of neurological disorders and intellectual disability. Also, no appropriate autism group recruited (some analyses for example were not calculated for autism participants as only two were reported to be verbal; p.581).	Yes.	Mostly, although it would benefit from more partialling out of children and adult samples, as well as intellectual disorder.	Medium.

Table 6.8. Characteristics of included studies for OCD psychometric studies.

Reference	Instrument	Design	Purpose of study	Participants	Quality appraisal
Storch et al. (2007)	Florida Obsessive-Compulsive Inventory	Cross-sectional questionnaire within-subjects design.	To analyse the following psychometric properties: internal consistency; convergent validity (against the Y-BOCS); divergent validity (against measures of depression and global severity of psychopathology)	113 outpatients with diagnosis of OCD (53.1% females; mean age 33.8 years, SD = 11.5).	High.
Stanley et al. (1993)	Leyton Obsessional Inventory	Cross-sectional questionnaire, between- and within-subjects design.	To evaluate psychometric properties, consisting of: internal consistency of 4 subscales; divergent validity (against measures of psychopathy and personality traits); and discriminant validity between the subscales.	77 patients with DSM-III diagnosed OCD. 18 participants with single (OCD) diagnosis, 59 with additional Axis I disorder. 74% female, mean age 37.5 years (SD = 0.8).	Medium.
Richter et al. (1994)		Cross-sectional questionnaire, within-subjects design.	To test convergent validity (against Y-BOCS and Maudsley Obsessional-Compulsive Inventory).	30 consecutive OCD diagnosed outpatients (36.7% female; mean age 33.8 years, SD = 9.2).	Medium.
Abramowitz and Deacon (2006)	Obsessive Compulsive Inventory-Revised (OCI-R)	Cross-sectional questionnaire, between- and within-subjects design.	To test internal consistency and divergent validity (against measures of anxiety and depression).	322 patients given a diagnosis of anxiety disorder (N = 167 OCD). Mean age of overall sample was 36.5 years (SD = 13.0). 95% Caucasian. None of the anxiety disorder participant has OCD diagnosis.	High.

Reference	Instrument	Design	Purpose of study	Participants	Quality appraisal
Cadman et al. (2015)	Obsessive Compulsive Inventory-Revised (OCI-R)	Cross-sectional questionnaire, between- and within-subjects design.	To analyse symptom profile of adults with autism, OCD and comorbid autism + OCD using OCI-R. Psychometric properties of OCI-R analysed, consisting of internal consistency and convergent validity (correlation between subscales).	152 participants with comorbid OCD and autism (19% female, mean age 31.19 and SD = 11.41, verbal IQ 100.66 and SD = 15.43). 108 participants with OCD (54% female, mean age 39.60 and SD = 11.69). 171 participants with only autism (11% female, mean age27.99 and SD = 9.07, verbal IQ 106.33 and SD = 15.31). 92 neurotypical controls (0% female, mean age 29.35 and SD = 7.29, verbal IQ 108.85 and SD = 13.30).	High.
Hajcak et al. (2004)		Cross-sectional questionnaire, within-subjects design.	To analyse psychometric properties of test-retest validity (1 month apart), convergent validity (against MOCI) and divergent validity (against measures of worry and depression)	395 undergraduate students (61.3% females) and no further demographic information.	Medium.
Moritz et al. (2012)		Cross-sectional questionnaire within-subjects design.	To test convergent validity (against Y-BOCS) in the measure in a cross-cultural (German) population, delivered online. Other purposes for the Y-BOCS psychometric assessment.	66 German participants with confirmed OCD (61.61% females; mean age 40.45 years – SD = 10.940; 86 with probable OCD (32.56% male; mean age 34.52 years – SD = 10.66); and 121 clinical experts (30.58% male; mean age 29.78 years – SD = 7.37) as controls to pretend they had OCD.	Medium to high – mainly due to the lack of clarity on the correlational analyses.

Reference	Instrument	Design	Purpose of study	Participants	Quality appraisal
Burns et al. (1996)	Padua Inventory (PI)	Cross-sectional questionnaire, between- and within-subjects design.	To test discriminant validity (against measure of worry and PI subscales) and test-retest reliability (between a 6 to 7 month interval).	5010 individuals. 2970 completed the PI and the Penn State Worry Questionnaire (55.3% female, mean age 19.34 years and SD = 2.29, 88.6% Caucasian).	Medium to high.
MacDonald and de Silva (1999)		Cross-sectional questionnaire, within-subjects design.	To test internal reliability and discriminant validity (against measures of personality).	1855 participants from twin studies. Age range 15-75 years (mean and standard deviation not stated). 84.5% females.	Medium to high.
Thordarson et al. (2004)	Vancouver Obsessional-Compulsive Inventory	Cross-sectional questionnaire, between- and within-subjects design.	To test various psychometric properties, consisting of test-retest reliability, internal consistency, discriminant validity (against measures of depression, anxiety, worry and personality traits), convergent validity (against Y-BOCS, Y-BOCS SR, Maudsley Obsessional Impulsive Inventory, Padua Inventory)	88 adults with OCD (91% as primary diagnosis). 63% female, mean age 35.3 years. 52% comorbid Axis 1 disorder. Anxiety/depression group of 60 participants, 60% female, mean age of 36.0 years. Neurotypical control groups of 39 community adults and 200 students (self-reported, not screened), 64% and 69% female and mean age 41.0 and 19.9 years, respectively.	Medium to high.

Reference	Instrument	Design	Purpose of study	Participants	Quality appraisal
Arrindell et al. (2000)	Yale-Brown Obsessive-Compulsive Scale (Y-BOCS).	Cross-sectional questionnaire, within-subjects design.	To analyse the following psychometric properties: cross-cultural validity; inter-rater agreement; concurrent validity; and divergent validity (with measures of anxiety and depression).	65 psychiatric inpatients in Netherlands, mean age 34 years (SD = 9), 69.2% females. 60% of sample with confirmed OCD diagnosis, with additional comorbidities (largest = 20% social phobia/panic disorder then dysthymia at 6.2%). 4 of this OCD sample randomly selected for inter-rater reliability.	High.
Goodman et al. (1989a)		Cross-sectional questionnaire within-subjects design.	To analyse convergent (against the Global Obsessive Compulsive Scale and the Maudsley Obsessional Compulsive Scale) and discriminant validity (against measures of depression and anxiety via the Hamilton Rating Scale for Depression and Anxiety) of the newly created Y-BOCS.	81 OCD patients in all. 3 groups: 16 in fluvoxamine group (75% female; mean age 36 years, SD = 12; "most patients depressed"); 45 in fluvoxamine group (53.3% female; mean age 38, SD = 10; 50% depressed; and 20 in clomipramine group (35% female; mean age 24 years, SD = 11; 0% depressed).	High.
Richter et al. (1994)		Cross-sectional questionnaire within-subjects design.	To test internal consistency, discriminant validity (against (Hamilton Rating Scale for Depression) and convergent validity (against Leyton Obsessional Inventory and Maudsley Obsessional-Compulsive Inventory).	30 consecutive OCD diagnosed outpatients (36.7% female; mean age 33.8 years, SD = 9.2).	Medium.
Storch et al. (2007)		Cross-sectional questionnaire within-subjects design.	To test the concurrent validity of the measure between the Florida Obsessive-Compulsive Inventory and the Y-BOCS.	113 outpatients with diagnosis of OCD (53.1% females; mean age 33.8 years, SD = 11.5).	High.

Reference	Instrument	Design	Purpose of study	Participants	Quality appraisal
Moritz et al. (2012)	Y-BOCS self-report	Cross-sectional questionnaire between subjects design.	To test assessment psychometric properties (test-retest, internal consistency, external and discriminant validity) in a cross-cultural (German) population, delivered online.	66 German participants with confirmed OCD (61.61% females; mean age 40.45 years – SD = 10.94; 86 with probable OCD (32.56% male; mean age 34.52 years – SD = 10.66); and 121 clinical experts (30.58% male; mean age 29.78 years – SD = 7.37) as controls to pretend they had OCD.	Medium to high – mainly due to the lack of clarity on the correlational analyses.
Steketee et al. (1996)		Cross-sectional questionnaire between subjects design.	To test the psychometric properties (test-retest, internal consistency, convergent validity) between self-report and clinician interview.	4 groups of participants: 46 non-clinical received either self-report Y-BOCS or interview format (age range 17-21 years; 100% female; 26% non-Caucasian); 70 additional non-clinical received both formats, counterbalanced in half for order effects (age range 17-22 years; 100% female); 36 treatment-seeking OCD confirmed group received both formats counterbalanced (55.56% females; mean age); and 10 treatment-seeking clinical non-OCD group (70% female; mean age 44.5 years).	Medium to high.
Mataix-Cols et al. (2004)	Y-BOCS Symptom Checklist.	Cross-sectional within-subjects design.	To test the convergent validity (against the Maudsley Obsessional Compulsive Inventory and the Padua Inventory) and discriminant validity (against State-Trait Anxiety Inventory, Beck Depression Inventory and Hamilton Rating Scale for Depression).	56 Spanish patients with primary confirmed diagnosis of OCD (41% female; mean age 28.3 years, SD = 8). Patients were treatment-seeking (54.7% were already receiving SSRIs and 51% benzodiazepines). 22% fulfilled criteria for depression, post-OCD diagnosis in all cases.	High.
Sulkowski et al. (2008)		Cross-sectional questionnaire within-subjects design.	To test convergent validity (against Obsessive Compulsive Inventory-Revised) and discriminant validity (against measures of anxiety and depression).	112 adults diagnosed with OCD (51% male; mean age 30.43 years, SD = 11.38) as a primary disorder.	High.

Reference	Instrument	Design	Purpose of study	Participants	Quality appraisal
Storch et al. (2010a)	Y-BOCS-II.	Cross-sectional questionnaire within-subjects design.	To test the following psychometric properties: internal consistency; construct validity; exploratory and confirmatory factor analysis; test-retest reliability; inter-rater reliability; convergent validity (against Obsessive Compulsive Inventory-Revised and Clinical Global Impression-Severity; and) discriminative validity (against Inventory of Depressive Symptomology-Self Report and Penn State Worry Questionnaire).	130 OCD confirmed clinic patients (49% female; mean age 31.76 years, SD = 12.70).	High.
Hiranyatheb et al. (2015)	Y-BOCS-II SR	Cross-sectional questionnaire within subjects design.	To analyse the following psychometric properties in a cross-cultural sample (Thai): internal consistency; convergent validity (against Florida Obsessive-Compulsive Inventory); and divergent validity (against measures of depression and quality of life).	52 Thai outpatients with confirmed OCD diagnoses (45.2% females; mean age 37 years, SD = 16.52).	Medium to high. (based on relatively small number of participants).
Storch et al. (2010b)	Y-BOCS-II-Symptom Checklist (SC).	Cross-sectional questionnaire within-subjects design.	To test the following psychometric properties: factor structure; internal consistency; inter-rater reliability; test-retest reliability; convergent (against Obsessive Compulsive Inventory-Revised) and divergent validity (against measures of anxiety and depression).	130 OCD confirmed clinic patients (49% female; mean age 31.76 years, SD = 12.70).	High.
Wu et al. (2016)		Cross-sectional questionnaire within-subjects design	To test the following psychometric properties: internal consistency; inter-rater reliability; test-retest reliability; convergent validity; and divergent validity (against measures of anxiety, depression and impulsiveness).	61 outpatients with principal OCD diagnosis (56% female; mean age 35.27 years, SD = 14.91). 57% were enrolled in medication-treatment and 31% in cognitive-behavioural therapy.	Medium to high.

Table 6.9. *Characteristics of included studies for Repetitive Behaviour psychometric studies.*

Reference	**Instrument**	**Design**	**Purpose of study**	**Participants**	**Quality appraisal**
Bangerter et al. (2017)	Autism Behavior Inventory	Cross-sectional within-subjects design.	To examine the psychometric properties: test-retest reliability and concurrent validity (between subscales).	23 participants in psychometric measures study (mean age 10 years, SD = 5.3; 8.70% female; 95.7% Caucasian).	Low to medium.
Inada et al. (2015)	Repetitive Behaviour Scale-Revised (RBS-R)	Cross-sectional within-and between-subjects design.	To analyse internal consistency and discriminant validity (against autistic symptoms, adaptive functioning, aberrant behaviour and sensory processing).	274 children with autism (mean age 15.0, SD = 6.3; 20.1% female; mean IQ = 80.3, SD = 30.0; 35% without intellectual disabilities). 36 neurotypical controls (mean age 13.1, SD = 7.3; 33.3% female; mean IQ = 66.0, SD = 31.7; 19.4% without intellectual disabilities).	High.
Lam and Aman (2007)		Cross-sectional within-subjects design.	To assess validity of RBS-R: internal consistency and inter-rater reliability.	307 people with autism (self- and informant-reported). Mean age 15.34 years (SD = 9.60), 16.9% females, 23.1% African American. 81.4% reported diagnosis of autism. 45.6% reported no intellectual disabilities.	Medium.
Martínez-González and Piqueras (2018)		Cross-sectional within-subjects design.	To examine the psychometric properties: internal consistency; test-retest reliability (after 1 month); concurrent validity (between subscales); divergent validity (social interaction, emotion regulation, social cognitive performance).	233 with autism recruited from appropriate centres. Age 13.34 years (SD = 9.79). 22.3% female. 65.3% without intellectual disabilities.	Medium.

Reference	Instrument	Design	Purpose of study	Participants	Quality appraisal
Moss et al. (2009)	Repetitive Behaviour Questionnaire	Cross-sectional between-subjects design.	To examine the psychometric properties (inter-rater reliability, internal consistency, concurrent validity and content validity).	707 participants with neurological conditions (60 excluded for various reasons): Angelman (N=104); Cornelia de Lange (N=101); Cri-du-Chat (N=58); Fragile X (N=191); Prader-Willi (N=198); Lowe (N=56); Smith-Magenis (N=42); and intellectual disability of heterogeneous aetiology (N=56). Participants between the ages of 10 and 28 years (mean age 17.6 years; SD = 3.7). 70.9% participants were male. 45.6% participants were verbal. Participants	Medium.
Barrett et al. (2018)	Repetitive Behaviour Questionnaire-2A	Cross-sectional within-subjects design.	To examine psychometric properties, including internal consistency and convergent validity (through subscale analysis).	275 adult autistic participants, recruited by opportunity sample through online advertising (60.8% female, 1.6% non-binary; 92.6% Caucasian; mean age 37.0 years, SD 12.32). 77.3% educated to at least post-16 and 41.4% to degree level. Mean AQ score 38.31, SD = 5.86, range 26-50.	Medium-High.
Jia et al. (2019)		Cross-sectional within-subjects design.	To test internal consistency and convergent (between scales) and discriminant validity (against Autism Quotient, personality measures and Systemizing Quotient).	Study 1: 207 undergraduate participants (mean age 21.86 years, 91.8% between 20-25 years; 35.3% female). Study 3: 442 more general population (56.3% females; 41% between 18 and 34 years, 41% between 45 and 54 years, 18% over 55 years).	Medium.

Table 6.10. *Psychometric data for OCD assessment studies.*

Measure	Study	Sample mean OCD tool score	Internal consistency	Inter-rater reliability	Test-retest reliability	Discriminant validity	Convergent	Concurrent validity
Florida Obsessive-Compulsive Inventory	Storch et al. (2007)	27.38 (SD = 5.43) (YBOCS)	FOCI Symptom Checklist (α = 0.83). Severity (α = 0.89)	NR	NR	Clinical Global Impression Scale (*r* = 0.29), Beck Depression Inventory (*r* = 0.35); Hamilton Depression Rating Scale (*r* = 0.34)	NR	Y-BOCS (*r* = 0.89)
Leyton Obsessional Inventory	Richter et al. (1994)	Symptom = 23.79 (SD = 7.88)	NR	NR	NR	NR	NR	LOI Symptoms versus Y-BOCS Total (*r* = 0.63)
	Stanley et al. (1993)	Symptom = 22.6 (SD = 7.1)	Symptom (α = 0.88). Trait (α = 0.75). Resistance (α = 0.88). Interference (α = 0.90).	NR	NR	Moderate correlation with psychoticism (*r* = 0.45; obsessive-compulsive versus LOI symptom) and with neuroticism (*r* = 0.37 with LOI symptom). Interference scores strongest predictor of OCD versus non-OCD assignment (R^2 = 0.51); Symptom scores also significant difference between two groups.	NR	NR

Nb. All correlations are significant, unless otherwise stated.

Measure	Study	Sample mean OCD tool score	Internal consistency	Inter-rater reliability	Test-retest reliability	Discriminant validity	Convergent	Concurrent validity
Obsessive Compulsive Inventory-Revised	Abramowitz and Deacon (2006)	Total OCI-R = 19.78 (SD = 13.82)	With OCD patients (α = 0.83)	NR	NR	Moderation correlation with measures of depression (*r* = 0.41), anxiety (*r* = 0.47) and Intolerance for Uncertainty (*r* = 0.39)	NR	Weak to moderate relationships with Y-BOCS total (*r* = 0.41).
	Cadman et al. (2015)	Total OCI-R 29.66, SD = 15.25 (OCD group), 26.96, SD = 14.01 (autism group), 34.06, SD = 15.11 (comorbid) and 9.18, SD = 6.86 (control).	For autism sample, excellent internal consistency, with total OCI-R (α = 0.92)	NR	NR	NR	Correlation between subscales moderate (*r* = 0.32-0.59), indicating they measure different aspects of OCD.	NR
	Hajcak et al. (2004)	Total OCI-R 18.91 (SD = 11.38)	Total OCI-R (α = 0.88)	NR	Excellent: OCI-R total (*r* = 0.70)	Moderate correlations with worry (*r* = 0.42) and depression (*r* = 0.39).	Moderate correlation with the MOCI (*r* = 0.56)	NR
	Moritz et al. (2012)	Total OCI-R 24.21 (SD = 11.27)	NR	NR	NR	NR	Moderate correlation with the Y-BOCS self-report (*r* = 0.59)	NR

Nb. All correlations are significant, unless otherwise stated.

Measure	Study	Sample mean OCD tool score	Internal consistency	Inter-rater reliability	Test-retest reliability	Discriminant validity	Convergent validity	Concurrent validity
Padua Inventory (PI)	Burns et al. (1996)	Total for OCD group 54.93 (SD = 16.72)	High levels of internal consistency between subscales α (0.77-0.88)	NR	Total (*r* = 0.76), scales between (*r* = 0.61-0.79).	Moderate correlations with worry: Total PI score and Penn State Worry Questionnaire score (*r* = 0.34).	NR	NR
	MacDonald and de Silva (1999)	Total 24.04 (SD 23.55)	PI total (α = 0.96)	NR	NR	Moderate correlations: anxious & depressive personality symptoms (*r* = 0.58); neuroticism (*r* = 0.48).	NR	NR
Vancouver Obsessional -Compulsive Inventory	Thordarason et al. (2004)	Not reported.	VOCI total (α = 0.94)	NR	Excellent – all above (*r* = 0.9)	Moderate correlations with measures of depression (*r* = 0.43), anxiety (*r* = 0.44) and worry (*r* = 0.59), neuroticism (*r* = 0.56) and extraversion (*r* = 0.32).	NR	Total with Y-BOCS-SR total (*r* = 0.64), Padua Inventory (*r* = 0.85) and MOCI (*r* = 0.74).
Yale-Brown Obsessive-Compulsive Scale (Y-BOCS)	Arrindell et al. (2000)	NR	Obsessions (α = 0.97) Compulsions (α = 0.95)	*W*-indices: 0.897 (Obsessions); and 1.00 (Compulsions).	NR	Anxiety (*r* = 0.1) Depression (*r* = 0.3)	NR	NR
	Goodman et al. (1989a)	Severity Y-BOCS = 25.1 (SD = 6)	NR	NR	NR	Anxiety *r* = 0.47; Depression *r* = 0.60	NR	MOCI *r* = 0.53; OC *r* = 0.67
	Moritz et al. (2012)	Total 19.50 (SD = 5.79)	Clinical group (α = 0.81) Deception group (α = 0.85)	NR	Clinical group (*r* = 0.68) Deception group (*r* = 0.27)	Obsessions versus Compulsions (*r* = 0.15)	NR	NR

Nb. All correlations are significant, unless otherwise stated.

Measure	Study	Sample mean OCD tool score	Internal consistency	Inter-rater reliability	Test-retest reliability	Discriminant validity	Convergent validity	Concurrent validity
Yale-Brown Obsessive-Compulsive Scale (Y-BOCS)	Richter et al. (1994)	Total 20.03 (SD = 6.7)	Y-BOCS total (α = 0.86)	NR	NR	Depression (*r* = 0.51)	NR	Y-BOCS total versus MOCI Total (*r* = 0.54)
	Storch et al. (2007)	Total 27.38 (SD = 5.43)	NR	NR	NR	Depression *r* = 0.61 and *r* = 32	NR	FOCI (*r* = 0.89)
Yale-Brown Obsessive Compulsive Scale-Symptom Checklist (Y-BOCS-SC)	Mataix-Cols, et al. (2004)	Total 25.5 (SD = 7.5)	NR	NR	NR	Non-significant correlations with measures of anxiety and depression.	NR	MOCI (*r* = 0.29) PI (*r* = 0.34)
	Sulkowski et al. (2008)	NR	NR	NR	NR	Non-significant for Depression measure (*r* = 0.07) and moderate significance for anxiety measure (*r* = 0.29).	NR	OCI-R (*r* = 0.47)
Y-BOCS Self Report (Y-BOCS-SR)	Steketee et al. (1996)	21.4 (SD = 5.4) self-report 23.9 (SD – 5.0) interview	Total Y-BOCS (α = 0.78)	NR	*r* = 0.40 to *r* = 0.89	NR	Versus interview: total Y-BOCS (*r* = 0.73).	NR
Y-BOCS-II	Storch et al. (2010a)	Total Y-BOCS-II 30.55 (SD 7.44); Total Y-BOCS 29.03 (6.78)	Severity (α = 0.89)	ICC = 0.96	ICC = 0.85	Depression (*r* = 0.35) Worry (*r* = 0.20)	NR	OCI-R (*r* = 0.22)

Nb. All correlations are significant, unless otherwise stated.

Measure	Study	Sample mean OCD tool score	Internal consistency	Inter-rater reliability	Test-retest reliability	Discriminant validity	Convergent validity	Concurrent validity
Y-BOCS-II-SC	Storch et al. (2010b)	20.48 (SD = 11.11)	KR-20 values 0.91 (total)	ICC = 0.97	ICC = 0.90	Anxiety (*r* = 0.29) Depression (*r* = 0.38)	NR	OCI-R (between *r* = 0.35 and *r* = 0.76)
	Wu et al. (2016)	26.54 (SD = 7.66)	α = 0.86	ICC between 0.97 and 0.99	*r* = 0.64 and *r* = 0.81	Non-significant for anxiety and impulsiveness Depression (*r* = 0.41)	NR	NR
Y-BOCS-II-SR	Hiranyatheb et al. (2015)	Total 20.71 (SD = 11.16)	Total scores (α = 0.94) Obsessions (α = 0.90) Compulsions (α = 0.89)	NR	NR	Depression measure (*r* = 0.41) Patient Health Questionnaire (*r* = 0.61) Quality of life (*r* = -0.44)	NR	FOCI (*r* = 0.90)

Nb. All correlations are significant, unless otherwise stated.

Table 6.11. *Psychometric data for Repetitive Behaviour assessment studies.*

Measure	Study	Sample mean repetitive behaviour score	Internal consistency	Inter-rater reliability	Test-retest reliability	Discriminant validity	Convergent validity	Concurrent validity
Autism Behavior Inventory	Bangerter et al. (2017)	None reported	Repetitive behaviour scale: α = 0.80 (full version) and α = 0.60 (short version). Restricted interests α = 0.63 (full version), not calculated for short version.	NR	Overall (full version), mostly *r* > 0.90 for parent-reported and *r* > 0.80 for clinician-reported.	No significant correlations between repetitive behaviour domain and measures of anxiety (*r* = 0.24; Child and Adolescent Symptom Inventory, Anxiety subscale). However, high correlations between repetitive behaviour domain and social communication questionnaire (*r* = 0.80) and social responsiveness scale (*r* = 0.83).	NR	Strong correlation with repetitive behaviour domain and RBS-R (*r* = 0.89).
Repetitive Behaviour Scale-Revised (RBS-R)	Inada et al. (2015)	Not reported	Overall α = 0.928, scales between α = 0.693 (restricted) and α = 0.877 (sameness)	NR	NR	Significant moderate correlation with IQ (*r* = 0.381 to 0.545), significant correlation with sensory processing (*r* = -0.627 to 0.617), significant correlations with adaptive skills (*r* = -0.754 to -0.431) and significant correlations with aberrant behaviours (*r* = 0.527-0.699).	NR	NR

Nb. All correlations are significant, unless otherwise stated.

Measure	Study	Sample mean repetitive behaviour score	Internal consistency	Inter-rater reliability	Test-retest reliability	Discriminant validity	Convergent validity	Concurrent validity
Repetitive Behaviour Scale-Revised (RBS-R)	Lam (2004)	In adults (20 years plus): 32.84 (SD = 20.84)	Satisfactorily high overall: subscales mean α = 0.83; between α = 0.78 to 0.91).	In adult sample ICC = -0.24 (stereotypic behaviour) to ICC = 0.72 (compulsive behaviour).	NR	NR	NR	NR
	Martínez-González and Piqueras (2018)	Not reported.	Total α = 0.93. Compulsive α = 0.70; ritualistic α = 0.80; self-injury α = 0.83; stereotypic α = 0.86; sameness α = 0.88.	NR	Total α = 0.83, with ICC between 0.95 (restricted behaviour) and 0.98 (self-injury).	(Selected results) Significant moderate correlations between total RBR-R and measures of: reciprocal social interaction (*r* = 0.36); communication (*r* = 0.32); attention (*r* = 0.15); social skills (*r* = 0.39); temperament (*r* = 0.32); and regulation (*r* = 0.41).	NR	Total RBS-R with repetitive behaviours from the Social Communication Questionnaire (*r* = 0.68)

Nb. All correlations are significant, unless otherwise stated

Measure	Study	Sample mean repetitive behaviour score	Internal consistency	Inter-rater reliability	Test-retest reliability	Discriminant validity	Convergent validity	Concurrent validity
Repetitive Behaviour Questionnaire	Moss et al. (2009)	M = 3.20 – 6.90 across groups for stereotyped behaviours and M = 1.29 to 7.21 for compulsive behaviours.	α > .80 (stereotyped behaviours) α > 0.70 for compulsive behaviours Lower for preferences, speech and sameness (α = 0.50, 0.54, and 0.64)	NR	Ranging from ICC = 0.61 to 0.93 at item level, with 52.6% of items above .80.	NR	NR	Good against the Autism Screening Questionnaire (*r* = .6).
Repetitive Behaviour Questionnaire-2 Adults	Barrett et al. (2018)	Total 2.11 (*M* = 0.38)	α = 0.70 (RSMB) α = 0.81 (IS)	NR	NR	NR	RSMB *r* = 0.25 IS *r* = 0.42	NR
	Jia, Steelman and Jia (2019)	Study 1: not reported Study 3: RSMB 2.91 (SD = 1.40), IS 3.22 (SD = 1.33)	Study 1: α = 0.80 (RSMB) α = 0.83 (IS)	NR	NR	RSMB: Extroversion (*r* = -0.28), neuroticism (*r* = 0.51) and systemizing quotient structure (*r* = 0.28). IS: Extroversion (*r* = -0.32), neuroticism (*r* = 0.40) , systemizing quotient structure (*r* = 0.14) and systemizing quotient technicity (*r* = 0.15).	NR	NR

Nb. All correlations are significant, unless otherwise stated.

6.3.1. OCD assessment tools

To compare the psychometric properties of OCD measurement tools, evidence from the systematic review is presented below. The data relating to the key psychometric properties (as stated in Table 6.1) are set out for clarity in Table 6.10. The following narrative synthesis first details evidence for the various versions of the Y-BOCS, as the tool regularly claimed as the gold standard. Psychometric evidence from the various Y-BOCS tools will be compared, to select the most appropriate tool based on the evidence. Following this, each of the other identified OCD tools will be compared to the selected Y-BOCS tool. It is notable the quality of the studies for the OCD tools are all medium to high, with only one study (Richter et al., 1994) demonstrating medium quality (on the basis of relatively small sample size and unclear recruitment procedures – see Table 6.8).

6.3.1.1. Yale-Brown Obsessive-Compulsive Scale.

The Yale-Brown Obsessive Compulsive Scale (Y-BOCS) has demonstrated robust psychometric properties cross-culturally, in both participants in the Netherlands (Arrindell et al., 2000) and Germany (Moritz et al., 2012). Subscale factors (e.g. Severity and Disturbance), appear to be extremely highly correlated, and two overall factors (Obsessions and Compulsions) have been reported to explain 81% of variance in OCD symptoms (Arrindell et al., 2000). Obsessions and Compulsions appear to be relatively independent, with low correlations reported between them (Arrindell et al., 2000). Internal consistency across both cross-cultural groups appear good, for both Obsessions ($\alpha = 0.97$) and Compulsions ($\alpha = 0.95$) (Arrindell, et al., 2000), as well as overall ($\alpha = 0.81$; Moritz et al., 2012). However, a notable issue for the present study –

delivered online – is the lack of accuracy in detecting deceitful answers (from non-OCD individuals), with identical internal consistency in an expert deception group (α = 0.81; Moritz et al., 2012). Discriminant validity appears reasonable, with relatively small associations of state anxiety (*r* = 0.1) and depression (*r* = 0.3; Arrindell et al., 2000). Additionally, test-retest appears reasonable (*r* = 0.68) (Moritz et al., 2012).

Additionally, the Y-BOCS has shown good concurrent validity in comparison to self-reported OCD measures. Strong correlations (*r* = 0.89) have been reported with the Florida Obsessive Compulsive Inventory (FOCI) severity scales (Storch et al., 2007). The Y-BOCS appears to be slightly better than the FOCI at discriminating against depression: correlations with the Beck Depression Inventory and the Hamilton Depression Rating Scale were *r* = 0.61 and *r* = 0.32 (Y-BOCS total) and *r* = 0.70 and *r* = 0.36, respectively (Storch et al., 2007). The Y-BOCS has been reported to demonstrate good discriminant depression against anxiety and depression, with Arrindell et al. (2000) reporting correlations between *r* = 0.1 and *r* = 0.3.

In a lower quality study – mainly due to much lower number of participants (*n* = 30, versus *n* = 113 in Storch et al., 2007) – Richter et al. (1994) reported the Y-BOCS (total score) to be moderately associated with the self-report Maudsley Obsessive Compulsive Inventory (*r* = 0.54). However, it is worth noting significant correlations were only found between the two self-report measures and Y-BOCS scores for female participants (*r* = 0.73 and *r* = 0.83), but not for males (*r* = 0.28 and *r* = 0.40; Richter et al., 1994).

6.3.1.2. Yale-Brown Obsessive Compulsive Scale: self-report.

Psychometric properties of self-report versions of the Y-BOCS (Y-BOCS-SR: Moritz et al., 2012; Steketee et al., 1996) and the Y-BOCS-II-SR (Hiranyatheb et al., 2015) have been generally good. For the Y-BOCS self-report, Steketee et al. (1996) reported good internal consistency (ranging from α = 0.55 for Obsessions, α = 0.71 for Compulsions and α = 0.78 for total Y-BOCS). This was either comparative, or even higher than the clinician-administered interview Y-BOCS (Obsessions α = 0.56; Compulsions α = 0.61; and Total Y-BOCS α = 0.74). Similarly, for the Y-BOCS-II-SR, Hiranyatheb et al. (2015) reported high internal consistencies for total scores (α = 0.94), Obsessions (α = 0.90) and Compulsions (α = 0.89). Furthermore, each item showed good factor loading with total score (between 0.54 and 0.94). Whilst Moritz et al. (2012) similarly found internal consistency was reported to be good (α = 0.81) for the OCD group, there was no difference between this score and the internal consistency in the deception group (α = 0.85), which does indicate some difficulties in identifying deceit.

For test-retest reliability, an online self-reported Y-BOCS appears to be adequate (*r* = 0.68; Moritz et al., 2012). Additionally, in a non-clinical group all self-reported Y-BOCS scores were at last moderately correlated (*r* = 0.40 to *r* = 0.89; total Obsessions *r* = 0.87; total Compulsions *r* = 0.88) (Steketee et al., 1996). Comparatively, these scores were much higher than for the interview OCD measures (ranging between *r* = 0.11 and *r* = 0.79; Steketee et al., 1996).

No significant differences have been reported for items endorsed or category scores between the self-report and the interview format, indicating good content validity

(Steketee et al., 1996). Similarly, Hiranyatheb et al. (2015) reported no significant differences between the clinician-administered and the self-report formats (p = 0.96). For convergent validity, Steketee et al. (1996) reported correlations between the self-report and interview to be strong, for: total Y-BOCS (r = 0.73); Obsessions (r = 0.78); and Compulsions (r = 0.79). Hiranyatheb et al. (2015) also reported good convergent validity, indicated by high correlation with Florida Obsessive-Compulsive Inventory (r = 0.90).

Whilst Steketee et al. (1996) indicated poor sensitivity of the Y-BOCS self-report, as significant numbers of non-OCD participants scored above the cut-off for OCD (40% for the self-report version and 50% for the interview), it is notable there were very few participants in the non-OCD clinical group (n = 10). Hiranyatheb et al. (2015) identified reasonable divergent validity, with moderate correlations with measures of depression (r = 0.41), health (r = 0.61) quality of life (r = -0.44).

6.3.1.3. Yale-Brown Obsessive Compulsive Scale-Symptom Checklist (Y-BOCS-SC).

A better understanding of the heterogeneity of OCD has been attempted by moving away from the overall severity (subscales) of OCD and focusing more specifically on the traits (Mataix-Cols et al., 2004). The focus, therefore, shifts to a self-report method of clinical assessment.

Mataix-Cols et al. (2004) identified generally poor convergent validity for the Y-BOCS-SC compared to the Maudsley Obsessional Compulsive Inventory (MOCI) (small: r = 0.29) and the Padua Inventory (PI) (moderate r = 0.34) in 56 participants, although

many of low correlations or non-significant comparisons may result from the lack of direct mapping between items on these scales (see Table 6.10). However, in a larger sample of 112 patients, Sulkowski et al. (2008) reported much more robust convergent validity between the measure and Obsessive Compulsive Inventory-Revised scores (r = 0.63), although checking symptoms appeared to generally mediate this significant correlation (see Table 6.10).

Discriminant validity of the Y-BOCS-SC appears to be good, suggesting it may be measuring OCD independently as a pure construct. The Y-BOCS-SC appears to perform better than the MOCI and PI for discrimination against some measures of anxiety and depression. Mataix-Cols et al. (2004) reported identified no significant correlations between anxiety and depression for the Y-BOCS-SC, whilst whereas significant moderate correlations were reported between the MOCI (washing subscale) and anxiety (r = 0.37) and for three of the MOCI subscales for depression (r = 0.33 to 0.37), with the PI also demonstrating significant moderate correlations across three of the four subscales with depression (r = 0.28 to 0.48). This is notable as the Y-BOCS-SC was reported to correspond poorly to the self-administered MOCI and PI, although this may reflect poorer validity in the latter tools, as reasonable psychometric properties have been reported for the Y-BOCS-Self-Report tool (Steketee et al., 1996).

Similarly, Sulkowski et al. (2008) reported small to moderate associations between the Y-BOCS-SC and the anxiety measures (r = 0.29) and even non-significant for depression measures (r = 0.07), indicating good divergent validity for anxiety and, particularly for

depression. Furthermore, these results were comparable between the "improved" Y-BOCS-II-SC (*r* = 0.41; Wu et al., 2016).

6.3.1.4. Yale-Brown Obsessive Compulsive Scale-II.

Storch et al. (2010a) adapted the Y-BOCS with various purported improvements, to form the Y-BOCS-II. Changes consisted of: changing "resistance to obsessions" to "obsession-free interval"; expanding the Likert Scale to more sensitively assess more severe reports; including avoidance items to provide measure of a previously hidden clinical factor; and rewording of certain traits.

The Y-BOCS-II has also been demonstrated to show generally robust psychometric properties Storch et al. (2010a), including cross-culturally, in a relatively small Thai OCD outpatient sample (Hiranyatheb et al., 2015). Demographic analyses have identified no significant differences between males and females, whilst older age was significantly related to increased Y-BOCS-II symptom severity (*r* = 0.22) (Storch et al., 2010a). Internal consistency has been indicated to be high (Storch et al., 2010a), across Y-BOCS-II Severity (α = 0.89), Obsessions (α = 0.86) and Compulsions (α = 0.84). Similar consistency has been reported cross-culturally for total Y-BOCS-II scores (α = 0.94), including Obsessions (α = 0.90) and Compulsions (α = 0.89) (Hiranyatheb et al., 2015).

Inter-rater (ICC = 0.96) and test-retest reliability (ICC = 0.85) have both been reported to be very high (Storch et al., 2010a). Convergent validity appears generally good, with strong correlations were identified between the Y-BOCS-II and Clinical Global Impression-Severity (*r* = 0.87), the National Institute of Mental Health Global Obsessive

Compulsive Scale (*r* = 0.85) (Storch et al., 2010a) and the Florida Obsessive-Compulsive Inventory (*r* = 0.90) (Hiranyatheb et al., 2015).

Discriminative validity appears to be reasonable, with Storch et al. (2010a) reporting only moderate correlations with Y-BOCS-II Severity to depressive symptoms (*r* = 0.35) and general worry (*r* = 0.20). Similarly, Hiranyatheb et al., (2015) reported moderate correlations with self-reported measures of depression (*r* = 0.41), patient health (*r* = 0.61) and quality of life (*r* = -0.44). Notably, no significant differences have been reported between the clinician-administered Y-BOCS-II-SR and self-report measures (*p* = 0.96) (Hiranyatheb et al., 2015).

6.3.1.5. Yale-Brown Obsessive Compulsive Scale-II-Symptom Checklist.

A symptom checklist version has also been created for the Y-BOCS-II. Storch et al. (2010b) analysed psychometric properties to identify whether the measure remains valid when the subscales are dropped. The researchers reported a four factor solution (symmetry/order; contamination/washing; hoarding; and sexual/religion/aggression), accounting for 60.78% of the variance. Internal consistency appears generally high, and adequate for the four individual factors (Storch et al., 2010a), with acceptable to good internal consistency scores of between α = 0.86 for total Severity to α = 0.75 for Compulsions Severity (Wu et al., 2016). Inter-rater consistency has been reported to be high (ICC between 0.97 and 0.99; Storch et al., 2010a; and Wu et al., 2016) and test-retest reliability adequate (ICC = 0.90; Storch et al., 2010a and acceptable (correlations between *r* = 0.64 and *r* = 0.81; Wu et al., 2016).

For convergent validity, the total Y-BOCS-II-SC score was found to correlate moderately to strongly with all the subscale measures, including the Obsessive-Compulsive Inventory-Revised (between $r = 0.35$ to $r = 0.76$), indicating the tool might be able to measure general distress from OCD. For divergent validity, significant moderate correlations have been reported with measures of anxiety ($r = 0.29$) and depression ($r = 0.38$; Storch et al., 2010a). Moreover, Wu et al., (2016) reported non-significant correlations reported between Y-BOCS-II total severity and measures of anxiety and impulsiveness. However, amongst the factors, Storch et al. (2010b) reported the symmetry/order factor did not significantly correlate with either anxiety or depression ($r = 0.17$ and $r = 0.12$, respectively), anxiety was not significantly associated contamination/washing ($r = 0.18$) and depression was not found to be significantly associated with hoarding ($r = 0.14$). Wu et al. (2016) also reported divergent validity was not as good for measures of depression, with a moderate correlation demonstrated against the Depression Anxiety Stress Scale ($r = 0.41$).

6.3.2. Comparison of psychometric properties for Y-BOCS versions.

Overall, psychometric properties for all the different Y-BOCS versions appear generally robust and all the reported studies were of at least medium to high quality (see Table 6.6). Internal consistency has been reported comparably high for the original Y-BOCS, with α(0.81) reported by Moritz et al. (2012) and α(0.86) reported by Richter et al. (1994), compared to α(0.86) reported by Wu et al. (2016) for the Y-BOCS-II-SC. Inter-rater reliability for all the Y-BOCS-II versions have been found to be very high, with intraclass correlations reported between 0.96 (Storch et al., 2010a) and 0.97 (Storch et al. ,2010b; Wu et al., 2016). Test-retest reliability is also comparable between the

original Y-BOCS (*r* = 0.68; Moritz et al., 2012), and the "improved" Y-BOCS-II (*r* = 0.64; Wu et al. 2016). The Y-BOCS-II appears to have much better properties for discriminant validity than the original Y-BOCS, although interestingly the discriminant validity against measures of depression appear to be better for the Y-BOCS-SC (Sulkowski et al., 2008) than the Y-BOCS-II-SC (Storch et al., 2010b; Wu et al., 2016). Concerns of poor concurrent validity between the Y-BOCS-SC and the self-administered MOCI and PI (Mataix-Cols et al., 2004) may be bolstered by good psychometric properties reported for the Y-BOCS-Self Report (Steketee et al., 1996). Finally, whilst there are no psychometric properties to report in cross-cultural samples using the Y-BOCS-II-SC, robust psychometric properties were reported for cross-cultural studies using the Y-BOCS-II-SR (Hiranyatheb et al., 2015), Y-BOCS (Arrindell et al., 2000), Y-BOCS self-report (Moritz et al., 2012) and the Y-BOCS Symptom Checklist (Mataix-Cols et al., 2014).

The Y-BOCS-II-SR would appear to be a reasonable tool to use in an adult sample, based on good comparative psychometric properties in particular for discriminant validity, concurrent validity and internal consistency (Hiranyatheb et al., 2015), in addition to robust properties for the all the underlying versions of the Y-BOCS (see Table 6.10).

6.3.3. Comparison of other OCD tools.

Before confirming the Y-BOCS-II-SR as a suitable tool for the present investigation, data on psychometric measures will be compared to the other OCD assessment tool studies indicated in the systematic search.

6.3.3.1. Florida Obsessive-Compulsive Inventory.

Storch et al. (2007) reported good internal consistency (α = 0.83) for the Florida Obsessive-Compulsive Inventory (FOCI) Checklist. However, this is lower (but possibly statistically comparable) than the internal consistency reported for the Y-BOCS, with alpha coefficients ranging from α(0.86; Richter et al., 1994) to between α(0.96 to 0.97) for the Obsessive and Compulsive trait items (Arrindell et al. 2000). This is comparable to the internal consistency scores reported by Hiranyatheb et al., (2015) for the Y-BOCS-II-SR, with total scored at α(0.94) (see Table 6.10). Storch et al. (2007) reported concurrent validity between the FOCI Severity scales and the Y-BOCS Total Severity to be strong (*r* = 0.89). The researchers also identified divergent validity to be reasonable, with more moderate correlations against measures of global clinical functioning (*r* = 0.29), and depression (between *r* = 0.34 and *r* = 0.35). This is comparable to concurrent validity data against depression measures reported for the Y-BOCS-II-SC by both Wu et al. (2016) and Storch et al. (2010b), which were between *r* = 0.38 and *r* = 0.41, respectively. However, the Y-BOCS-SC has been reported to demonstrate even stronger discriminant validity properties, with non-significant correlations reported by Mataix-Cols et al. (2004) and Sulkowski et al. (2008).

6.3.3.2. Leyton Obsessional Inventory.

Psychometric properties of the Leyton Obsessional Inventory (LOI) have been reported to be generally adequate (Stanley et al., 1993). Comparable internal consistency has been reported between the LOI, reported between α(0.75 to 0.90) by Stanley et al. (1993) and various versions of the Y-BOCS, including: α(0.78) for the Y-BOCS self-report

(Steketee et al., 1996); and α(0.86) for total Y-BOCS (Richter et al., 1994), which is lower than overall internal consistency of α(0.94) reported by Hiranyatheb et al., (2015) for the Y-BOCS-II-SR. Little other evidence of psychometric properties of the LOI was identified in the present systematic search. No discriminant validity can be reported for measures of anxiety and depression. However, whilst the LOI has been reported to demonstrate good divergent validity compared to measures of psychoticism and two of the three personality measures (extraversion and lie), moderate significant correlations have been reported between LOI Symptom scores and obsessive-compulsive psychoticism measures (r = 0.45) and neuroticism (r = 0.37). These are comparative, if not higher, than the anxiety and depression correlations across the Y-BOCS measures (e.g. Arrindell et al., 2000; Storch et al., 2010a; Sulkowski et al., 2000; Wu et al., 2016), although they clearly lack direct comparison.

6.3.3.3. Vancouver Obsessional Compulsive Inventory.

Internal consistency has been reported higher for the Vancouver Obsessional Compulsive Inventory (VOCI; Thordarson et al., 2004) at α(0.94), than many of the scores across the Y-BOCS versions. Test-retest reliability has also been reported higher in the VOCI, with correlations above r = 0.9 reported by Thordarson et al. (2004), compared to correlations at r = 0.68 for the Y-BOCS (Moritz et al., 2012), between r = 0.40 to 0.86 for the Y-BOCS-Self-Report (Steketee et al., 1996) and between r = 0.64 to 0.81 for the Y-BOCS-II-SC (Wu et al., 2016). Test-retest validity for the Y-BOCS-II-SR was also high, with ICC = 0.85 reported by Hiranyatheb et al., (2015). The Y-BOCS-II-SR appears have similar discriminant validity to the VOCI. Hiranyatheb et al. (2015) reported measures of depression (r = 0.41) and quality of life (r = 0.44) similar to the

comparative correlations between the VOCI and measures of depression (*r* = 0.43), anxiety (*r* = 0.44) and, in particular, worry (*r* = 0.59), as reported by Thordarson et al. (2004). Comparatively, discriminant validity against measures of depression for the Y-BOCS measures have been reported to vary across this range: between α(0.3; Arrindell et al., 2000) to α(0.7; Sulkowski et al., 2008) for Y-BOCS versions; and between α(0.35; Storch et al., 2010a) to α(0.41; Hiranyatheb et al., 2014; Wu et al., 2016) for Y-BOCS-II versions.

6.3.3.4. Padua Inventory.

The internal consistency scores for the Padua Inventory (PI) have also been reported to be higher (α = 0.96; MacDonald and de Silva, 1999) than evidence across the Y-BOCS measures. Evidence of test-retest scores are comparative, with moderate correlations between *r* = 0.61-0.79 for the PI (MacDonald and de Silva, 1999), the Y-BOCS (*r* = 0.68; Moritz et al., 20120) and reported at between *r* = 0.64-0.81 for the Y-BOCS-II-SC (Wu et al., 2016).

However, discriminant validity appears not to be as robust for the PI. Whilst correlations between worry have been reported by Storch et al. (2010a) at *r* = 0.20 for the Y-BOCS-II, higher correlations have been reported for the PI (*r* = 0.34; Burns et al., 1996). Additionally, much higher correlations have been reported by MacDonald and de Silva (1999) for correlations between the PI with anxious and depressive personality symptoms (*r* = 0.58) and trait neuroticism (*r* = 0.48), which is less discrimant than the measures of depression (r = 0.41) and quality of life (r = 0.44) for the Y-BOCS-II-SR as reported by Hiranyatheb et al. (2015).

The overall evidence would indicate the PI to be a less robust measure of OCD than the Y-BOCS measures. This is further strengthened by criticisms which have been aimed at the PI. For example, Thordarson et al. (2004) argued some symptoms clinically believed to be important to OCD (such as hoarding) are not covered, which is not an issue in the Y-BOCS.

6.3.3.5. Obsessive-Compulsive Inventory-Revised.

Internal consistency for the Obsessive-Compulsive Inventory-Revised (OCI-R), reported between α(0.83) by Abramowitz and Deacon (2006) and α(0.88) by Hajcak, Huppert, Simons and Foa (2004), appears comparable to internal consistency scores across the Y-BOCS versions (see Table 6.10). However, test-retest reliability has been reported to be inferior in the OCI-R (r = 0.70; Hajcak et al., 2004) compared to the Y-BOCS-II-SC (r = 0.81; Wu et al., 2016), although comparable to over versions of the Y-BOCS (Moritz et al., 2012; Steketee et al., 1996).. Abramowitz and Deacon (2006) reported a moderate significant correlation (r = 0.41) between the OCI-R and measures of depression, whilst for the Y-BOCS-SC, Sulkowski et al. (2008) found correlation with depression to be non-significant (r = 0.07), indicating better discriminant validity in the latter.

Additionally, Thordarson et al. (2004) criticised the OCI-R on the basis of procedural problems, arguing the “double rating” of each item (in terms of frequency and distress) to be confusing to OCD participants, and problematic in the extra length of time required to complete the assessments. This is not a concern in the Y-BOCS. On the

basis of this, in addition to the comparative psychometric data, it would not seem more valid or reliable to use the OCI-R over the Y-BOCS.

6.3.4. Summary OCD assessment psychometric properties.

The overall evidence indicates the Y-BOCS, and in particular both the Y-BOCS-II-SC and the Y-BOCS-II-SR, has robust psychometric properties and is a valid measure of OCD in adult participants. As detailed above, psychometric properties of this measure appear to exceed those of the other OCD tools identified in this systematic search and would be the most suitable tool to assess OCD in an adult population.

Finally, in the context of the present thesis, the distinct lack of evidence for samples of autistic adults is also notable. Only one study recruited from this population, in which Cadman et al. (2015) reported excellent internal consistency in autistic adult participant for the OCI-R ($\alpha = 0.92$). As there is a large and wide ranging evidence-base of comorbidity between OCD and autism, it would appear crucial to investigate psychometric properties of OCD measurement tools in adults with autism. However, as assessments of Repetitive Behaviours have been studied in the autistic population, this evidence may provide insight into what may be a relevant framework between the disorders: Compulsive and Repetitive Traits.

6.4. Repetitive Behaviour assessment tools

Of the sevem studies of psychometric properties of Repetitive Behaviour tools, only one of these studies recruited an adult-only sample (Barrett et al., 2018). Across a high number of autistic adult participants, Barrett et al. (2018) found the RBQ-2A to be a

reliable and valid self-report measure of Repetitive Behaviours in adults with autism, in an online delivered study. Two components were identified, explaining 49.1% of the variance: repetitive sensory and motor behaviours (RSMB); and insistence on sameness (IS). Whilst there were no significant differences between gender, RSMB (but not IS) was negatively correlated with age, especially in over 50 year olds (mean = 2.05 for 18-34 years, to mean = 1.76 for 50-66 years). Barrett et al., (2018) reported internal consistency was acceptable for the RSMB (α = 0.70) and good for the IS (α = 0.81) component and both components were found to be significantly moderately correlated: RSMB (*r* = 0.25); and IS (*r* = 0.42).

Moss et al. (2009), was included in this systematic review as it was the study which developed the Repetitive Behaviour Questionnaire, which was used in the empirical investigation in Chapter 7 (for reasons explained in 7.1.3). Overall, psychometric properties were mixed (see Table 6.11). Internal consistency was as comparably high to Barrett et al. (2018), for stereotyped and compulsive behaviours, though much lower for other repetitive traits, including Insistence on Sameness. Test-retest reliability was good, but only in just over half of the neurological groups (ICC = 52.6% of items above .80). Moss et al. (2009) was the only study in this systematic review to not recruit an autism group (hence why it was not yielded in the search). However, the Autism Spectrum Quotient (ASQ) was used to screen for presence of autistic traits and identified high traits across groups. Concurrent validity of the Repetitive Behaviour Questionnaire was reported as good against the ASQ (*r* = .6). Overall, whilst concurrent validity and internal consistency (on the full-scale level) was good, in general the robustness of such results are difficult to compare with the other studies reported in

this chapter due to comorbidity issues between autism, other neurological disorders and high levels of intellectual disabilitiy/nonverbal communication.

In a non-clinical sample, Jia Steelman and Jia (2019) reported significant moderate correlations between the RBQ-2A and measures of personality (both extroversion and neuroticism), and systemizing in undergraduate participants (see Table 6.9). Whilst this indicates a lack of discriminant validity in the measure, it is notable the sample in non-clinical. Further investigation is necessary to identify whether the RBQ-2A is a psychometrically robust measure in adults with autism, specifically evidence of the following in this target sample, particularly relating to: test-retest reliability; discriminant validity (against anxiety, depression); convergent validity (against other autistic symptoms); and concurrent validity (against other Repetitive Behaviour measures).

Evidence from the other studies may provide some basis for the autistic population generally. The remaining four studies provided evidence of the psychometric properties of repetitive behaviour tools in samples of children. Overall, internal consistency was indicated to be highest for the Repetitive Behaviour Scale-Revised (RBS-R), with alpha Coefficients being reported between 0.83 (Lam & Aman, 2007) and 0.93 (Inada et al., 2015; Martínez-González and Piqueras, 2008). This is both higher than the internal consistency scores (α = 0.6 to 0.80) produced for the Autism Behavior Inventory (ABI: Bangerter et al., 2017) and for the RBQ-2A (Barrett et al., 2018; Jia et al., 2019). Inter-rater consistency has been reported to be variable for the RBS-R, with low intraclass correlations (ICC = -0.24) for stereotypic behaviour, and much higher

(ICC = 0.72) for compulsive behaviours (Lam & Aman, 2007); although overall inter-rater consistency for the RBS-R has been demonstrated high (ICC= 0.95 to 0.98) by Martínez-González and Piqueras (2008).

For discriminant validity, significant moderate correlations have been reported across the three Repetitive Behaviour measures. For the ABI, Bangerter et al. (2017) identified significant moderate correlations on the lower end for anxiety (r = 0.24), whereas high correlations were identified for the social communication (r = 0.80) and social responsiveness (r = 0.83). The RBS-R has been reported to show better discriminant validity against measures of social skills (r = 0.39) and social interaction (r = 0.36), as reported by Martínez-González and Piqueras (2008). However, Inada et al. (2015) found higher significant correlations with IQ (r = 0.381 to 0.545), sensory processing (r = -0.627 to 0.617), adaptive skills (r = -0.754 to -0.431) and significant correlations with aberrant behaviours (r = 0.527-0.0.699).

Overall, the Repetitive Behaviour measures appear generally robust, although they all appear to lack high discriminant validity against a range of other measures. The RBQ-2A (Barrett et al., 2018), is the only scale validated in autistic adults, although there is still a lack of evidence across a range of psychometric properties in this population.

6.5. Discussion

The results appear to indicate generally sound psychometric properties of the Y-BOCS, across various versions, in an adult population. The Y-BOCS demonstrates robust psychometric properties cross-culturally (Arrindell et al., 2000; Moritz et al., 2012).

Demonstrating the validity in utilising the Y-BOCS without subscales, robust psychometric properties have also been reported when using the individual trait items alone, via the Y-BOCS-Symptom Checklist (Mataix-Cols et al., 2004; Sulkowski et al., 2008). An improved Y-BOCS, the Y-BOCS-II, also appears to be robust (Hiranyatheb et al., 2015; Storch et al., 2010a), including the symptom checklist format (Storch et al., 2010a; Wu et al., 2016), again demonstrating the validity in the measure when the subscales are dropped. Finally, self-report versions of the Y-BOCS appear to be psychometrically sound, from the Y-BOCS (Moritz et al., 2012; Steketee et al., 1996), to the Y-BOCS-II-SR (Hiranyatheb et al., 2015), and both the Y-BOCS-SC (Moritz et al., 2012) and the RBQ-2A would appear to be suited to be tested in online investigations (Barrett et al., 2018).

In the scope of the present investigation, it is notable there was no evidence identified of validation of Y-BOCS measures in autistic adult samples. It would be more likely for Repetitive Behaviour assessments to be validated in this population, having being largely created for this group. However, to date only the RBQ-2A appears to have been validated in autistic adults (Barrett et al., 2018). Whilst this evidence available indicates psychometric properties to be adequate, there is still a lack of psychometric properties from this population.

Having presented evidence of the validity in comparing adults with OCD and with autism within a symptomological (repetitive trait) framework in Chapters 3 to 5, and having presented evidence of robust psychometric properties for measure of OCD

traits and Repetitive Behaviours, the next chapter will present an empirical study to test this framework.

Chapter 7. Empirical Investigation: Comparing Autism and Obsessive-Compulsive Disorder within a Compulsive and Repetitive Trait Framework

Overview

The previous chapters provide evidence indicating both the potential validity and the utility of comparing OCD and autism within a shared framework. Research indicates OCD and autism have overlapping features (Anholt et al., 2010; Bejerot et al., 2001; Hutton et al., 2008; Hollander et al., 2003; Ivarsson & Melin, 2008; Russell et al., 2005). Based on the importance of repetitive traits within both disorders, a Compulsive and Repetitive Trait (CaRT) framework is proposed.

Repetitive traits are central diagnostic features of both autism and OCD (DSM-5, APA, 2013). Furthermore, as there is evidence of links between the two disorders (e.g. Anholt et al., 2010; Bejerot et al., 2001; Hutton, Goode, Murphy, Le Couteur, & Rutter, 2008; Hollander et al., 2003; Ivarsson & Melin, 2008; Russell et al., 2005), it is relevant to determine the nature of the interaction of traits between autism and OCD. The aim of this study was to explore these relationships across autism, OCD and neurotypical development.

The evidence presented so far indicates the validity in a combined symptomological framework to compare autism and OCD. A Compulsive and Repetitive (CaRT) framework, illustrated in Figure 7.1, is proposed based on the evidence presented in the preceding chapters, indicating: validity in comparing repetitive traits between autism and OCD (Chapter 2); evidence of symptomological comorbidity between autism and OCD (Chapter 3); and potentially both ego-dystonic and ego-syntonic

repetitive traits in adults with autism (Chapters 4 and 5). Figure 7.1 depicts how this CaRT framework may be considered within (and between) both autism and OCD, based on the evidence presented in Chapters 2 to 5. This evidence presents a potential dichotomy between OCD traits and Repetitive Behaviours, with OCD traits defined as negative in origin, being unwanted symptoms. Repetitive Behaviours, however, have been historically considered a (non-negative) part of the autistic condition. Using this evidence, there would appear to be little overlap in these traits between autism and OCD (see Figure 7.1).

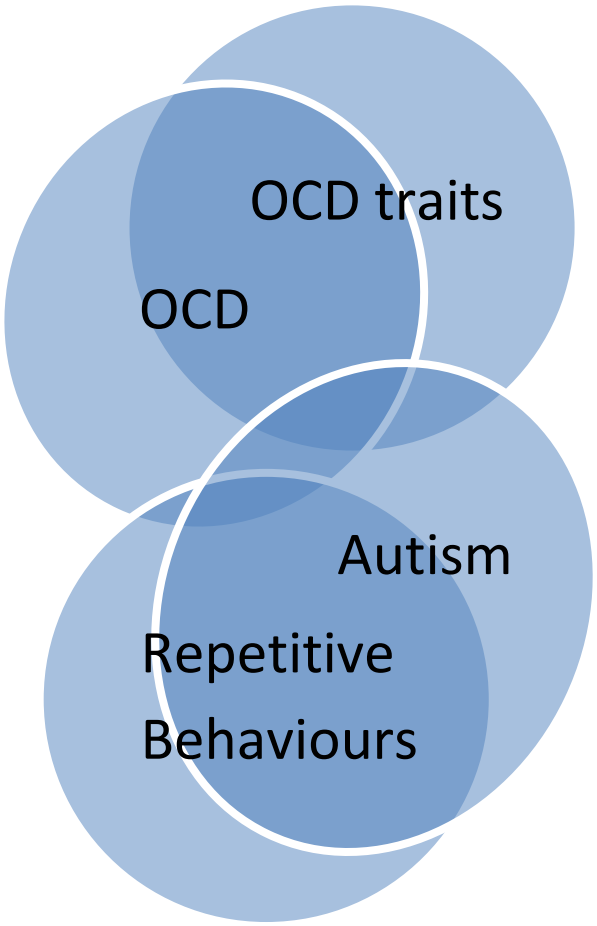

Figure 7.1. The conceptual framework demonstrating the potential relationship -- between Compulsive and Repetitive Traits in autism and obsessive-compulsive disorder.

To understand the similarities and differences between OCD and autism, the assessment of repetitive traits for both disorders will be explored. This chapter tests this framework, with a Compulsive and Repetitive trait questionnaire as a basis for this investigation. The creation of this questionnaire is described below.

7.1. Creation of the Compulsive and Repetitive Trait Questionnaire

A Compulsive and Repetitive Trait (CaRT) questionnaire was created to combine existing measures of OCD traits with existing measures of Repetitive Behaviours, alongside novel additional measures of these items. The CaRT questionnaire is described below, following an outline of OCD trait and Repetitive Behaviour measurement and the rationale behind decisions for the specific tools chosen.

The following sections outline the three constituent parts of the CaRT questionnaire. First, the measurement of obsessive-compulsive traits, including the rationale behind using the Yale-Brown Obsessive Compulsive Scale-II-Self Report (Y-BOCS-II-SR; Baer, 1993), in a self-report format. Second is the measurement of Repetitive Behaviours, including the rationale behind using the Repetitive Behaviours Questionnaire (Moss et al., 2009). Finally, the novel item-level measurement of ego-dystonic and ego-syntonic related properties is outlined.

7.1.1. Measurement of obsessive-compulsive traits and Repetitive Behaviours.

The Compulsive and Repetitive Trait questionnaire, described below, was used to distinguish between potential ego-dystonic and ego-syntonic properties of a comprehensive set of repetitive traits, using two existing measures: obsessive-

compulsive traits from OCD research; and Repetitive Behaviours from autism/intellectual disabilities research.

Obsessive-compulsive traits are distinguished by their ego-dystonic nature: being inconsistent with the individual's sense of self they are unwanted, unpleasant and associated with distress. The study of Repetitive Behaviours – originating from research on autism and intellectual disability (e.g. Lewis & Bodfish, 1998; and Moss et al., 2009) – has less certain ego-related origins. The traditional view of such behaviours being consistent with the individual's sense of self (e.g. Baron-Cohen, 1989) is definitively ego-syntonic. However, evidence has indicated there may be ego-dystonic repetitive traits in autism (e.g. Saddington, 2013).

Whereas ego-dystonic traits should be associated directly with measures of low mood, ego-syntonic traits should be associated with positive, or at least neutral mood. Comparing obsessive-compulsive traits and Repetitive Behaviours may clarify the relationship between OCD and autism. Assessing mood for every repetitive trait across the two measures would allow for a more sensitive measure of ego-dystonic and ego-syntonic properties, rather than assuming ego-syntonic or ego-dystonic properties of an entire set of repetitive traits.

7.1.2. Measurement of Obsessive-Compulsive Disorder in autism.

There is little research on the efficacy of using OCD measures in adults with autism (see section 6.2). What evidence there is almost exclusively focuses on pharmacological treatments, based on small samples, and lacks comparison groups (Brugha, Doos, Tempier, Einfield, & Howlin, 2015). This is surprising given the range of

evidence comparing OCD and autism (e.g. Anholt et al., 2010; Bejerot et al., 2001; Deramus, 2009; Hollander et al., 2003; Hutton et al., 2008; Ivarsson & Melin, 2008; Lehnhardt, 2013; Russell et al., 2005).

Whilst the Y-BOCS has been specifically adapted to assess OCD traits in an autistic sample (Scahill et al., 2006), there are some fundamental issues with this measure. Low discriminant validity suggested this measure is poor at differentiating between repetitive and maladaptive behaviours (Scahill et al., 2006). The validity of the measure is further questionable through the methods in which it was derived from the Y-BOCS: the obsession checklist was dropped; and some Repetitive Behaviours known to be prevalent in autism (such as repetitive water play), seemingly unsystematically added to the compulsions checklist.

There is some evidence OCD symptoms may be unrelated to core Repetitive Behaviour symptoms in adults with autism (Cadman et al., 2015), therefore it would appear relevant to identify an assessment tool with the adequate discriminant validity to differentiate between OCD traits and Repetitive Behaviours. In a study of children with autism, Anagnostou et al. (2011) has demonstrated that some of the Y-BOCS categories may be related to ego-syntonic Repetitive Behaviours (pleasure-related) in autism, whereas others may be related to ego-dystonic Repetitive Behaviours (anxiety-related). Potentially, a measure which more accurately distinguishes between ego-dystonic and ego-syntonic (for pleasure) Repetitive Behaviours would create a more valid measure of OCD in the autistic population. Clearly there is a need for more research to be conducted to assess OCD symptoms in adults with autism and how they are associated with Repetitive Behaviours.

7.1.2.1. Item level investigations of the Yale-Brown Obsessive Compulsive Scale.

Recognising the Y-BOCS was not originally created as scale – being rationally, rather than empirically, driven (Goodman, 1989a) – recent studies have been investigating OCD traits at the item-level. Evidence so far has demonstrated mixed success. Wu et al.'s (2007) slightly adapted version of the Y-BOCS was reported to adequately fit a sample of participants with OCD, a psychiatric sample and non-clinical controls. However, whilst Pertusa, Fernández de la Cruz, Alonso, Menchón and Mataix-Cols, (2012) concluded their Dimensional Y-BOCS shows generally robust psychometric properties, the measure still demonstrates low discriminant validity for anxiety and depression (Grabill et al., 2008). Continuing to investigate item-level properties of OCD traits may provide clearer evidence of the subtle differences between the Repetitive Behaviours in autism and OCD. Furthermore, a direct comparison between measures of OCD traits and measures of other Repetitive Behaviours may be significant in understanding the distinction between ego-dystonic and ego-syntonic repetitive traits.

7.1.2.2. Obsessive Compulsive Disorder measurement in adults with autism.

Despite its wide application, there are potential improvements which could be made to the Y-BOCS. Firstly, it was originally rationally (rather than empirically) derived (Goodman et al., 1989a). It is possible, therefore, that there are unidentified traits which are relevant to the OCD spectrum. Secondly, the Y-BOCS is created to assess severity on a global, rather than item level scale. Clinicians are expected to measure the impact of severity for just a small number of OCD traits. More recent investigations have suggested that a greater understanding can be gained from analysis of severity at the item-level (e.g. Pertusa et al., 2012; Wu et al., 2007), although such assessment has

not been undertaken in participants with autism. Finally, a review by Grabill et al. (2008) highlighted the poor discriminant validity of the Y-BOCS, particularly for depression and anxiety. It has been argued this is a consequence of comorbidity issues (Grabill et al., 2008), with shared symptomology between OCD and autism potentially being specifically implicated (e.g. Barber, 2015; Deramus, 2009; Hutton et al., 2008; Russell et al., 2005). Measures of OCD in autism have typically excluded significant items – such as obsessions (e.g. Scahill et al., 2006) – and have received little study in autistic adults.

7.1.2.3. Obsessive-compulsive disorder measurement in the present investigation.

The Yale-Brown Obsessive Compulsive Scale (Y-BOCS; Goodman et al., 1989b) is widely considered to be the "gold standard" assessment tool for identifying OCD symptoms (Deacon & Abramowitz, 2005; Wu et al., 2014). The Y-BOCS-Self Report (Y-BOCS-SR; Baer, 1992) was chosen because, despite dropping the subscales (a benefit to the present investigation due to the length of the survey), it has been shown to demonstrate robust psychometric properties in an adult population, particularly in relation to discriminant validity (Mataix-Cols et al., 2004; Sulkowski et al., 2008). The systematic literature review of psychometric properties of the Y-BOCS-SR were found to be good in comparison to other OCD measures and other Y-BOCS versions (see section 6.3.4), with robust psychometric properties as a self-report measure (Steketee et al., 1996), which was the intended method for the present investigation. The validity of self-report formats of the Y-BOCS appear to be at least comparable to clinician-administered versions (Federici et al., 2010); it has even been suggested that self-report measures may be more reliable that clinician-report versions (Steketee et al., 1996).

7.1.3. Repetitive Behaviour Measurement

A range of measurement tools have also been designed to assess what is referred to simply as Repetitive Behaviours. These Repetitive Behaviours are distinguished from the types of Repetitive Behaviours in OCD in that they are not implicitly ego-dystonic i.e. unwanted (see review by Turner, 1999). That some measures of Repetitive Behaviour have been designed to assess these traits during normal development, in early childhood (Evans et al., 1997; Leekam et al., 2007), or as core part of the symptomology of autism or learning disabilities (Lewis & Bodfish, 1998; and Moss et al., 2009), indicates they may be ego-syntonic (consistent with the individual's sense of self).

The systematic review of psychometric properties in of Repetitive Behaviour assessment tools in adults (see section 6.4) indicates only one Repetitive Behaviour measurement has been validated in a sample of adults with autism (Barrett et al., 2018). However, at the time of running the present investigation even that study had not been published. The measurement tool chosen for the present investigation (in light of the lack of research) was the Repetitive Behaviour Questionnaire (RBQ: Moss et al., 2009). The RBQ is a different assessment tool to the comparably named tool by Leekam et al. (2007). It is an 18-item informant-reported questionnaire, based on operationally defined features of behaviours.

This measure was chosen for a variety of reasons, all demonstrated in a study by Moss et al. (2009). Firstly, the measure appears to show robust psychometric properties, with Moss et al. (2009) reporting: with Spearman's coefficient of test retest reliability of items between 0.61 to 0.93; and good full-scale internal consistency ($\alpha = 0.80$).

Secondly, it has been validated for use with adult participants, the target sample for the current investigation. Thirdly, the questionnaire was based on operational definitions of behaviour, therefore accessible across a wider range of individuals. Fourthly, the items were derived from a comprehensive range of other Repetitive Behaviour tools: Stereotyped Behaviour Scale; Compulsive Behaviour Checklist; Childhood Routines Inventory; the Repetitive Behaviour Scale-Revised; and the Children's Yale Brown Obsessive Compulsive Scale. Finally, despite not recruiting any participants with autism, Moss et al. (2009) reported good concurrent and content validity between the measure and the Repetitive Behaviour subscale of the Autism Screening Questionnaire (r = 0.6), suggesting it may be a valid measure in an autistic population. To be administered as a self-report tool, the items from the original tool (see Appendix 12) were changed to first-person questions (see Appendix 3). It must be acknowledged this is another non-validated aspect of the measure and will be discussed within the limitations section (see section 7.6.1).

7.2. Compulsive and Repetitive Trait Questionnaire

The Compulsive and Repetitive Trait (CaRT) questionnaire consists of all the items from both the Y-BOCS-SR (Baer, 1992) and the Repetitive Behaviour Questionnaire (RBQ, Moss et al., 2009), two self-report questionnaires on OCD traits and Repetitive Behaviours, respectively (see Appendices 12 and 13). Appendices 2 and 3 outlines the exact items used in the CaRT questionnaire. All 58 checklist items from the Y-BOCS-SR (Baer, 1992) were included in the CaRT questionnaire, including questions on obsessions and compulsions. All 18 items from the RBQ were included (except for the control group, who answered 16 items, as described in section 7.4.8). Participants reported frequency and mood for each trait (see Appendix 1).

Items were rationally driven following the first stage of literature review for the present thesis. Whilst there were some established mood assessment measures, these were based on more in-depth analysis of mood such as the Brunel Mood Scale, which consists of 24 mood descriptors (Terry et al., 1999). For the present investigation, the most basic assessment of mood was required as the demands on the participant from the quantity of items was already large. The scale designed is a simplified Likert scale – a standard tool used widely in psychological research, from mood assessment (e.g. Terry et al., 1999) to Repetitive Behaviour (e.g. Bodfish et al., 2000; Moss et al., 2009) and OCD trait measures (e.g. Goodman et al., 1989a). As recommended by Oppenheim (1992, p.47), a pilot study was initially run on a neurotypical sample to trial the validity in the questions. In this pilot phase of the Compulsive and Repetitive Trait measure, participants were originally asked how they felt "before, during and after" each trait. This was intended to test the ego-dystonic/ego-syntonic origins of traits by understanding the preceding mood. However, feedback across the 276 neurotypical participants identified such a question was phenomenologically vague: eight participants from this pilot specifically reported being unable to use this measure. Clearly a less subjective measure was needed. The final tool made as simple assessment of mood as possible: "how did you feel when doing this?" (see Appendix 1). To assess the possibility of indirect mood (e.g. a trait may be associated with positive mood due to relief, but caused by an underlying anxiety), alternative questions were also designed for each item: for behaviours/compulsions the question was "how do you feel if you're unable to do this"; and for thoughts/obsessions the question was "how do you feel when thinking of this fear" and "how do you feel when this does happen?". Participants were prompted to report any difficulties in answering

questions – these free responses are analysed in the results section (see section 7.5.9) and used as a critique of the measure in the discussion.

The two main elements of the Y-BOCS-II-SR (Baer, 1992) consists of a checklist of potential symptoms, in addition to subscales designed to assess the severity (e.g. distress, frequency, inflexibility) of a small selection of the more prevalent obsessions and compulsions. For the present investigation, the checklist of symptoms was retained, but the severity subscales were dropped to focus on a more fine-grained analysis of specific severity assessments in line with the research question. These item-level scales consist of frequency, social context and measure of mood (see Appendix 1). Being unwanted and unpleasant, ego-dystonic traits (such as in OCD) should be related to distress; conversely, ego-syntonic behaviours are likely to be associated with pleasure, or at the very least an absence of negative (neutral) mood.

Mood is measured on two levels: firstly, when the thought/behaviour is occurring; and secondly, either when the individual is not able to do this (in the case of behaviours) or, if the obsessive content does happen (in the case of obsessive traits). The purpose of these two levels of mood assessment is to provide greater insight into whether the trait may be ego-dystonic (inconsistent with one's sense of self) or ego-syntonic (consistent with one's sense of self). Mood may not only be fixed at the time of the occurrence of the trait, but a trait may be driven by the mood which would occur in the absence of the trait. For example, an individual who repetitively washes their hands in response to a germ obsession may display neutral or positive mood when the action is occurring (e.g. in relief of alleviating the distressing obsessional content), which would erroneously give the impression in the analysis that the trait is ego-

syntonically driven (i.e. associated with positive mood). However, a more comprehensive picture can be obtained by including an assessment of mood if the trait cannot occur (i.e. how would the individual feel if they were unable to wash their hands). Clearly, this same questioning is irrelevant in the case of obsessional traits: it makes no sense to ask an individual how they would feel if they were unable to think of the obsession. In this latter case, the logical comparative question takes the form of "how do you feel if x was to happen". An example would be: "how do you feel if you do harm yourself" as a response to the item "I fear I might harm myself".

To keep consistency between both scales, for each item participants are asked to report whether they experienced each trait "never/hardly ever", "in the past but, not in the past 7 days" or "in the past 7 days". This is the scale used in the Y-BOCS-II-SR (Baer, 1992), but differs from the RQB, which measures which behaviours have occurred in the past month. For any traits endorsed by participants, they are then required to report the frequency of this trait, the situation in which this happens, and then the mood experienced during and after the trait (see Appendix 1).

7.3. Aims and Hypotheses of the Study

The overall aim of the study is to test the proposed Compulsive and Repetitive Trait framework. To establish the extent to which this framework may differentiate clinical symptoms in autism and OCD, the following hypotheses were postulated:

For the mean Compulsive and Repetitive Trait scores:

H_0 There will be no differences (number or frequency) in Repetitive Behaviours between any of the groups.

H_1 Mean scores in Repetitive Behaviours between the groups will differ: the autism group will demonstrate the highest Repetitive Behaviours (number and frequency); OCD group and control group scores will be comparable.

Rationale behind hypothesis 1: Repetitive Behaviour research has arisen from autism and learning disability research (e.g. Lewis & Bodfish, 1998; Moss et al., 2009), therefore they would be expected to be specifically prevalent in autism.

H_0 There will be no difference between the groups in mood scores for Repetitive Behaviours.

H_2 Repetitive Behaviour scores will be associated with positive mood. This will be highest for the autism group. OCD group and control group Repetitive Behaviour mood scores will be comparable.

Rationale behind hypothesis 2: These traits have been originally viewed as part of the expression of autism (Baron-Cohen, 1989), rather than a consequence of distress. If such behaviours are separate in function to ego-dystonic OCD traits, they may therefore be diametrically opposed.

H_0 There will no differences (number or frequency) in OCD traits between any of the groups.

H_3 Mean scores in OCD traits between the groups will differ. The OCD group will display the highest OCD traits (number and frequency), followed by the autism group then the control group.

Rationale behind hypothesis 3: Comorbidity between OCD and autism has been reported to be higher than in the general population (e.g. Hutton et al., 2008; Lehnhardt, 2013).

H_0 There will be no difference between the groups in mood scores for OCD traits.

H_4 OCD trait scores will be associated with negative mood. This will be highest for the OCD group, followed by the autism group then the control group.

Rationale behind hypothesis 3: OCD traits are diagnostically ego-dystonic (APA, 2013), therefore should be associated with measures of negative mood such as anxiety, distress and unpleasantness.

H_0 There will be no differences in mood between higher-order mood scores compared to lower-order mood scores.

H_5 Higher-order Repetitive Behaviours will have more positive mood scores than lower-order Repetitive Behaviours. This effect will be specific to the autism group.

Rationale behind hypothesis 5: Higher-order repetitive traits have been reported to be specific to autism (Baron-Cohen, 2008; Carcani-Rathwell et al., 2006).

H_0 Repetitive Behaviours will not differ (number or frequency) function of different contexts in participants with autism.

H_6 Participants with autism will demonstrate significantly higher Repetitive Behaviours when the context is “around people”.

Rationale behind hypothesis 6: Social anxiety and problems with social interaction are supposedly high in autism, as indicated in the DSM5--5 (APA, 2013). Comorbidity levels with social anxiety disorder appears to be high in autism, with Maddox and White (2015), for example, reporting comorbidity in 50% of an autistic sample. Depressive and anxiety symptoms have been also reported to be associated with measures of social anxiety (Liew, Thevaraja, Hong & Magiati, 2015). Longitudinal evidence suggests social and communication difficulties appears to be a risk factor to the later development of social anxiety: Pickard, Rijsdijk, Happé and Mandy (2017), report this effect strongest between the ages of 7 to 10 years. Whilst there appears to be a lack of evidence comparing social anxiety to repetitive traits in autism, this relationship in autism would be consistent with evidence repetitive traits in autism appear to occur as a consequence of anxiety generally (e.g. Rodgers et al., 2012), with social problems even indicated to be mediated through anxiety (Deramus, 2009).

For the correlation between the Y-BOCS-II-SR and the Repetitive Behaviour Questionnaire items:

H_0 There will be no correlation between OCD traits and Repetitive Behaviours.

H_7 There will be a correlation between OCD traits and Repetitive Behaviours.

In line with the research by Cadman et al. (2015) – reporting no relationship between Repetitive Behaviours and OCD symptoms in adults with autism – the null hypothesis is expected for the correlation between the Y-BOCS-II-SR and the Repetitive Behaviour Questionnaire.

Finally, data was collected to identify how difficult each participant found answering the questions, whereby participants were given a free response. These answers were expected to show all participants could answer the questions without any major difficulty, for two main reasons. Firstly, there is some evidence of both OCD trait (e.g. Cadman et al., 2015) and Repetitive Behaviour measurement (e.g. Scahill et al., 2006) in autistic samples. Additionally, following a pilot trial (albeit recruiting exclusively neurotypical participants - see section 7.2), the mood items were changed to make them easier to interpret and answer.

7.4. Method

A Compulsive and Repetitive Trait (CaRT) framework was used to compare the repetitive symptomology between OCD and autism. This study is presented below, followed by results and a discussion of implications of the key findings.

7.4.1. Design.

The study employed a cross-sectional design to compare self-reported Compulsive and Repetitive Traits (CaRTs) at a single time point. A between-groups method was used to test the dependent variable (CaRTs) using ordinal measures across a range of Likert Scales (see Appendix 1) with three levels of independent variable (disorder: autism, OCD and neurotypical control). This design is a standardised procedure (e.g. Cadman,

2015; Scahill et al., 2006) to compare clinical measures between different disorders (e.g. autism and OCD and a neurotypical control group). Specifically, this design is a widely accepted method for comparing repetitive traits in clinical populations including OCD (e.g. Goodman, 1989a; Storch et al., 2010a), autism (e.g. Scahill et al., 2006), or autistic traits (e.g. Moss et al., 2009).

7.4.2. Measure.

The Compulsive and Repetitive Trait questionnaire is a structured self-report rating which measures OCD traits (Yale-Brown Obsessive Compulsive Scale II; Storch et al., 2010a) and Repetitive Behaviours (Repetitive Behaviour Questionnaire; Moss et al., 2009). As described in section 7.2, additional novel constructs were added to these two batteries of repetitive items to measure simple potential ego-dystonic (unwanted) and ego-dystonic (pleasure-related) properties of item-level measures of mood.

7.4.3. Sample size and power calculation.

Due to the limited evidence of measurement of OCD in adults with autism, and the two papers which use the Y-BOCS-II-SR in the population failed to employ a comparative control group (Russell et al., 2005; and Ruta et al., 2010), the effect size calculation was based on the study by Cadman et al. (2015) which used a measure derived from the Y-BOCS-II-SR. This data gave an effect size of 1.34, which indicates a minimum of 10 participants need to be recruited in each group to test the null hypothesis the two groups come from populations with equal means, to have an 80% power of getting a statistically significant difference (based on an independent samples t-test using a 5% significance level). Based on the limited statistical data to base this calculation on and the novel nature of the present measure (see Chapter 5), a more cautious approach

was undertaken to improve the likelihood of the study being sufficiently powered to detect a significant effect. As detailed in Appendix 4, a sample size calculation was performed to detect a more modest medium effect size of 0.25 using a one-way ANOVA with three groups, with 80% power and using a two-sided significance level. This calculation indicates a sample size of 159, split equally between the three groups. As the power of the analyses is reduced by the lack of equal numbers across the three groups, a larger overall number of participants was sought.

7.4.4. Participants.

Three groups of participants consisted of adults with OCD (*n* = 35), autism (*n* = 39) and neurotypical controls (*n* = 170). The expected sample size expected within each clinical group did not meet the expectations (*n* = 53) from the power calculation (see 5.5.3), however, the overall number of participants was in excess to accommodate the unequal split between the groups. The limitation of this potentially underpowered sample is discussed in section 7.6.1.

Participants were included if they were aged 18 years and over and had a single diagnosis of an autism spectrum disorder, OCD, or no clinical diagnosis (control). Exclusion criteria consisted of: a diagnosis of both autism and OCD; a diagnosed psychiatric disorder; a diagnosed learning disability (IQ below 70); a diagnosis of Fragile X; structural brain abnormalities; tuberous Sclerosis complex; or Smith-Lemli-Optiz syndrome. Participants were also required to have a fluent use of the English language to be able to complete the self-report questionnaires.

7.4.5. Setting.

All elements of the study were accessed online, via Online Surveys (JISC, 2018). This was designed to increase the accessibility for autistic participants who may find social situations overwhelming: experimenter-delivered tasks have been demonstrated to affect performance (e.g. Ozonoff, 1995). Due to widespread advertisement via social media and access to email distribution (see section 7.4.7), this may have taken place at any time of day and potentially in any location to suit the participant (whether via computer or smartphone), further supporting the ecological validity of the study. Research has indicated response rates of web-based versus paper-based questionnaires appear to be comparable (Horevoorts, Vissers, Mols, Thong & van de Poll-Franse, 2015). Such response rates have been reported as high as 95.33% (Horevoorts et al., 2015) and 58% (Burgess, Nicholas & Gulliford, 2012). However, typical response rates are unclear for questionnaire methods in autistic samples (e.g. Cadman et al., 2015; Scahill et al., 2006).

7.4.6. Ethical approval.

Ethical approval was obtained by the Faculty of Health and Social Care Research Ethics Committee at the University of Hull (see Appendix 8). All participants were provided with a detailed Participant Information Sheet (see Appendix 9) giving a full outline of the study prior to providing consent. No personally identifiable information was held on the participants in the data collection. The only potentially personally identifiable information related to the email addresses individuals supplied to be considered for the prize: this information was held securely under password protection (University emails) on a locked computer until the data was no longer required. Anonymity was further supported through the online-only procedure: the services and organisations

supporting to forward the advertisements were able to act as gatekeepers to the identities of any individuals who participated in the study.

7.4.7. Recruitment.

Neurotypical adult controls were recruited via opportunity access to online pools of potential applicants across three different types of forums: personal; voluntary and community; and University. A link to the survey was posted on the lead researcher's personal Facebook and Twitter account, with a simple advert outlining the study (see Appendix 5). The local community was engaged through posting the same link and advert onto local voluntary and community electronic mailing lists in the East Riding: Volcom; Neighbourhood Networks; and East Riding Voluntary Action Service. Finally, the lead researcher contacted the graduate office at the University of Hull, which supported by forwarding the link and advert to participate to all postgraduate at the University of Hull – chosen as most likely candidates the University to support in a request to participate in a postgraduate research project. Whilst there was a bias to the opportunistic way these networks were identified (particularly the personal and University networks), these networks were chosen with most likely potential to respond to and decrease the bias which occurs when uptake is more limited. Furthermore, these three forums were also chosen to represent a potential widespread network which were unlikely to significantly overlap. Five-hundred and forty-nine potential participants accessed the study (as reported by the number of "counts" of individuals who left page 1 on the survey), although this is a maximum figure as some of the participants may have accessed this page on more than one occasion. This first stage of recruitment (n = 170) commenced on 17th August 2014, closing on 25th August 2015.

Recruitment began for the autistic and OCD participants from 1st October 2015. The gap between the first stage of neurotypical participant recruitment and the recruitment of the two clinical (disorder) groups was due to an initial decision to include clinical groups from local NHS clinics, which was later disregarded. On near completion of the NHS's ethics procedure (IRAS), it was ultimately decided a more widespread recruitment drive may be more representative of the wider clinical populations. During this second recruitment drive, initial analyses of the responses of neurotypical controls identified two items had been omitted from the Repetitive Behaviour Questionnaire assessment tool (see section 7.4.8). A second stage of recruitment of neurotypical participants (*n* = 30) was undertaken, repeating the above procedure of advertising via the lead researcher's personal Facebook account (see section 7.4.7).

Adult participants with OCD and autism were recruited through advertisement (see Appendices 6 and 7) relayed by their respective services. As an online study, a co-ordinated approach was taken to recruit as widespread a population for each of these groups as possible via online forums. For the autism participants, the two leading organisations for autism agreed to post these adverts: the National Autistic Society (NAS), which claims to be the leading UK charity for autistic people; and Research Autism, which claims to be the only UK charity exclusively dedicated to research into interventions in autism. Each NAS branch in England, Wales, Scotland and Northern Ireland working with adults on the autism spectrum (approximately 50 services) was contacted twice (the second as a reminder) to request permission to support in advertising the study. Subsequently, these groups mailed out the advert to service users and some advertised it on their websites. Research Autism advertised the study

on their site. Additionally, local (Hull and East Riding) services for adults with autism (Matthew's Hub, FiND and Autism Plus) advertised the study within their premises amongst their members. Finally, all relevant groups (with "autism, autistic or OCD" in their name) on Twitter and Facebook (in both the United Kingdom and the United States) were contacted with the advertisement information. Participants with OCD were recruited via contact on Facebook and Twitter to relevant OCD services in the United Kingdom and the United States. The national charity, OCD Action, advertised the study on their website. Additionally, adverts were placed via the charity AnxietyUK, reaching a potential membership base of around 6000 (magazine and website) and online social media following of more than 60,000 individuals. Whilst the exact reach of potential participants is unknown, the approach to recruit participants was a co-ordinated effort to reach the largest national and local online networks possible.

Despite wide distribution, recruitment was slow in the first three months. This is likely due to the large amount of time it could take for participants to complete the study: the 76 items would likely have taken a long time, particularly for the OCD and autism groups as more information was required for items which were positively endorsed. Although a potential 440 participants accessed this study (the number of "counts" on the first page), only 28 participants completed the questionnaire. Therefore, a slight amendment was made to the recruitment via advertisement of the study to include an incentive to participate (following a new successful Ethics Committee application). Subsequent participants were made aware of a new £50 cash prize randomly allocated to one individual. Contact was made again to all the aforementioned services and adverts were widely distributed as before. Following the addition of this incentive, 47 participants were recruited (27 with autism and 24 with OCD). One-thousand two-

hundred and five potential participants accessed this new questionnaire (approximately 553 for the survey advertised to services for people with autism and approximately 652 for the survey advertised for people with OCD). Following the second round of advertisements, to minimise the likelihood of participants completing the survey again, those who came forward to say that they had completed the questionnaire previously were included in the cash prize draw.

7.4.8. Procedure.

All elements of the study were accessed online, via Online Surveys (JISC, 2018). Participants were required to declare they had read and understood a comprehensive Participant Information Sheet (see Appendix 9) before giving consent to participate. Contact email addresses were supplied for the lead researcher and study supervisor for individuals to contact with any queries. Participants were required to self-report all information, including diagnosis. The restricted procedure (online-only access) increased the selection bias and potentially reduced the representativeness of the sample. Whilst this remained a limitation of the study, the online method was selected to reduce social desirability bias (and effects caused by social anxiety of autistic participants), whilst recruitment was aimed widely.

Thirty-nine adults with a self-reported diagnosis of an autism spectrum disorder and 35 adults with a self-reported diagnosis of obsessive-compulsive disorder completed the full version of the CaRT questionnaire. One-hundred-and-seventy neurotypical control participants completed version one of the Compulsive and Repetitive Traits (CaRT) questionnaire, which consisted of all the items as outlined in Appendices 1 and 2, with the exception of questions 17 (“I organise objects into categories according to various

characteristics such as colour, size or function") and 18 ("I line up or arrange objects") of the Repetitive Behaviour Questionnaire. These items were inadvertently omitted during the creation of the survey but amended before recruitment of the clinical groups. A further neurotypical control group (*N* = 30) was recruited to administer the full set of CaRT items; t-tests were performed to compare whether the two neurotypical control groups were equivalent. All questions were mandatory, which may have increased the drop-out rate but improved the statistical analyses.

7.4.9. Statistical analysis.

Data were analysed using the statistical software IBM SPSS 22.0 for windows. No data was excluded as the design of the online survey ensured this. However, as described in section 7.4.8, two questions from the Repetitive Behaviour Questionnaire were missing from the control group. To establish any differences between these two control groups, t-tests were performed on these interval level data, with "equal variances not assumed" reported where there was identified to be inequality of variance (using Levene's statistic), a process recommended by Brace, Kemp and Snelgar (2003).

To compare the mean results across the three groups of participants (based on disorder) on various measures of Repetitive Behaviours and OCD traits, one-way ANOVAs were performed. Field (2018, p.248) suggests tests of normality are irrelevant: for large samples they are unnecessary and for small samples they lack the power to detect non-normality. However, the sample size recruited was between these sizes, therefore data were explored for normality using Shapiro-Wilk test calculations (for relatively small samples). Where these analyses indicated the dataset

was not normally distributed, more robust analyses were performed via random sampling of the data (bootstrapping). Where relevant, these bootstrapped results are reported. This procedure was followed for analyses of the data for hypotheses 1 to 6.

To evaluate how participants felt about answering the questionnaire, qualitative answers were obtained. At the end of completing the study participants were asked to "explain any difficulties you had in answering any of the questions". This was included particularly to assess the validity of the mood-related questions. These responses were scored by the lead researcher by hand, blinded at first to the disorder group. To support the statistical analysis, each free response was coded based on the severity of how difficult the participants reported the questions were: 0 = blank (may relate to no difficulty, but also may be left by the participant who was keen to complete the experiment); 1 = none; 2 = minor difficulty with items; minor difficulty for personal reasons; 3 = major difficulty in items; and 4 = major difficulty for personal reasons. The full range of major difficulties are reported in Appendix 10. These coded responses were analysed as described above for hypotheses 1 to 6. Between group comparisons were calculated with Dunnett's T3 comparisons (bootstrapped).

To analyse the relationship between OCD traits and Repetitive Traits, bivariate correlations were performed for hypothesis 7. Additionally, T-tests were performed to compare the significance of the mean lower- and higher-order Repetitive Behaviours.

7.5. Results

This section provides the results of the comparative analyses of various measures of Compulsive and Repetitive Traits (in addition to analysis of the difficulties reported by

each participant to answering the questions) between the two clinical groups (autism and OCD) and the neurotypical control group. Following this results section, the implications of these results are presented in the discussion section, in context of the Compulsive and Repetitive Trait framework. First, definitions of key terms are outlined to provide meaning to the various datasets collected within this investigation.

Definition of key terms

Total number of Repetitive Behaviours/OCD traits: Count of all behaviours/traits reported for both past (ever) and current (within the last 7 days).

Weighted: To quantify differences between qualitative reports on frequency and severity, adjustments were made to the dataset as follows. With no standardised way to make adjusted calculations, these were rationally – rather than empirically – derived.

Weighted frequency: Each Repetitive Behaviour and OCD item endorsed as occurring either past or current was also rated for frequency. Weighted total relates to the count of the number of items after the following weighting was applied: counts of “often” (less than 1 hour per day) were multiplied by a factor of 1; counts of “frequently” (between 1 and 3 hours per day) were multiplied by a factor of 2; counts of “often” (between 3 and 8 hours per day) were multiplied by a factor of 3; and “very often” (more than 8 hours per day) were multiplied by a factor of 4.

Weighted mood: This weighted score was calculated by: multiplying the counts of “very good” and “very bad” by a factor of 2; adding the adjusted “very good” counts to

the “good” counts and the adjusted “very bad” scores to the “bad” scores; then subtracting the overall “bad” from the overall “good” scores. A positive number relates to an overall positive mood associated with these traits, and a negative number related to an overall negative mood associated with these traits.

Current behaviours/traits: Count of items reported as occurring “within the past 7 days”.

Lower-order Repetitive Behaviours: Sum of responses to questions 1 through to 3 on the Repetitive Behaviour Questionnaire (see Appendix 3), as recommended by Moss et al. (2009).

Higher-order Repetitive Behaviours: Sum of responses to questions 4 to 18 on the Repetitive Behaviour Questionnaire (see Appendix 5), as recommended by Moss et al. (2009).

Alternative questions: Measuring mood associated with a trait is challenging; indirect mood scores can contaminate direct measurement. For example, an individual may report positive mood to performing a compulsion as relief (indirect), whilst the overall feeling may be one of distress and/or anxiety (due to the overall unwanted nature of the act). To control for this effect, an alternative question was created for each item to assess both direct and indirect responses. For behaviours (compulsions) the indirect (alternative) question was “how do you feel when you are unable to perform this act?” For thoughts (obsessions), the indirect (alternative) question was “how does this make you feel if you do this?”

7.5.1. Comparison between the two control groups.

As it was likely the same participants could have been recruited in each of the two neurotypical groups, one of the data sets needed to be omitted. Parametric t-tests were computed for the Repetitive Behaviour analyses between the two control groups, to analyse whether the Repetitive Behaviour score was affected by the omission of the two Repetitive Behaviour items. If the data between the two group are not comparative, the data for the large group (with the missing items) would need to be removed from subsequent analyses.

Independent sample t-tests were run to compare Repetitive Behaviour scores between the control group 1 (*N* = 170) and control group 2 (*N* = 30). These analyses revealed no significant differences in any mean Repetitive Behaviour measure between control group 1 (*N* = 170) and control group 2 (*N* = 30), i.e.: total number of Repetitive Behaviours (*t* = -1.53, *p* = .14, two-tailed), equality variances not assumed; weighted mood of Repetitive Behaviours (*t* = -1.20, *p* = .24, two-tailed), equality variances not assumed; current Repetitive Behaviour score (*t* = -1.00, *p* = .32, two-tailed); and weighted frequency of Repetitive Behaviours (*t* = -1.57, *p* = .13, two-tailed). Of all the measures compared, only number of Repetitive Behaviours occurring under negative mood was significantly different between these two control groups (t = -2.3, *p* = 0.03, two-tailed); equality variances not assumed. To increase the power of the study, the analyses for the smaller control group (*N* = 30) was dropped in favour of retaining the data from the larger control group (*N* = 170): in all subsequent analyses, the control group refers only to this larger group.

7.5.2. Demographics.

Fischer's exact test of independence was performed (due to small expected cell frequencies) to examine the relationship between demographic characteristics and disorder. This relationship was significant for: gender ($\chi^2 = 12.6$, $p = .002$); country of residence ($\chi^2 = 19.2$, $p < .001$); and first language ($\chi^2 = 7.98$, $p = .009$). Chi-square analyses indicated a significant relationship between disorder and age ($\chi^2 = 26.5$, $p = .001$). Fischer's exact test revealed no significant association between disorder and ethnicity i.e. Caucasian ($\chi^2 = 3.34$, $p > .05$).

Two-hundred and forty-four adult participants completed the study, consisting of 170 neurotypical controls, 39 participants with a self-reported diagnosis of autism and 35 with a self-reported diagnosis of OCD. There was a fairly even distribution of participants across the age ranges (see Figure 7.3). Within the autism sample, 21 (54%) were male, whereas 7 (20%) and 44 (26%) of the OCD and control sample, respectively, were male. Figure 7.3 demonstrates the age distribution across the groups as percentages as age responses were provided within age brackets, rather than as an actual value. As Figure 7.2 illustrates, relatively few (15; 9%) control participants were between 18- to 25-years, compared to the OCD and autism groups (12 and 10; 34% and 26%, respectively). A disproportionate distribution of participants within the control group (21; 12%) were 60-years plus, compared to the OCD (1; 3%) and autism group (2; 5%). A slight administrative error is worth noting: two of the age brackets inadvertently include 45 years. Whilst relatively few participants with autism and controls resided outside of the United Kingdom (4 and 6; 10% and 4%, respectively), a much larger proportion of the participants with OCD resided outside of the United Kingdom (10; 29%). Within the OCD group, these additional countries of residence

consist of: United States (*n* = 5); Taiwan; Canada; Italy; Ukraine; and India. There was a similar trend for first language, with relatively few participants with autism and controls speaking a language other than English (2 and 4; 5% and 2%), whereas a larger proportion of participants with OCD spoke an alternative first language (5; 14%).

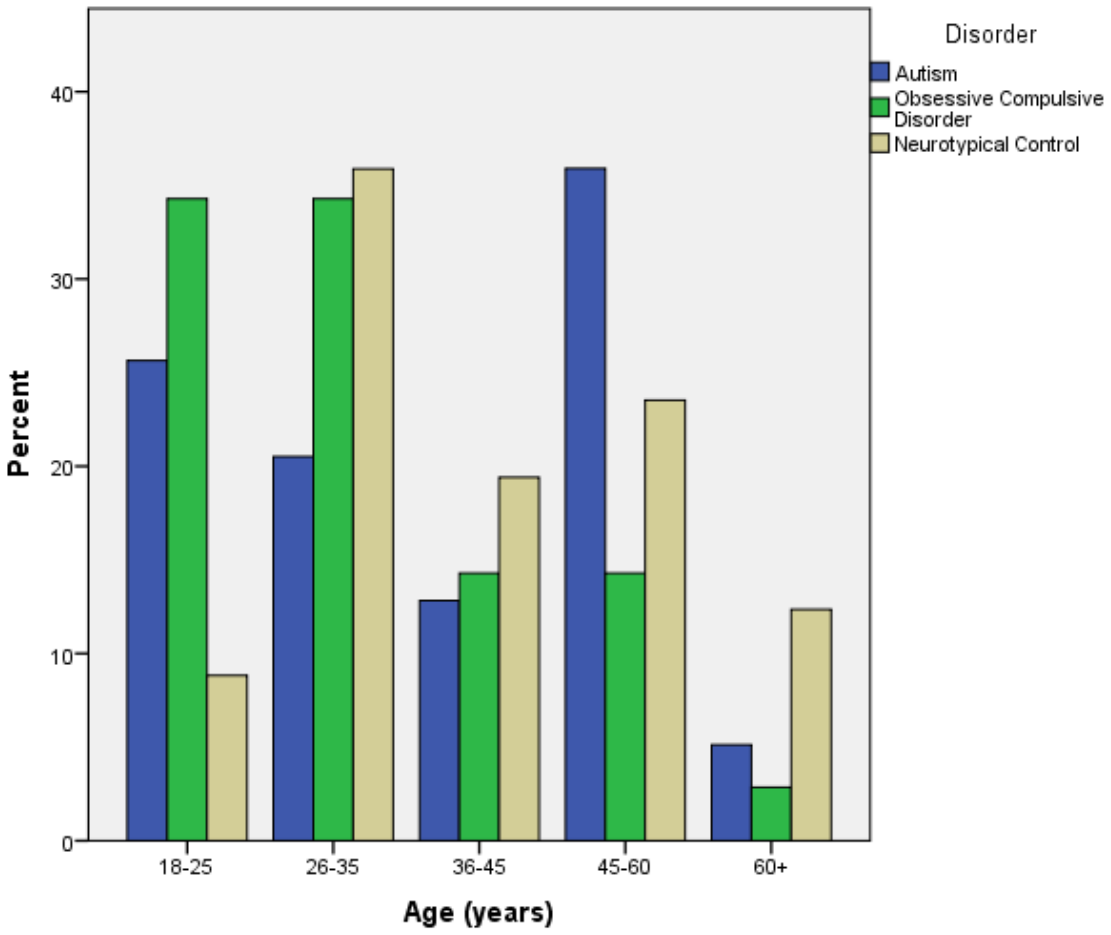

Figure 7.2. Percentage number of participants within each age bracket for each disorder.

Table 7.1. *Frequencies and percentages for demographic variables.*

Demographic variable	Category	Disorder		
		Autism (n = 39)	OCD (n = 35)	Neurotypical (n = 170)
Age	18-25 years	10 (25.6)	12 (34.3)	15 (8.8)
	26-35 years	8 (20.5)	12 (34.3)	61 (35.9)
	36-45 years	5 (12.8)	5 (14.3)	33 (19.4)
	45-60 years	14 (35.9)	5 (14.3)	40 (23.5)
	60+ years	2 (5.1)	1 (2.9)	21 (12.4)
Gender	Male	21 (53.8)	7 (20)	44 (25.9)
	Female	18 (46.2)	28 (80)	126 (74.1)
Country	United Kingdom	35 (89.7)	25 (71.4)	164 (96.5)
	Other	4 (10.3)	20 (28.6)	6 (3.5)
Language	English	37 (94.9)	30 (85.7)	166 (97.6)
	Other	2 (5.1)	5 (14.3)	4 (2.4)
Ethnicity	White (British)	34 (87.2)	25 (71.4)	157 (92.4)
	White (Irish)	0 (0)	0 (0)	2 (1.2)
	Other white	4 (10.3)	6 (17.1)	4 (2.4)
	Other black	0 (0)	0 (0)	1 (0.6)
	Indian	0 (0)	1 (2.9)	0 (0)
	Chinese	0 (0)	1 (2.9)	0 (0)
	Mixed	1 (2.6)	0 (0)	6 (3.5)
	Other	0 (0)	2 (5.7)	0 (0)

Percentages appears in parentheses.

7.5.3. Overview of Compulsive and Repetitive Traits.

The data was first observed for signs of skew. These observations were made on created Q-Q plots, as suggested by Field (2018, p.250). Compared to the expected quartiles, there were no signs of significant skew in any of the Q-Q plots of the Repetitive Behaviour or OCD trait scores displayed in Table 7.1. Therefore, all averages are reported as means and standard deviations (rather than median and interquartile ranges). For weighted mood scores, negative scores indicate low mood (such as distress or anxiety) and positive scores indicate high mood (such as pleasure). Total

number of Repetitive Behaviour scores are out of a maximum score of 18 items. Total number of OCD traits is out of a maximum score of 58 items. It is notable the standard deviation across most of these scores are high, demonstrating a huge variability in the scores within each of the samples and indicating the sample means are likely to not to accurately reflect the population mean for each disorder (Field, 2018, p.64).

Table 7.2. *Means and standard deviations for Compulsive and Repetitive Traits scores.*

Measure	Variable	Disorder		
		Autism (n = 39)	OCD (n = 35)	Neurotypical (n = 170)
Repetitive Behaviour	Total number	7.79 (4.19)	6.71 (5.44)	2.09 (2.64)
	Total number of current	4.51 (3.07)	4.37 (4.55)	1.26 (2.18)
	Weighted frequency	16.1 (10.2)	15.3 (15.3)	3.94 (6.73)
	Weighted mood	5.92 (5.63)	2.77 (6.95)	1.46 (2.74)
	Total number associated with negative mood	0.38 (0.54)	1.37 (2.20)	0.047 (0.24)
OCD trait	Total number	16.4 (9.89)	24.5 (13.3)	6.36 (6.61)
	Weighted frequency	27.5 (21.4)	49.3 (36.5)	8.83 (13.0)
	Weighted mood score	-6.18 (6.52)	-17.5 (12.0)	-1.94 (3.92)
	Total number occurring under positive or neutral mood	7.94 (5.86)	9.17 (7.41)	3.59 (4.05)
	Alternative question total number thoughts positive or neutral mood	1.98 (1.98)	2.49 (3.37)	0.96 (1.19)
	Alternative question total number behaviours positive or neutral mood	1.18 (1.64)	1.03 (1.81)	0.69 (1.21)

Standard deviation appears in parentheses.

7.5.4. Frequency and mood of Repetitive Behaviours (hypotheses 1 and 2).

Firstly, it was hypothesised measures of Repetitive Behaviours (as measured by the RBQ, Moss et al., 2009) would be significantly highest in the autism group, whilst the OCD and control groups' scores would be comparable. Secondly, Repetitive Behaviour scores were predicted to be associated with positive mood.

Figure 7.3 illustrates the distribution of Repetitive Behaviours reported by the two clinical disorder groups. For the sample of participants with autism, the distribution is fairly normal, largely centred around a midpoint. However, the distribution of Repetitive Behaviours for the OCD group was skewed more towards reporting fewer Repetitive Behaviours than the autism group. The large variability is affected by the four participants with OCD who endorsed 17 and 18 (the maximum) of the Repetitive Behaviours.

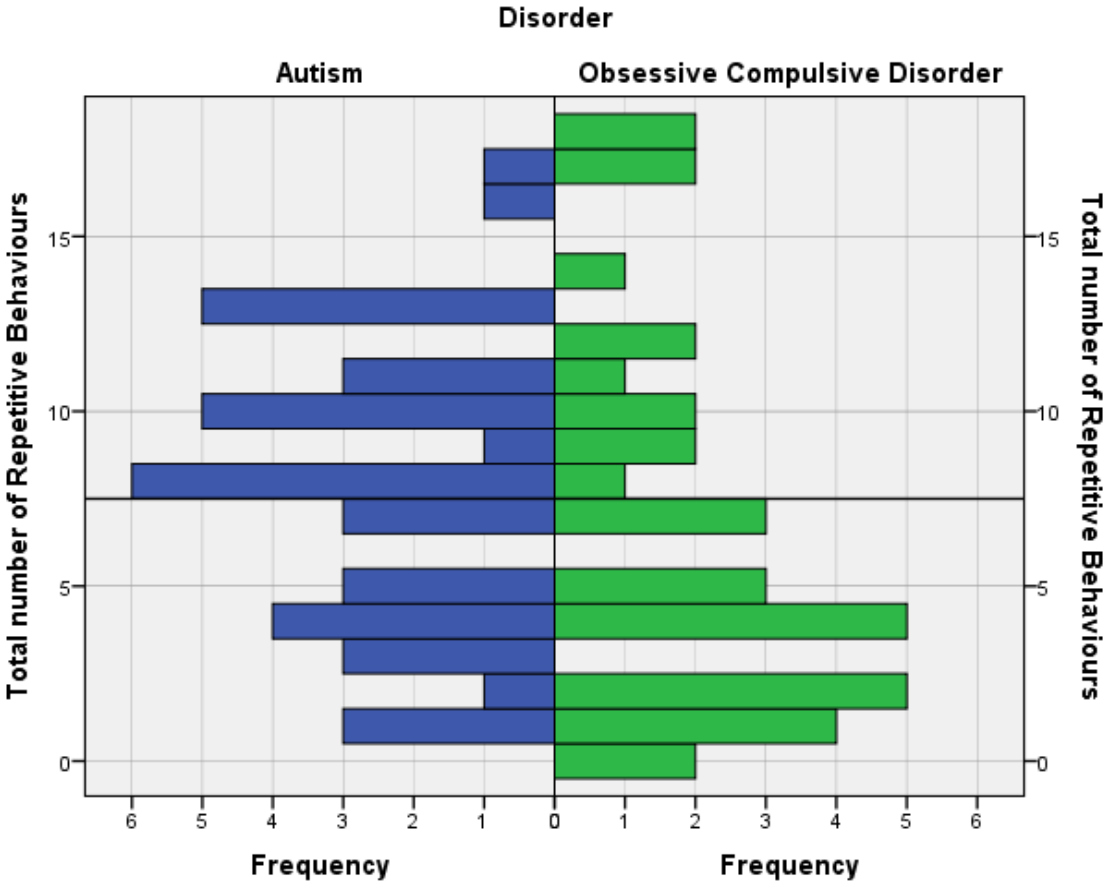

Figure 7.3. Population pyramid frequency total number of Repetitive Behaviours by disorder.

As Figure 7.4 illustrates, the range of weighted mood scores of Repetitive Behaviours was much larger for the autism group, which is in line with the second hypothesis. All the groups contained outliers, with the neurotypical group (disorder: none) showing five extreme scores (illustrated by the asterisks), potentially a reflection of undiagnosed disorders in this group (see discussion). For the autism group, mood was all within the positive range. However, for the OCD group and the neurotypical group all the lower quartile contained negative (such as low, bad, distress or anxiety).

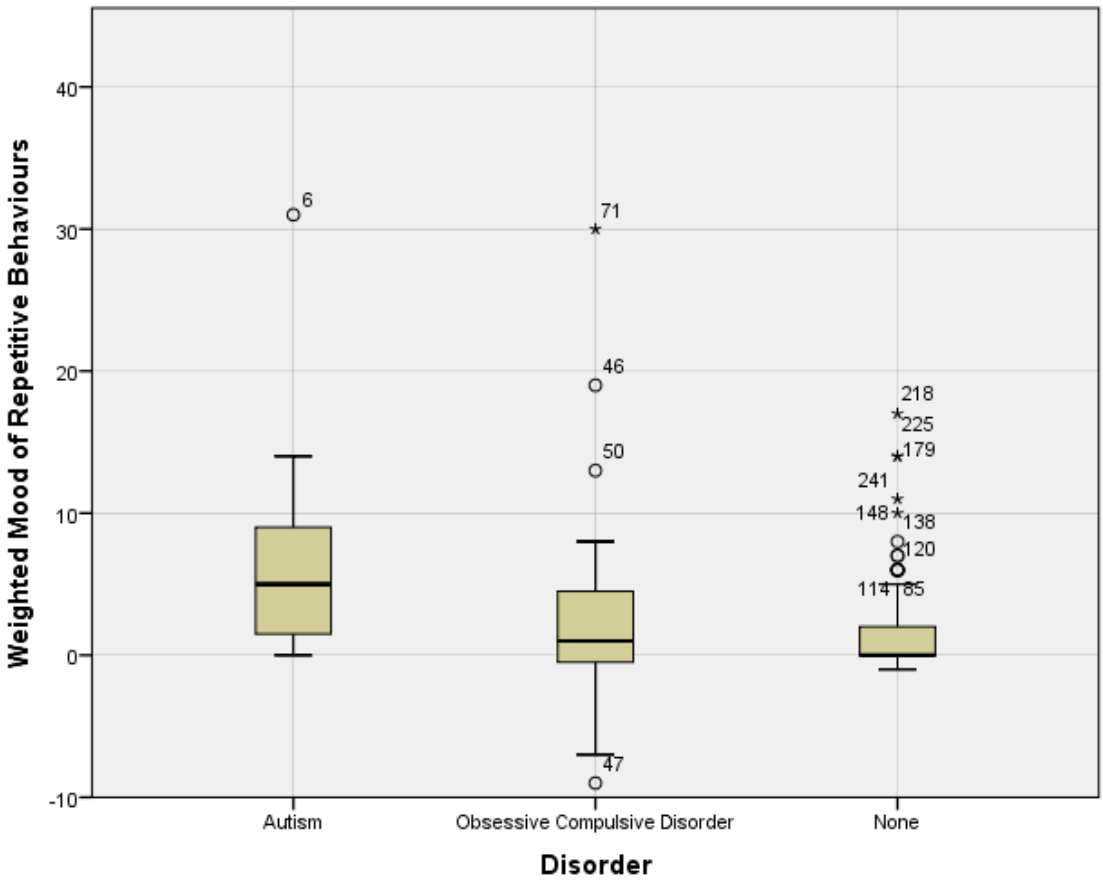

Figure 7.4. Simple boxplot of weighted mood of Repetitive Behaviours by disorder.

The data only partially supported the first hypothesis, where it was expected Repetitive Behaviours would only be significantly elevated in autism and comparable between the OCD and the neurotypical control groups. One-way analysis of variance (ANOVA) were performed to compare differences in the means between the number, frequency and mood of Repetitive Behaviours between the three groups. Group differences were identified for total number ($F(2, 241) = 59.6$, $p < .001$, $\eta_p^2 = .33$), currently occurring ($F(2, 241) = 33.7$, $p < .001$, $\eta_p^2 = .22$) and weighted frequency of Repetitive Behaviours ($F(2, 241) = 44.2$, $p < .001$, $\eta_p^2 = .27$). Group difference were also identified for both weighted mood score ($F(2, 241) = 18.7$, $p < .001$, $\eta_p^2 = .14$), alternative question weighted mood score ($F(2, 241) = 41.2$, $p < .001$, $\eta_p^2 = .26$) and total number of Repetitive Behaviours associated with negative mood ($F(2, 241) = 33.4$, $p < .001$, $\eta_p^2 = .22$). Bootstrapped Dunnett T3 post-hoc comparisons were

performed to control for non-normally distributed data (see Table 7.3). These analyses indicated both the autism (*M* = 16.1) and the OCD group (*M* = 15.3) reported more frequent Repetitive Behaviours than the control group (*M* = 3.94).

Repetitive Behaviours were expected to be significant to autism, therefore it was expected these traits would be highest in number in this disorder (hypothesis 1). These analyses revealed no significant difference ($p > .05$) between the autism group and OCD group for total number, number of current or weighted frequency of Repetitive Behaviours.

If Repetitive Behaviours are ego-syntonic they are likely to be related to positive mood (hypothesis 2). The data suggested Repetitive Behaviours were associated with more positive mood, specifically in the autism group. Weighted mood was significantly more positive for the autism group (*M* = 5.92) than the OCD group (*M* = 2.77). Notably, there were no significant differences ($p > .05$) identified for weighted mood score between the OCD group (*M* = 2.77) and the control group (*M* = 1.46).

Finally, if Repetitive Behaviours are solely ego-syntonic, it would be expected no participants would report negative mood associated with these traits. However, all three groups reported some negative mood for Repetitive Traits. For the control group this seemed minimal, with an average of *M* = 0.047 items (out of 18 Repetitive Behaviour items) endorsed as associated with negative mood. Bootstrapped analyses identified the OCD group reported most items associated with negative mood (*M* = 1.37), which was significantly higher than in the autism (*M* = 0.38) group. The autism

group reported significantly more Repetitive Behaviours associated with negative mood than the control group.

In summary, the data only partially supported hypothesis 1. Whilst the autism and OCD samples reported significantly more Repetitive Behaviours than the control group, there were no significant differences in these traits between the autism and OCD group (number or frequency). However, the evidence is largely in line with the Hypothesis 2. Repetitive Behaviour mood was significantly more positive for the autism group, compared to both the OCD and the control group.

Table 7.3. *Bootstrapped post-hoc Dunnett's T3 comparisons for Repetitive Behaviour scores.*

Repetitive Behaviour scores	Comparison	Mean difference	Standard error of difference	95% Confidence Interval[a]
Total number	ASD vs. OCD	1.08	1.14	(-1.23,3.19)
	ASD vs. Control	5.71*	0.71	(4.38,7.14)
	OCD vs. Control	4.63*	0.94	(2.94,6.68)
Total number of current	ASD vs. OCD	0.14	0.91	(-1.69,1.87)
	ASD vs. Control	3.25*	0.51	(2.32,4.28)
	OCD vs. Control	3.11*	0.79	(1.71,4.79)
Weighted frequency	ASD vs. OCD	0.82	3.06	(-5.30,6.94)
	ASD vs. Control	12.16*	1.70	(9.08,15.6)
	OCD vs. Control	11.3*	2.65	(6.58,16.9)
Weighted mood	ASD vs. OCD	3.15*	1.53	(0.091,6.31)
	ASD vs. Control	4.46*	0.92	(2.85,6.47)
	OCD vs. Control	1.31	1.23	(-0.83,4.00)
Total number associated with negative mood	ASD vs. OCD	-0.99*	0.37	(-1.75,-0.28)
	ASD vs. Control	0.34*	0.09	(0.17,0.50)
	OCD vs. Control	1.32*	0.37	(0.66,2.10)

[a] based on 1000 bootstraps

* $p < .05$

7.5.5. Frequency and mood of obsessive compulsive traits (hypotheses 3 and 4).

OCD-traits were expected to be significantly highest in the OCD group, whilst the autism group were expected to report significantly more OCD-traits than the neurotypical controls (hypothesis 3). Furthermore, OCD traits were expected to be associated with negative mood, highest again in the OCD group, followed by the autism and then the neurotypical controls (hypothesis 4).

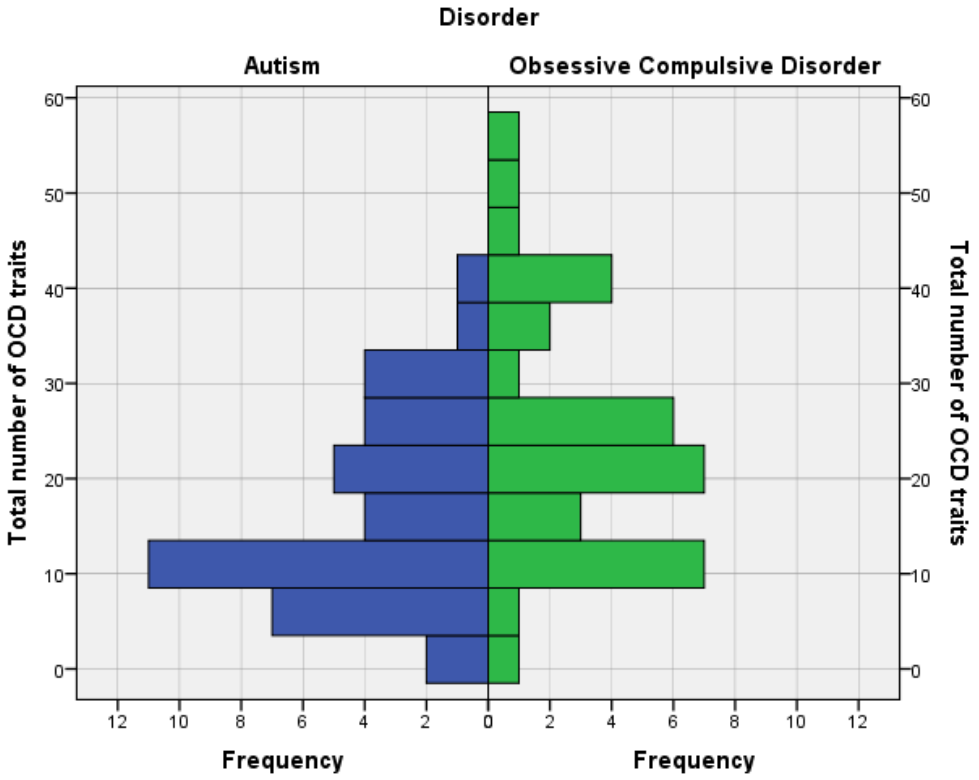

Figure 7.5. Population pyramid frequency total number of OCD traits by disorder.

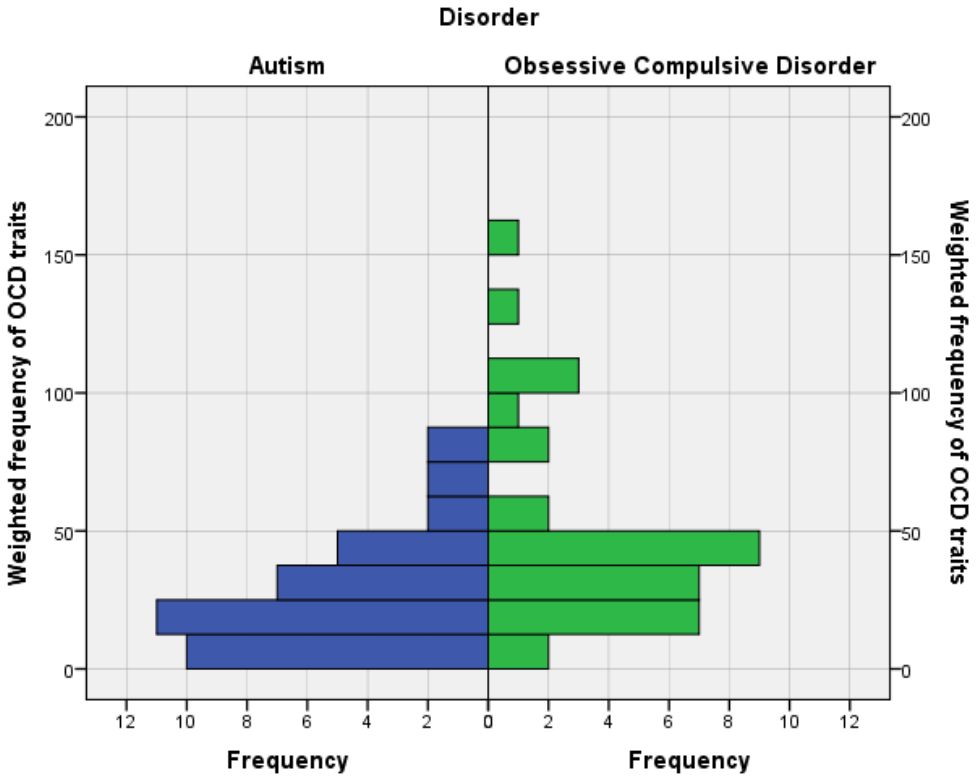

Figure 7.6. Population pyramid frequency illustrating weighted frequency of OCD traits by disorder.

For total number of OCD traits, the frequency pyramid demonstrated the opposite pattern to the Repetitive Behaviours (see Figure 7.5). For this dataset, the distribution of OCD traits was fairly normal for the OCD group, whereas it was more skewed towards fewer reported OCD traits in the autism group. For (weighted) frequency (see Figure 7.6), both clinical groups demonstrated a negative skew of distribution towards more participants reporting less frequent OCD traits. Participants with autism more frequently reported fewer OCD traits (more negatively skewed), with the OCD group containing more extreme scores of frequency of OCD traits.

In line with hypothesis 4, the converse pattern to that identified for Repetitive Behaviours was identified for mood of OCD traits. As can be seen in Figure 7.7, participants with OCD endorsed more negative mood (such as distress or anxiety) for OCD traits than both other groups. Extreme scores (illustrated by asterisks) for the

neurotypical group inflated the negative scores towards low mood in the lower quartile. However, it was notable the neurotypical and – this time – the autism group contained reported positive mood of OCD traits (most of the upper quartile when endorsed by both groups). Very few of the OCD traits were associated with positive mood (as would be expected, being clinically ego-dystonic), in the OCD group.

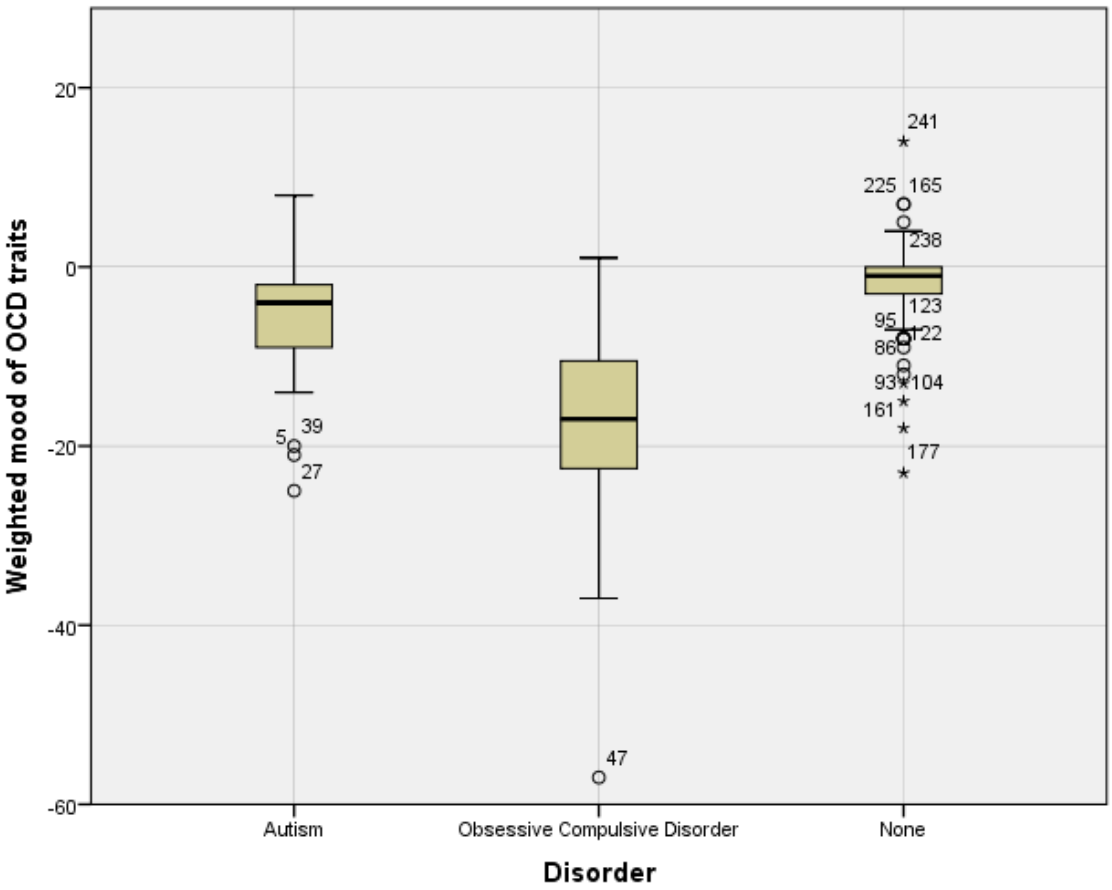

Figure 7.7. Simple boxplot of weighted mood of OCD traits by disorder.

One-way ANOVAs indicated group difference for total number of OCD traits ($F(2, 241) = 78.1$, $p < .001$, $\eta_p^2 = .39$) and weighted frequency of OCD traits ($F(2, 241) = 68.9$, $p < .001$, $\eta_p^2 = .36$). Group differences were identified for total number of OCD traits occurring under positive or neutral mood ($F(2, 241) = 26.0$, $p < .001$, $\eta_p^2 = .18$) and total

number of OCD thoughts occurring under positive or neutral mood for alternative questions ($F(2, 241) = 13.5$, $p < .001$, $\eta_p^2 = .10$). However, there were no group differences identified in total number of OCD behaviours occurring under positive or neutral mood for alternative questions ($F(2, 241) = 2.49$, $p = .085$, $\eta_p^2 = .02$).

The data generally supported hypothesis 3. With evidence of comorbidity between OCD and autism, it was expected OCD traits would be highest in OCD, but significantly higher in autism than the neurotypical control group. Bootstrapped post-hoc comparisons for these analyses are presented in Table 7.4. The OCD group ($M = 24.5$) reported significantly more ($p < .05$) OCD traits than both the autism group ($M = 16.4$) and the control group ($M = 6.36$), whilst the autism group also reported significantly more OCD traits than the control group ($p < .05$). Similarly, the OCD group ($M = 49.3$) reported spending significantly more time ($p < .05$) engaged in OCD traits than both the autism ($M = 27.5$) and the control group ($M = 8.83$), with the autism group also reporting significantly more time spent engaging in OCD traits than the control group ($p < .05$).

Similarly, the data generally supported hypothesis 4. Mood scores of OCD traits followed an identical pattern. The OCD group ($M = -17.5$) experienced significantly ($p < .05$) more negative mood associated with OCD traits compared to both the autism group ($M = -6.18$) and the control group ($M = -1.94$), whilst the autism group reported significantly more negative mood for OCD traits compared to the control group ($p < .05$). These significant group differences also remained for the revised OCD trait mood scores (see Table 7.1 for descriptive statistics).

However, the ego-dystonic (unwanted and unpleasant) nature of OCD traits suggests these traits should only be related to negative mood. As illustrated in Table 7.1, OCD traits associated with positive mood were identified for all groups. To control for positive mood as an indirect effect, alternative question responses were analysed (see definition of key terms, section 7.5). Bootstrapped Dunnett T3 analyses revealed no significant differences between the OCD and autism groups for any OCD trait (obsession or compulsion) for initial or alterative question ($p > 0.5$). However, both OCD and autism groups reported significantly more OCD traits associated with positive mood than the control group (see Table 7.3). For initial response ("how do you feel when you do this"), the OCD ($M = 9.17$) and the autism group ($M = 7.94$) reported significantly more OCD traits occurring under positive or neutral mood than the control group ($M = 3.59$). For the alternative response ("how you feel when this happens"), the OCD ($M = 2.49$) and the autism group ($M = 1.98$) reported significantly more OCD traits occurring under positive or neutral mood than the control group ($M = 0.96$).

In summary, the data only largely supported hypothesis 3. The OCD samples reported significantly more OCD traits than the autism group, who in turn reported significantly more OCD traits than the control group. The evidence is largely in line with hypothesis 4, with OCD trait mood was significantly more negative for the OCD group, compared to both the autism and the control group.

Table 7.4. *Bootstrapped post-hoc Dunnett's T3 comparisons for frequency of OCD trait scores.*

OCD trait score	Comparison	Mean difference	Standard error of difference	95% Confidence Interval[a]
Total number	ASD vs. OCD	-8.13*	2.76	(-13.7,-2.50)
	ASD vs. Control	10.0*	1.68	(6.76,13.4)
	OCD vs. Control	18.1*	2.32	(13.8,22.9)
Weighted frequency	ASD vs. OCD	-21.8*	7.16	(-36.3,-8.02)
	ASD vs. Control	18.7*	3.58	(12.2,26.4)
	OCD vs. Control	40.5*	6.37	(28.9,36.3)
Weighted mood score	ASD vs. OCD	11.4*	2.33	(6.81,15.94)
	ASD vs. Control	-4.24*	1.08	(-6.64,-2.14)
	OCD vs. Control	-15.6*	2.07	(-19.8,-11.4)
Weighted revised mood score	ASD vs. OCD	14.8*	3.60	(7.33,21.3)
	ASD vs. Control	-9.97*	1.83	(-13.9,-6.51)
	OCD vs. Control	-24.8*	3.07	(-31.0,-18.7)
OCD traits occurring under positive or neutral mood	ASD vs. OCD	-1.22	1.61	(-4.67,1.73)
	ASD vs. Control	4.35*	0.97	(2.50,6.31)
	OCD vs. Control	5.58*	1.33	(3.09,8.59)
Alternative question total number thoughts positive or neutral mood	ASD vs. OCD	-0.51	0.67	(-1.98,0.60)
	ASD vs. Control	1.02*	0.32	(0.38,1.62)
	OCD vs. Control	1.53*	0.59	(0.58,2.84)

[a] based on 1000 bootstraps

* $p < .05$

7.5.6. Higher- and lower-order Repetitive Behaviour differences (hypothesis 5).

Higher-order Repetitive Behaviours (i.e. those which require more cognitive demand, as opposed to more motor/sensory traits) were predicted to be associated with elevated mood. This was expected both in comparison to lower-order Repetitive Behaviours and specific to the autism-group.

Table 7.5 illustrates the descriptive statistics for the lower- and higher-order Repetitive Behaviours. Group difference were identified for both number of lower-order

Repetitive Behaviours ($F(2,241) = 25.6$, $p < .001$, $\eta_{p2} = .18$) and number of higher-order Repetitive Behaviours ($F(2,241) = 59.3$, $p < .001$, $\eta_{p2} = .33$).

As the Repetitive Behaviour measure contained only 3 items for lower-order Repetitive Behaviours (compared to 15 for higher-order Repetitive Behaviours), average values were calculated to allow for simpler comparison. These (number of Repetitive Behaviour) average values were calculated by dividing the lower-order Repetitive Behaviour scores by a factor of three and dividing the higher-order Repetitive Behaviour scores by a factor of 15: a score of -1 would mean all items were associated with a negative mood and a score of 1 would mean all items were associated with a positive mood. Group differences were identified for mood of lower-order Repetitive Behaviours ($F(2,241) = 4.97$, $p = .008$, $\eta_{p2} = .040$), but not for mood of higher-order Repetitive Behaviours ($F(2,241) = 1.02$, $p = .36$, $\eta_{p2} = .008$). However, for the alternative questions, group differences were identified for both higher-order Repetitive Behaviours ($F(2,241) = 34.4$, $p < .001$, $\eta_{p2} = .22$) and mood of lower-order Repetitive Behaviours ($F(2,241) = 22.0$, $p < .001$, $\eta_{p2} = .15$).

The autism-specific effect was rejected, with the higher-order Repetitive Behaviours being generally comparable between both clinical groups. As illustrated by the bootstrapped post-hoc comparisons (see Table 7.6), number of higher-order Repetitive Behaviours were significantly higher ($p < .05$) in both the autism group ($M = 5.95$) and the OCD group ($M = 5.31$) than the control group ($M = 1.37$). Similarly, number of higher-order Repetitive Behaviours were significantly higher ($p < .05$) in the autism group ($M = 1.85$) and the OCD group ($M = 1.4$), than the control group ($M = 0.72$). In contrast to group differences, these post-hoc analyses identified no significant

differences in mood for either lower-order or higher-order Repetitive Behaviours between any of the groups ($p > .05$). For alternative questions, there was no significant difference in mood for lower-order Repetitive Behaviours between the autism group ($M = -0.27$) and the OCD group ($M = -0.31$), whilst mood was significantly lower compared to the control group ($M = -0.039$) for both the autism and OCD groups ($p < .05$). This trend was also identified for mood related to higher-order alternative questions.

In summary, the evidence is largely in line with the null hypothesis for Hypothesis 5, with no autism-specific effects identified for the higher- and lower-order Repetitive Behaviours, signified by no significant differences reported in mood between the autism and OCD group.

Table 7.5. *Means and standard deviations for lower- and higher-order repetitive behaviour traits.*

Measure	Variable	Disorder		
		Autism (n = 39)	OCD (n = 35)	Neurotypical (n = 170)
Lower-order Repetitive Behaviour traits	Total number	1.85 (1.11)	1.40 (1.22)	0.72 (0.86)
	Average mood	0.56 (1.19)	0.06 (1.17)	0.10 (0.49)
	Average mood (alternative question)	-0.82 (1.30)	-0.94 (1.24)	-0.12 (0.54)
Higher-order Repetitive Behaviour traits	Total number	5.95 (3.51)	5.31 (4.56)	1.37 (2.12)
	Average mood	3.26 (2.66)	2.31 (2.73)	0.75 (1.67)
	Average mood (alternative question)	0.26 (0.75)	-0.06 (0.34)	-0.01 (0.26)

Standard deviation appears in parentheses.

Table 7.6. *Bootstrapped post-hoc Dunnett's T3 comparisons for higher- and lower-order Repetitive Behaviours.*

Repetitive Behaviour scores	Comparison	Mean difference	Standard error of difference	95% Confidence Interval[a]
Number of lower-order	ASD vs. OCD	0.47	0.27	(-0.058,0.99)
	ASD vs. Control	1.13*	0.20	(0.75,1.53)
	OCD vs. Control	0.68*	0.22	(0.28,1.11)
Number of higher-order	ASD vs. OCD	0.63	1.00	(-1.24,2.47)
	ASD vs. Control	4.58*	0.59	(3.51,5.80)
	OCD vs. Control	3.94*	0.79	(2.52,5.63)
Average mood of lower-order traits	ASD vs. OCD	0.17	0.093	(-0.018,0.35)
	ASD vs. Control	0.13	0.066	(-0.002,0.27)
	OCD vs. Control	-0.040	0.068	(-0.17,0.10)
Average mood of higher-order traits	ASD vs. OCD	-0.029	0.062	(-0.16,0.091)
	ASD vs. Control	0.22	0.051	(-0.085,0.12)
	OCD vs. Control	-0.051	0.040	(-0.029,0.13)
Average mood of lower-order alternative question traits	ASD vs. OCD	0.041	0.097	(-0.17,0.23)
	ASD vs. Control	-0.23*	0.070	(-0.37,-0.090)
	OCD vs. Control	-0.28*	0.070	(-0.40,-0.13)
Average mood of higher-order alternative question traits	ASD vs. OCD	0.044	0.060	(-0.070,0.16)
	ASD vs. Control	-0.18*	0.039	(-0.26,-0.10)
	OCD vs. Control	-0.22*	0.048	(-0.33,-0.14)

[a] based on 1000 bootstraps

* $p < .05$

The data also failed to support the predicted difference in mood between the higher- and lower-order Repetitive Behaviours. No significant differences ($p < 0.05$) were identified in difference in mood between these two constructs for either of the clinical groups (see Table 7.6). Only the control group demonstrated a significant difference ($p < .05$) in mood between the lower- and higher-order Repetitive Behaviours, with a higher mood score for lower- ($M = 0.059$) compared to higher-order ($M = -0.004$) Repetitive Behaviours.

Table 7.7*. T-tests for higher- and lower-order Repetitive Behaviours.*

Repetitive Behaviour scores	Disorder	Mean difference	Standard error of difference	95% Confidence Interval[a]
Number of lower-order versus number of higher-order	Autism	0.17	0.10	(-0.031,0.38)
	OCD	-0.029	0.092	(-0.20,0.14)
	Control	-0.020	0.014	(-0.048,0.007)
Mood of lower-order versus mood of higher-order	Autism	0.17	0.10	(-0.031,0.38)
	OCD	-0.03	0.092	(-0.20,0.14)
	Control	0.06*	0.023	(0.017,0.12)

[a] based on 1000 bootstraps

7.5.7. Group differences for Social Context (hypothesis 6).

Table 7.8*. Means and standard deviations for Repetitive Behaviours occurring during differing social contexts.*

Measure	Variable	Disorder		
		Autism (n = 39)	OCD (n = 35)	Neurotypical (n = 170)
Repetitive Behaviours when alone	Mean	1.56 (2.17)	1.23 (2.26)	0.31 (1.23)
Repetitive Behaviours when around people	Mean	1.41 (2.04)	0.83 (1.71)	0.23 (1.23)

Standard deviation appears in parentheses.

Table 7.9. *Bootstrapped post-hoc Dunnett's T3 comparisons for Repetitive Behaviours during different social contexts.*

Repetitive Behaviour scores	Comparison	Mean difference	Standard error of difference	95% Confidence Interval[a]
Repetitive Behaviours when alone	ASD vs. OCD	0.34	0.51	(-1.32,0.72)
	ASD vs. Control	1.25*	0.35	(0.63,2.00)
	OCD vs. Control	0.92*	0.39	(0.20,1.71)
Repetitive Behaviours when around people	ASD vs. OCD	0.58	0.45	(-0.31,1.42)
	ASD vs. Control	1.12*	0.38	(0.52,1.78)
	OCD vs. Control	0.54	0.31	(-0.03,1.21)

[a] based on 1000 bootstraps

* $p < .05$

The negative effect of social context was predicted to be autism-specific. However, this was not demonstrated by the data (see Tables 7.8 and 7.9). Whilst group difference were identified for Repetitive Behaviours occurring when alone ($F(2,241) = 12.6$, $p < .001$, $\eta_{p2} = .095$), and occurring when around other people ($F(2,241) = 9.77$, $p < .001$, $\eta_{p2} = .075$), bootstrapped Dunnett's T3 post-hoc comparisons revealed no significant difference between the autism and OCD group in number of Repetitive Behaviours occurring when either alone ($p > .05$, 95% CI [-1.32,0.72]), or when around people ($p > .05$, 95% CI [-0.31,1.42]). Compared to the control group, the autism group reported significantly more Repetitive Behaviours when both alone ($p < .05$, 95% CI [0.63, 2.00]) and when around people ($p < .05$, 95% CI [0.52, 1.78]). Whilst the OCD group reported significantly more Repetitive Behaviours compared to the control group when alone ($p < .05$, 95% CI [0.20, 1.71]), there was no significant difference between the OCD and control group in the number of Repetitive Behaviours when around people ($p > .05$, 95% CI [-0.03, 1.21]).

7.5.8. Correlation between Repetitive Behaviours and Obsessive-Compulsive Disorder traits (hypothesis 7).

Finally, there was expected to be no correlation between Repetitive Behaviours and OCD traits (hypothesis 7). However, Spearman's rho calculations identified, across all participants, a strong correlation between total Repetitive Behaviour and total OCD trait scores ($r(244) = .691$, $p < .001$). Additionally, a small negative correlation was identified between mood associated with Repetitive Behaviours and mood associated with OCD traits ($r(244) = -.130$, $p = .042$). This correlation indicated a small association between positive mood of negative Repetitive Behaviour, with negative mood of OCD traits. Figure 7.8 demonstrates an almost identical interaction for all three groups, as designated by the similar steepness of the lines of best fit for the OCD, autism and neurotypical (disorder: none) groups.

There was found to be a strong correlation between number of Repetitive Behaviour and number of OCD trait scores for each of the groups: autism ($r(39) = .647$, $p < .001$); OCD ($r(35) = .610$, $p < .001$); and the control group ($r(170) = .535$, $p < .001$). However, no significant correlations were identified between the mood associated with Repetitive Behaviours and mood associated with OCD traits between for any of these groups: autism ($r(39) = -.053$, $p = .748$); OCD ($r(35) = -.185$, $p = .289$); or the control group ($r(170) = -.103$, $p = .182$).

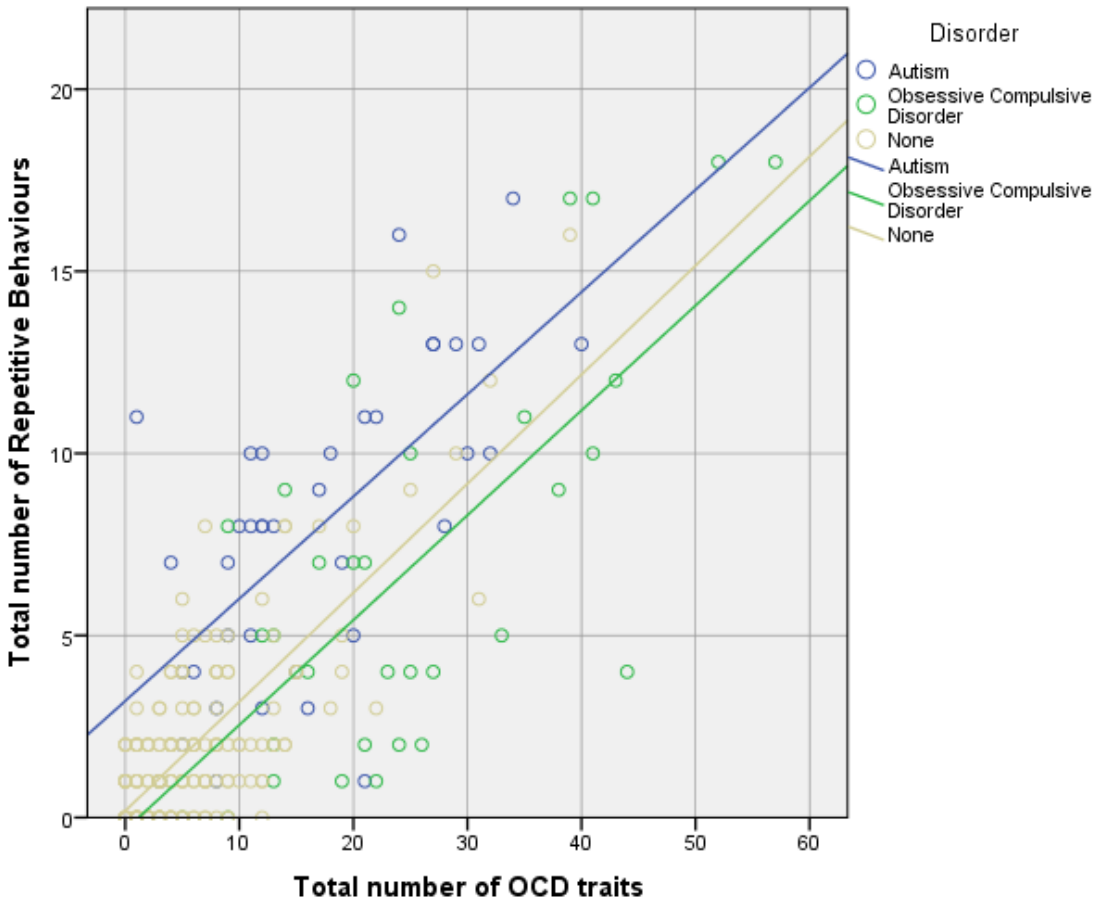

Figure 7.8. Grouped scatterplot of total number of Repetitive Behaviours by total number of OCD traits by disorder.

7.5.9. Difficulty answering the questions

Bootstrapped post-hoc comparisons were performed for these analyses and are presented in Table 7.11. There were no significant differences in the reported difficulties in answering the items between the autism and the OCD groups ($p > .05$). However, both groups reported significantly more difficulties in answering the questions than the control group ($p < .05$). For example, as can be seen in the descriptive statitics in Table 7.10, a high proportion of control participants reported no ("blank" or "none") difficulties answering questions (132 or 77.7% of control participants), compared to both clinical groups, with only 18 (46.2%) autistic participants and 16 (45.7%) of OCD participants reporting such difficulties.

Table 7.10. *Number and percentage of participants who reported difficulties in answering items.*

Reported Difficulty	Percentage		
	Autism Group (n = 39)	OCD Group (n = 35)	Neurotypical Group (n = 170)
Blank	15 (38.5)	12 (34.3)	103 (60.6)
None	3 (7.7)	4 (11.4)	29 (17.1)
Minor difficulty with items	7 (17.9)	5 (14.3)	16 (9.4)
Minor difficulty for personal reasons	5 (12.8)	8 (22.9)	14 (8.2)
Major difficulty with items	5 (12.8)	4 (11.4)	8 (4.7)
Major difficulty for personal reasons	4 (10.3)	2 (5.7)	0 (0.0)

Percentages appear in parentheses.

Table 7.11. *Bootstrapped post-hoc Dunnett's T3 comparisons for difficulty question response.*

	Comparison	Mean difference	Standard error of difference	95% Confidence Interval[a]
Coded difficulty response	ASD vs. OCD	-0.17	0.411	(-0.81,0.77)
	ASD vs. Control	1.10*	0.31	(0.51,1.72)
	OCD vs. Control	1.12*	0.30	(0.54,1.69)

7.6. Discussion

Overview

Overall, the evidence presented indicates the relevance of a Compulsive and Repetitive Trait (CaRT) framework to compare disorders such as OCD and autism. This framework appears to be complex, although it is not clear how much this is affected by the methodological limitations within this study (see section 7.6.1).

An overlapping symptomology between Repetitive Behaviours and OCD traits was indicated throughout the analyses. Firstly, Repetitive Behaviours did not significantly differ between the autism or OCD groups in relation to number or frequency of these traits, suggesting that Repetitive Behaviours may reflect general pathology within a CaRT framework. However, the mood of Repetitive Behaviours seemed to distinguish the OCD and autism groups; these traits were associated with a significantly higher positive mood in the autism group. Secondly, OCD traits were relevant to the autism group, although all measures of OCD traits were significantly highest in the OCD group.

However, the ego-syntonic (consistent with the individual's sense of self, wanted and related to pleasure) assumptions of Repetitive Behaviours were challenged by the evidence. All groups reported negative mood associated with some Repetitive Behaviours, an effect which was much more pronounced in the OCD group and negligible in the neurotypical control group. Similarly, whilst OCD traits are clinically defined by their ego-dystonic properties (i.e. being unwanted and related to distress), there were OCD traits associated with positive mood in all groups of participants. This indicates a complexity to the CaRT framework as a function of mood.

Finally, adding to this complexity were the strong correlations identified between the number of Repetitive Behaviours and the number of OCD traits, despite a lack of correlations between any mood scores between these two measures. Whilst untested mediating variables (e.g. depression, anxiety, frustration) are likely to be important, it also indicates Repetitive Behaviours and OCD traits may be somewhat interconnected. However, all the results can only be very tentatively stated due to the various limitations with the methodology, as described below.

Overall discussion

For hypothesis one, it was expected Repetitive Behaviours would be significantly higher in the autism group (increased in number and frequency). This was expected as the origins of Repetitive Behaviours are from autism and intellectual disability research (e.g. Bodfish et al., 2000; Leekam et al., 2007; Moss et al., 2009) and have been specifically related to autistic symptoms (e.g. Bodfish et al., 2000). However, contrary to this expectation, the analyses indicated there was no significant difference between the autism sample and the OCD sample with respect to number or frequency of Repetitive Behaviour, either past or current.

Notably, there is a key difference in the distribution of the means of Repetitive Behaviours between these two groups. The normal distribution around the midpoint for the autism group was not observed for the OCD group. Instead, the distribution of mean Repetitive Behaviours for the OCD group was bimodal, with peaks scores mostly in the low frequency of reported Repetitive Behaviours, but also a slight peak in high frequency of these traits (see Figure 7.5). Although it cannot be ascertained from the available evidence, if this is due to undiagnosed autism in the OCD group (i.e. of those

participants reporting higher frequency of Repetitive Behaviours), it possible the data is masking the fact that Repetitive Behaviours in OCD may be significantly less frequent in type, compared to in autism (which would be in line with the hypothesis).

Regardless, there was a significant difference between the two disorders and the neurotypical control group on frequency and number of Repetitive Traits. Whilst repetitive traits are a common part of everyday life (Keren et al., 2010), the finding indicates Repetitive Behaviours may reflect general pathology and is supportive of the validity in a Compulsive and Repetitive Trait (CaRT) framework to compare OCD and autism. This is somewhat consistent with evidence in Chapter 4: whereby repetitive behaviours have been reported to be common across other disorders (Boyer& Liénard, 2006; Evans et al., 1997; Happé, 1994). The phenomenology of CaRTs across the groups appears to be varied, suggesting there may be considerable overlap in symptomology, as the standard deviation and error was reasonably large across all the groups (see Table 7.2). Furthermore, the results may be compounded by undiagnosed comorbidity, potentially indicated by a substantial number of outliers in the control group (see Figure 7.3). Although with the prevalence rates of autism and OCD at just over 1% in the general population, it could potentially be expected for as many as three or four of the (*n* = 170) control group to have undiagnosed disorders. Overall, for hypothesis 1, the evidence indicates we are not able to reject the null hypothesis: Repetitive Behaviours appear to be equally high in autism and OCD.

As previously described, mood analyses may be fundamental to identifying ego-dystonic and ego-syntonic repetitive traits, a core feature of the CaRT framework. For the second hypothesis it was proposed Repetitive Behaviours would be associated

with positive mood. This was expected as Repetitive Behaviours in autism have been assumed to be more ego-syntonic (not unwanted: typically for enjoyment, pleasure or arousal), perhaps more of a historic assumption (e.g. Baron-Cohen, 1989). The analyses supported this, with all groups reporting overall positive mood for Repetitive Behaviours. Furthermore, as expected, the autism group demonstrated significantly higher overall positive mood associated with these Repetitive Behaviours compared to both the OCD and the control group. Most of the Repetitive Behaviours reported by participants with autism were associated with a positive mood. This is consistent with previous research. Ruta et al. (2010), for example, reported individual with autism to not demonstrate distress in response to repetitive traits. Similarly, in the most relevant study indicated in the systematic literature review section (Chapter 5), Rice (2009) identified significantly higher pleasure-seeking and soothing qualities related to Repetitive Behaviours in autistic versus OCD participants.

Overall mood scores for Repetitive Behaviour scores reported by the OCD group were both non-significantly different from the control group and much closer to neutral mood. This again was largely in line with previous literature (e.g. Baron-Cohen, 1989; Rice, 2009). These results further an understanding of Repetitive Behaviours in adults with autism, indicating Repetitive Behaviours in autism are not distinct in frequency, but are specifically associated with positive mood in the disorder. This is consistent with research and indicates certain repetitive traits may be products of the unique functions of the autistic mind, such as Insistence on Sameness and restricted interests (e.g. Smith et al., 2009; Lam & Aman, 2007; Lam et al., 2008). However, the relatively high number of outliers (4 out of 35) in the OCD group is potentially problematic, as is the large measure of variance (see Figure 7.7). Whilst the latter point may reflect the

highly heterogeneous nature of OCD (which itself may problematic to establishing a comprehensive framework), the outliers may indicate the OCD sample as not being truly representative of the wider OCD population.

Furthermore, it must be emphasised these findings do not demonstrate Repetitive Behaviours in autism are definitively ego-syntonic. All groups reported negative mood associated with some Repetitive Behaviours. Whilst this was negligible in the neurotypical control group, comparatively there were a significant number of Repetitive Behaviours associated with negative mood for the OCD (mean score of 1.37 out of 18 items) and, to a lesser extent, the autism groups (mean score of 0.38 out of 18 items). Additionally, mood may be positive during a repetitive act, but driven by a prior negative mood, like in OCD. To infer a possible alternative cause of the Repetitive Behaviour, participants were also asked how they would feel if they were unable to complete the repetitive trait (alternative question). Three significant things occurred. Firstly, overall negative mood was reported across all the groups. Secondly, mood scores were now not significantly different between the autism and the OCD groups. Thirdly, mood for the control group was less extreme than for the clinical groups. Although common variables such as frustration may have affected all participants (anxiety has been demonstrated to increase repetitive traits in neurotypical individuals: Lang et al. 2015), it is notable that participants with a disorder were most affected – although these traits are more relevant to both clinical groups. Furthermore, it is interesting the participants with OCD were most affected by not being able to complete the repetitive trait. As the need for consistency and sameness appears to be significant to autism (e.g. Bodfish et al., 2000; Baron-Cohen, 2008; Factor et al., 2016; Lidstone et, 2014), this group may be expected to be specifically

affected. However, this considerable effect in OCD needs further investigation. It is possible the suggested shared symptomology between autism and OCD includes a need complete these repetitive traits, rather than just for a desire to do so. If this is confirmed, – and the cognitive pathways involved will be complex – then it is possible even Repetitive Behaviours may be more ego-dystonic (inconsistent with the sense of self; unwanted) than they have previously been assumed. This perspective is in line with more recent research (see review by Barber, 2015).

With regards to the third and fourth hypotheses, in line with evidence of links between OCD and autism (e.g. Barber, 2015; Deramus, 2009; Hutton et al., 2008; Russell et al., 2005), it was proposed OCD traits would be significantly higher in the autism group, compared to the neurotypical control group. The data supported this. Whilst all measures of OCD traits (including number, frequency and negative mood) were significantly higher in the OCD group, all measures of OCD were significantly higher in the autism sample compared to the neurotypical controls. This appears to demonstrate clinical significance of OCD in adults with autism and was in line with increasing evidence of links between the disorders (e.g. Anholt et al., 2010; Bejerot et al., 2001; Hutton et al., 2008; Hollander et al., 2003; Ivarsson & Melin, 2008; Russell et al., 2005). The frequency of the distribution of OCD traits were similarly in line with previous reports: lower frequency of OCD traits in the autism group (see Figure 7.5) reflects findings indicating the lower severity of OCD traits in this group (e.g. Russell et al., 2005). Like the Repetitive Behaviour scores, the OCD group again demonstrated high variability in reported OCD traits (see error bars in Figure 7.7). Whilst this again may reflect the heterogenous nature of OCD, it may also be a consequence of the relatively low sample size (as was determined originally by the power calculation – see

limitations section below). The decision to omit the severity subscales (to reduce the overall number of items in an already large questionnaire) from the Y-BOCS-II-SR meant a clinical assessment of OCD could not be obtained. Nevertheless, the findings seem to add to increasing evidence of shared symptomology between autism and OCD and is consistent with evidence of similar patterns of obsessions and compulsions in OCD and autism (e.g. Russell et al., 2005). This finding potentially indicating the validity in a Compulsive and Repetitive Trait spectrum, with both overlapping symptomology as well as disorder-specific differences.

Notably, there were OCD traits associated with positive mood in all groups of participants, significantly more in the two clinical groups. As OCD traits are diagnostically ego-dystonic (inconsistent with the self, therefore unwanted and related to negative mood), OCD traits associated with positive mood should, clinically defined, not exist. Unless there can be demonstrated to be additional cognitive processes occurring, it is tentatively proposed these OCD traits associated with positive mood are indicative of ego-syntonic Repetitive Behaviours, not OCD traits: certain individual traits may be wrongly defined. This analysis would be missed using most variations of the Y-BOCS (Goodman, 1989), where OCD traits are generally measured on global scale and clinicians are required to assess overall OCD based on severity (e.g. distress, frequency, duration etc.) of three of the most salient traits. However, a greater clarity is gained from assessing severity for each item, in line with approaches by Pertusa et al., (2012) and Wu et al. (2007).

For hypothesis five, higher-order Repetitive Behaviours were expected to be more related to positive mood than lower-order Repetitive Behaviours, specifically in the

autism group. This hypothesis was largely rejected. Previous evidence had suggested higher-order Repetitive Behaviours are autism-typical (e.g. Baron-Cohen, 2006; Carcani-Rathwell et al., 2006), whereas motor and sensory (lower-level) Repetitive Behaviours are likely a consequence of lower developmental age and/or level of functioning (Carcani-Rathwell et al., 2006; Militerni et al., 2002; Richler et al., 2010), in addition to the proposition of Repetitive Behaviours being fundamentally ego-syntonic in autism (Baron-Cohen, 1989; Rice, 2009). However, differences in mood were not demonstrated between higher- and lower-order Repetitive Behaviours in this investigation. Notably, an autism-specific effect was not found: there were no significant differences in mood between higher- and lower-order Repetitive Behaviours for any of the groups. Mood was positive across all groups for both higher- and lower-order Repetitive Behaviours, with no significant differences between either clinical group. The only significant difference in mood was for alternative responses; when participants reported how they felt if they were unable to undertake the Repetitive Behaviours, the autism and OCD groups both reported significantly lower mood than the control group. Again, both clinical groups appear to be similarly affected by an inability to complete repetitive traits, indicating complex and possibly shared mediating variables (e.g. anxiety). Evidence has suggested, for example, Insistence on Sameness (a higher-order Repetitive Behaviour) is related to anxiety in children with autism (Factor et al., 2016; Lidstone et, 2014), but specifically only in individuals with high-anxiety (Rodgers et al., 2012).

For the sixth hypothesis, OCD traits were anticipated to be more prevalent in social situations, specifically for the autism group. It is generally accepted social situations typically cause stress in autism (e.g. Baron-Cohen, 2008; Deramus, 2009), with reports

of high social anxiety in the disorder (e.g. Liew et al., 2015; Maddox & White, 2015; Pickard et al., 2017), and this finding may add to evidence of ego-dystonic repetitive traits in the disorder i.e. in response to a stressful situation. However, no significant differences were identified between the autism and OCD group for OCD traits occurring in either social situations (i.e. around people), or when alone. It is unclear why this surprising result was demonstrated. Possibly repetitive traits are less driven by social anxiety in adults with autism as they learn other coping strategies. This would be in line with the indication in the DSM-5 (APA, 2013) which regards repetitive traits as being less relevant in older age in autism. Whilst this may reflect general complexity of repetitive traits in autism, it could be interpreted as further support of a shared symptomology between autism and OCD.

The analyses did not support the final hypothesis, where there was expected to be no correlation between OCD traits and Repetitive Behaviours. This would have been indicative of a traditional view of Repetitive Behaviours in autism being ego-syntonic in origins (Baron-Cohen et al., 1989) and in line with some empirical evidence (e.g. Cadman et al., 2015; Rice, 2009). For each group, analyses demonstrated strong correlations between the number of Repetitive Behaviours and the number of OCD traits. Interestingly, no correlations were identified for any of the groups on mood scores between the two measures. This is perhaps indicative of the effect of any number of unstudied mediating variables (e.g. depression, anxiety, frustration). Again, it can be also interpreted as further evidence of the validity in combining these measures within one framework, suggesting Repetitive Behaviours and OCD traits are not on opposite ends of a continuum but are, perhaps, relatively interconnected (Fineberg et al., 2010).

Finally, with regards to the difficulty in completing the questionnaire, there were significant numbers of difficulties reported by participants with answering the questions. Notably, 21 (54.4%) of the OCD group, and 19 (53.8%) of the autism group reported some difficulties with 6 (17.1%) and 9 (23.1%) participants in the OCD and autism groups, respectively, reporting major difficulties in answering the questions. These reports indicate the questionnaire may be a completely valid measure of Compulsive and Repetitive Traits across in these groups (see limitations section 7.6.1). Whilst both clinical groups reported significantly more problems answering the questions, there was no significant difference between the autism and OCD groups. As detailed in Appendix 10 (a full set of all these free responses) typical answers for minor difficulties included grammatical mistakes in one or two of the items or responses (minor difficulties with items) and feelings of true answers as being slightly different from the narrow choices available (minor difficulties for personal reasons). Major difficulties were more varied, but typical responses included frequency being hard to assign for some items (major difficulty with items) and difficulty in reporting emotional responses (major difficulty for personal reasons), particularly in the autism group. Notably, there were a high proportion (12.2%) of clinical participants reporting major issues with answering questions.

Summary

Overall, these findings add to evidence of both ego-dystonic and ego-syntonic repetitive traits in autism (e.g. Rice, 2009; Saddington, 2013), which indicates the validity and utility of interpreting disorders such as autism and OCD within a Compulsive and Repetitive Trait spectrum. Rutter and Caron (1991) suggest such a dimensional approach to be more valid than the simplistic labelling of comorbidity, which itself may be a statistical phenomenon, devoid of meaning (see Rutter 1997. Instead, a more comprehensive perspective of the individual mechanisms (traits) would appear necessary to build a "cluster of symptoms approach" to the individual (Bentall, 2004). A range of hypotheses were presented to indirectly assess ego-dystonic and ego-syntonic repetitive traits in adults with autism, OCD and neurotypical controls. Whilst Repetitive Behaviours were comparable in both OCD and autism, the presence of more positive mood was a feature of autism. OCD traits were generally more severe in the OCD group, although may be potentially of clinical significance in autism. Other evidence appeared to suggest CaRTs may contain largely overlapping symptoms between autism and OCD, adding to trends identified in recent empirical research (Anholt et al., 2010; Bejerot et al., 2001; Deramus, 2009; Hollander et al., 2003; Ivarsson & Melin, 2008; Russell et al., 2005). This evidence supports the validity of using a combined Compulsive and Repetitive Trait spectrum as well as the clinical relevance of continuing to understand symptoms at the item-level (particularly with regards to mood). Figures 7.9 and 7.10 illustrate how the present evidence has changed the framework. This was based on the historical view suggesting a relative dichotomy between ego-dystonic (negative-mood related) OCD traits and what was originally viewed as more ego-syntonic Repetitive Behaviours, typical of autism. However, the evidence presented in the present study suggests a much more complex

relationship between ego-dystonic and ego-syntonic repetitive traits in OCD and autism. Repetitive Behaviours were not significantly different between the OCD and autism groups in number, although they were associated with increased positive mood in the autism group. OCD traits, however, were significantly higher in the OCD group, although they were also significantly higher in the autism group compared to the control group, consistent with increasing evidence to suggest comorbidity of OCD in autism. With repetitive traits (both OCD traits and Repetitive Behaviours) being heterogenous across both OCD and autism, the "cluster of symptoms" approach is supported. The main implications for practise is the necessity for the clinician to consider ego-dystonic traits in adults with autism, and to undertake more fine-tuned analysis to understand the impact (and function) of repetitive traits across disorders.

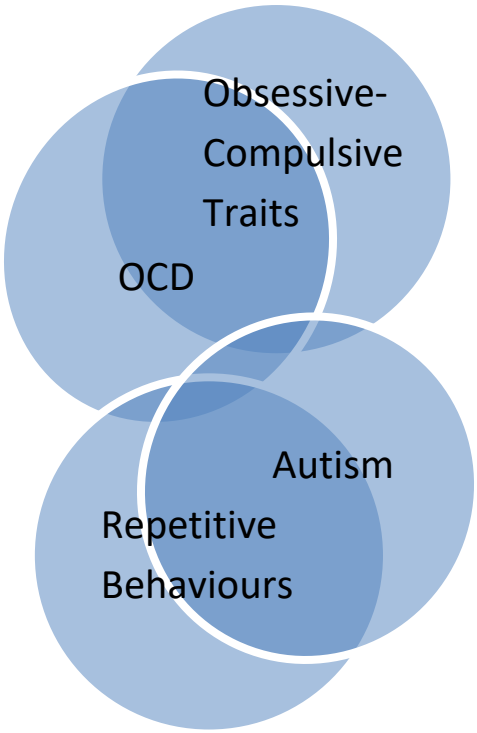

Figure 7.9. Original framework as illustrated in Figure 7.1.

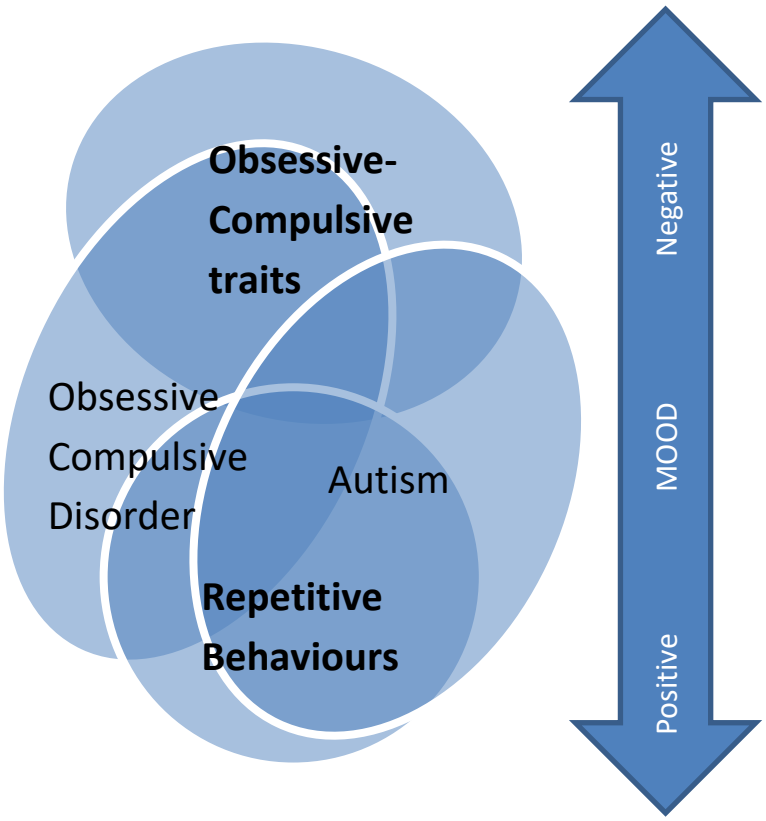

Figure 7.10. New Compulsive and Repetitive Trait framework with continuum of mood.

7.6.1. Research limitations and strengths.

Comparing OCD traits and Repetitive Behaviours within an overall framework (Compulsive and Repetitive Traits) adds to the recent understanding of OCD as a spectrum (DSM-5, APA, 2013) and to the growing knowledge of cross-disorder comparisons. The current research addresses methodological shortfalls in previous investigations by avoiding cross-comorbid OCD and autism issues (e.g. Russell et al., 2005). It also increases empirical data within a limited field of repetitive traits in adults with autism, particularly where no studies have attempted to assess and compare OCD traits and Repetitive Behaviours together in this population. Whilst the Y-BOCS was not originally designed to provide an item-level understanding of OCD traits, this study adds to the recent trend in attempting to understand OCD at the item-level (e.g. Pertusa et al., 2012; Wu et al., 2007). Additionally, a relative strength is the use of

validated measures of repetitive traits; whereas some studies directly assess ego-dystonic and ego-syntonic perceptions of repetitive traits (e.g. Rice, 2009), previous measures used have lacked psychometric validation.

A major limitation is that ego-dystonic and ego-syntonic qualities can only be inferred from this data. It is not possible to determine the exact cause of mood from this evidence without undertaking more complex investigations directly related to ego-syntonic and ego-dystonic perceptions of the functions of these traits (e.g. Rice, 2009). Originally, it was intended for the CaRT questionnaire to assess mood before, during and after each repetitive trait. However, after careful consideration – and reflection on the pilot study - it was decided this would be phenomenologically vague (particularly for thoughts) and potentially highly subjective. It was decided, therefore, to simplify the concept by asking the participant how they feel during the thought/behaviour. However, a major part of the original concept was lost: one may feel neutral when checking the door, but this may be driven by a prior feeling of anxiety.

However, simple mood assessments were chosen for two main reasons. Firstly, as a comparison between Repetitive Behaviours and OCD traits would require 78 items, there was a reluctance to add too many extra questions: minimising fatigue in participants should improve the reliability and validity of data, in addition to improving recruitment rates. Secondly, complex introspection (particularly ego-dystonic) may be inaccessible to some individuals, particularly with autism (Baron-Cohen, et al., 1989). Creating simple clinical tools should, therefore, improve accessibility across a greater proportion of individuals.

There are three main concerns about the samples of participants. Firstly, there were insufficient numbers within the two clinical groups to sufficiently power the statistical analysis (see 7.4.3). Regardless of the lack of relevant studies for the power calculation, any conclusions can only be tentatively made. Secondly, it appears likely the sample was unrepresentative of the wider population. For example, the overall proportion of female participants was very high (70.5%). This is particularly skewed for the autism sample, which would be expected to be about 80% males (Fombonne, 2003). Additionally, control participants were generally older than the clinical groups, whilst there was a lack of ethnic diversity throughout (95% of the entire sample were Caucasian). It appears that administering the study only on the internet increased sampling bias. However, there were advantages to recruiting in this way. Online access and widespread advertisement (even on an international scale) enabled an ecological validity not usually demonstrated across comparable literature, where participants are generally recruited via access to specialised clinics (e.g. Russell et al., 2005). Also, removing the social demands in studies may also improve the validity of the findings, particularly for participants with autism (e.g. Benford, 2008; Ozonoff, 1995). However, the skewed demographic characteristics may have affected any number of the analyses. It has been suggested, for example, that repetitive traits in autism are differently presented in females (Van Wijngaarden-Cremers et al., 2014). Whilst repetitive traits in autism have been demonstrated to be stable across cultures (e.g. Georgiades & Papageorgiou, 2010; Inada et al., 2015), there is some suggestion this may not always be the case (Magana and Smith, 2013).

One final major criticism can be made of the samples of participants. A conscious decision was made to attempt to recruit participants without comorbid OCD and

autism. Exclusion criteria for autistic participants included a previous diagnosis of OCD, and similarly dual-diagnosis of autism was an exclusion criterion for the OCD sample. The rationale behind this was to attempt to understand autism and OCD separately, in their "pure" forms, i.e. in isolation from other disorders. This was an extra safeguard as the format of this self-report study was unable to employ screening procedures or confirmation of diagnoses. However, use of these exclusion criteria may be deeply flawed. Comorbid autism and OCD have been regularly demonstrated to occur (e.g. Pertusa et al., 2012; Roy et al., 2015; Rydén & Bejerot, 2008). It could be argued that this set of participants is perhaps the most useful in terms of understanding the similarities, if not the differences, between autism and OCD with respect to Compulsive and Repetitive Traits. Researchers have warned against a skewed focus towards tiny atypical samples (Rutter & Caron, 1991). Furthermore, individuals in this study may have had comorbid OCD and autism; one of the disorders may have just been undiagnosed at the time of investigation: Wikramanayake et al. (2018) reported 46% of their sample with OCD met the diagnostic threshold for autism, but had not previously received this dual diagnosis. This recruitment method affected the external validity of the study as it is likely caused the sample to be unrepresentative of the wider populations they come from. A recruitment method employing consecutive referrals, and screening for comorbidity, may be much more valid to improve this issue.

The systematic literature review of psychometric properties (Chapter 6) demonstrates the Y-BOCS-II-SR used in this investigation to be a psychometrically robust measures in an adult population. No firm conclusions can be made from the current investigation with regards to OCD traits in the autistic sample until there is evidence of reliability

and validity of the Y-BOCS-II-SR in autistic adults. Furthermore, the OCD assessment in the present investigation combined the method of dropping the subscales from the Y-BOCS-II-SR, which has been validated in adult participants (Mataix-Cols et al., 2004; Sulkowski et al., 2008), along with the self-report formation of the Y-BOCS-II-SR (Baer, 1992), which has been validated in adult participants by Steketee et al. (1996). Similarly, the use of the Repetitive Behaviour Questionnaire-2 (Moss et al., 2009) in current investigation is even more tentative in an adult autistic population. At the time of investigation there were no studies identified which validated the use of Repetitive Behaviour assessment tools in adults, and the decision was made to use the RBQ-2 (Moss et al., 2009) based on the rationale stated in section 7.1.3. Taken together, all the results must be tentatively taken until psychometric evidence is available for the RBQ-2 (Moss et al., 2009) and the Y-BOCS-SR in autistic adults. Similarly, the self-report method used in this study for the Repetitive Behaviour Questionnaire, where the items were changed from third-person (see Appendix 12) to first-person (see Appendix 3) has not been validated in any sample and would require testing of psychometric properties before these results can hold acceptable scientific weight.

Although the Compulsive and Repetitive Trait (CaRT) questionnaire used in this study largely consisted of items from two validated assessments tools, there were novel constructs, specifically related to measure of mood. Recruitment was particularly difficult for the clinical groups (see section 7.4.7), likely due to the lengthy time needed for these participants to complete the items. This resulted in an insufficient number of participants for factor analysis, meaning the CaRT questionnaire cannot be validated from this data. However, at the end of each investigation, participants were asked to record any difficulties they had in completing the study. Adaptations following the

initial pilot (see section 7.2) indicated an improvement in the design of the measure. Of the two clinical groups (*N* = 74), 15 (20.2%) participants reported significant difficulties in completing the questionnaire, which is a high proportion. 9 of these participants (12.2%) reported major issues were the questions themselves, typically relating to the scales not being accurate enough to record their actual experiences (see Appendix 10). The remaining 6 (8.1%) participants with OCD and autism reported major difficulties with answering the questions due to personal reasons, of which 4 (5.4%) reported very significant problems such as "sometimes I didn't understand the question" or "some of my behaviours I am not aware of". Although further research is necessary to test the psychometric measurements of the properties and assess the validity of this measure, these responses indicate a substantial proportion of the clinical participants found it difficult to complete the questionnaire.

Another concern was the length of the CaRT questionnaire. The measure consisted of 76 individual items (see Appendices 2 and 3), each of which contained 1 to 5 levels (see Appendix 1). Every attempt was taken to minimise this being an issue. Participants were given the opportunity to save after each question and finish later, in addition to a percentage complete bar being displayed on each page. The data appears to show that participants remained motivated to answer throughout, with no obvious pattern of random answering. However, it is possible the time and effort required to complete the study may have been a barrier some individuals. Accordingly, the response rate was extremely low, which again may have led to the unrepresentative participants' characteristics. This response rate was a maximum of 4.89% and 3.69% in the autism and OCD samples, respectively. Furthermore, these figures are based on the number of people who accessed the first page of the Bristol Survey and doesn't include those

individuals who received the advert but chose not to access the study at all. Typical response rates are unclear for questionnaire methods in autistic samples (e.g. Cadman et al., 2015; Scahill et al., 2006) and, anecdotally, autism services are regularly approached to complete studies (which can reduce the response rate in the group). However, the huge discrepancy between the response rates in other web-based questionnaire studies (e.g. Horevoorts et al., 2015; Burgess, Nicholas & Gulliford, 2012) is likely to be an issue, affecting the likely representativeness of the samples to their wider populations.

Finally, the self-reporting procedure may be inaccurate, most notably as, participants' answers were taken on face value: there was no clinical confirmation of diagnoses, for example. Furthermore, whilst there may not be concerns with the validity compared to informant-reports (e.g. Federici et al., 2010), the study did not allow for any assessing or even screening of comorbid disorders. Thsis is may be significant, particularly as OCD and autism are reported to be largely overlapping disorders (Anholt et al., 2010; Bejerot et al., 2001; Deramus, 2009; Hollander et al., 2003; Hutton et al., 2008; Ivarsson & Melin, 2008; Lehnhardt, 2013; Russell et al., 2005). Additionally, there were also many confounding variables which were not investigated, such as potential effects of medication, anxiety and depression, in addition to subgroup membership (e.g. early- versus late-onset, or categorical type of OCD; High Functioning Autism versus Asperger syndrome in autism). This is notable as both autism and OCD are highly heterogeneous disorders and it is not certain whether the samples are truly representative of either of the populations.

Chapter 8. Pilot Investigation of Free Will and Compulsive and Repetitive Trait Correlates

Background

Whilst links between autism and OCD have been reported across a variety of investigations (e.g. Anholt et al., 2010; Bejerot et al., 2001; Hollander et al., 2003; Lehnhardt, 2013; Russell et al., 2005), the relationship between these two disorders is not clear. Evidence of both ego-dystonic and ego-syntonic repetitive traits in autism (e.g. Buckby, 1999; Rice, 2009; Saddington, 2013) suggests repetitive traits in the disorder may be part of a complex pathway; variables involved in this pathway are likely to include sensory processing (Brodsky, 2014), developmental age (Carcani-Rathwell et al., 2006; Militerni et al., 2002) and intelligence (Richler et al., 2010). Chapters 2 to 5 present an argument indicating a validity in comparing OCD and autism within a Compulsive and Repetitive Trait framework, whilst the empirical investigation in Chapter 7, further suggests the validity in such an approach. However, the lack of clarity (perhaps thrown up by limitations – see 7.6.1), may be compounded by the complex pathway of repetitive traits. Deeper understanding of the individual mediating and moderating variables is likely necessary: the study of free will and its potential links to compulsive behaviour (as described below) may provide this to some extent.

Compulsive behaviours are typically measured within the diagnostic boundaries of OCD: these repetitive traits are performed to reduce the anxiety caused by obsessions, or to rigidly applied rules (APA, 2013). However, there is another view of compulsions as irresistible urges to behave in a certain way – behaviour which some claim may be

"carried out against the will" (Heather, 2017). Free will, therefore, maybe another missing variable in the Compulsive and Repetitive Traits pathway.

This section on free will and Compulsive Repetitive Traits is presented as a separate chapter due to the exploratory nature of the investigation. A rationale for the inclusion of measures of free will belief is presented, following an outline of the theoretical principles in relation to the clinical framework presented (i.e. Compulsive and Repetitive Traits).

8.1. Introduction

8.1.1. Do humans have Free Will: arguments for and against.

The concept and nature of free will is one of the longest standing philosophical debates. Determinism claims it is an illusion. As every thought or behaviour is preceded by a previous event, determinism contends we can never truly control our actions, as is only ever one possible outcome caused by the summation of all previous events. However, compatibilist approaches reject the arguments on which determinism are based (e.g. Sartorio, 2016). Ryle (1949) convincingly argues the premises on which this determinist argument is erroneous; framed incorrectly. Theorists across various fields argue free will does exist but is often misperceived because of the sheer complexity of our cognitive machinery (e.g. Dennett, 1991; Levy, 2003). This philosophical debate is subject to endless claims and counter-claims, all of which are beyond the scope of this current investigation.

We can, however, turn to empiricism. Swinburne (2013) argues the weight of our scientific knowledge strongly opposes a determinist view to free will; just as no two

brains have been shown to be in the same state, neuroscience can predict, but not determine our decisions. The vast body of evidence in psychology would lead us to believe free will exists amongst a myriad of concepts including: choice; agency; volition; intention; cognitive control; and autonomy.

The understanding of whether we have free will is as strenuously disputed in psychology as it has been in philosophy. The dispute starts at the very definition of what free will is. A common experimental psychological paradigm is the study of voluntary actions. Libet (1999), for example, ignited this topic through the investigation of awareness of actions, whereby the concept of Readiness Potential was studied. Readiness Potential relates to the measure of brain activity, via which Libet (1999) reported participants appear to demonstrate electroencephalography (EEG) signals milliseconds before their conscious awareness of acting. The interpretation by Libet (1999) was that volitional actions are initiated by the unconscious mind. However, Armstrong, Sale and Cunnington (2018) reviewed the range of experiments investigating the Readiness Potential, a concept which has caused argument over the lines between volition and free will (as conscious awareness). The reviewers found the complexity of the brain precludes our ability to adequately test it within the frame of "free will", and the study of voluntary action lacks such clarity. Armstrong et al. (2018) conclude the current evidence does not determine any causal links between neural activity and intentional behaviour.

Evidence indicating brain function as a highly deterministic framework (Leisman, Machado, Melillo, & Mualem, 2012), would appear to strongly oppose the possibility of free will. However, free will is saved through the indication conscious choice may

play a small part in the production of movement. In support of this, Hallet (2007) neurologically demonstrated the seemingly illusory nature of conscious awareness. Movement appears to be initiated in the frontal lobe and later a sense of volition registered in regions including the parietal lobe and insular cortex. In fact, Libet's (1999) emphasises even evidence of the Readiness Potential does not exclude free will, as later voluntary awareness seems to allow a "veto" of movement. Clearly, evidence from brain activity can be misleading and a strict deterministic view of such a complex system has been indicated to be erroneous: in this incredibly developed system free will has the potential to exist.

Whilst free will may have many meanings, Swinburne (2013) suggests two are specifically relevant for use in psychology: agency (causing something to happen); and moral responsibility. The study of moral responsibility (in particular, the judgement of others) may lack relevance to the clinical investigation of Compulsive and Repetitive Traits. However, the personal construct of agency may be significant to this study. Agency may appear to be retrospectively understood, whereby an individual processes their sense of feelings of control in relation to later external events. This may be problematic to the view of volition and free will, much in the same way the concept of Readiness Potential (i.e. voluntary awareness after an act), may somewhat challenge an understanding of volition and free will. However, Chambon, Sidarus and Haggard (2014) reflect on evidence from neuroimaging and discover the likelihood agency may be linked prospectively. Unsurprisingly, agency appears to be affected by other psychological mechanisms. For example, agency has been reported to be constrained by attention (see review by Hon, 2017). There is a whole psychological network,

therefore, potentially linked to the understanding of free will; much of this research is discovered within the study of neurological disorder (see section 8.1.2).

Volition may also be a significant process in the psychology of free will. Volition has been defined to be goal-oriented, internally driven, cognitive processes leading to actions (Haggard, 2019). As volition requires an ability to consider future goals, this concept may appear to be a challenge for neuroscience. However, Fried, Haggard, He and Schurger (2017) explain how volition, as a concept of (internally driven) goal-directedness, aligns philosophical enquiry into psychological investigation. The researchers indicate goal-directedness is linked to neuroanatomical functioning, and also to a sense of agency through the motivational and subjective appraisal. Haggard (2019) goes further and argues how neurocognitive mechanisms related to volition may be central to issues such as subjectivity and voluntary action. It would appear, therefore, the philosophical enquiries of free will may be aligned to volition and agency.

8.1.2. Psychopathology of free will.

A definition of free will recommended by Miles Cox, Klinger and Fadardi (2017) is the "capacity for free action [which means] that the person could do different things in the same situation" (p.94). Using this definition, the researchers argue even individual suffering from addition (typically viewed as a condition lacking will) have free will, as they process goal-directed behaviours. Even severely addicted individuals are understood to have capacity to make a conscious decision-making based choice when the subjective costs are judged to outweigh value of continuing (Miles Cox et al., 2017).

From a cognitive neurocognitive perspective, Blair (2007) suggests a compatibilist approach to understanding the possibility of free will is consistent with the physiological and theoretical understanding of neurological disorder. Similar to the view by Miles Cox et al. (2017) in addictions and free will, Blair (2007) claims even in psychopathy, individuals still have a choice of actions and goals available to them, and the ability to process information according to these parameters. As Miles Cox et al. (2017) emphasise, there is a complexity of factors (genetic, neurobiological, social, personality and psychological), which all create a context for various goal-oriented choices, no matter how limited they may be. Whilst free will may be constrained, even in such restricted circumstances there still appears room for free will to exist.

Haggard's (2017) review of sense of agency considers the pathology of agency. Sense of agency is the study of the conscious awareness of voluntary actions. Such studies are typically social in nature i.e. they measure agency in relation to the self or another. Haggard (2017) reports a general cognitive bias in sense of agency: a self-serving bias where an individual will overestimate their sense of agency when an outcome is positive. Disorders of agency, Haggard (2017) reports, tend to lie on either end of a dichotomous scale. Hyperagentic conditions are those whereby an individual has excessive sense of one's own sense of causation and control. In hypoagenetic disorders an individual has a reduced experience of causation and control, such as in psychosis and schizophrenia, where an individual may overstate action as being caused by external agents.

Kranick and Hallett (2013) also argue for a spectrum of pathology of agency. Disorders such as Early Huntington's disease and anosognosia are considered to be hyperagentic,

with patients claiming agency for movements. On the other end of the spectrum Kranick and Hallett (2013) report Tourette's syndrome as being hypoagentic. The role of agency in Tourette's is interesting, as it implies an opposing role for will (or volition), with the researchers claiming these patients acknowledge responsibility for the action (sense of agency), but they are in response to an urge; therefore lack agency.

8.1.2.1. Free Will and Obsessive Compulsive Disorder.

Glannon (2012) considers how, philosophically, OCD patients can be impaired in their ability to execute free will; they are said to fail to meet the three conditions of free will. Firstly, their desires are misaligned and, therefore, do not meet the conditions for responsibility for mental states. Secondly, an individual with OCD is said to not satisfy conditions for receptivity and reactivity, being they are impaired by their absolute need to complete an action. Thirdly, the necessity for identification of actions is not met, since their ego-dystonic feelings towards their actions lack such identity (and is therefore not held to be a genuine source of the actions related to the intrusive thoughts). However, saving free will in OCD (and all disorders) is the idea free will is not an all-or-nothing (absolute) condition, and itself is believed to lie along a spectrum (Glannon, 2012).

Pathologically, cognitive components such as control and agency may be important. Pickard (2015) claims obsessions are one of several "disorders of agency", in which choice and free will are not dichotomies in opposition to a compulsion to act. The relationship between OCD and control is disputed. For example, Meynen (2012)

describes OCD as a disorder of excessive control, or a lack of control. Regardless, the importance of control in OCD is consistent.

The “not just right” experiences reported in OCD checking subgroups, has been suggested to potentially be related to an altered sense of agency. Giuliani et al. (2017) empirically investigated this hypothesis through a study of gaze agency (and associated causal attributions) in OCD patients with checking compulsions. The researchers found poorer performance in OCD participants compared to neurotypical controls, which they reported to be indicative of a difficulty with causal attribution in OCD. Whilst consistently dysfunctional in OCD, the potential role of agency was reported to be multi-faceted, with over-attribution to external causes in cognitive tasks, and an increased tendency to inwardly ascribe agency in perceptual tasks. This view is supported by a review on sense of agency in OCD by Szalai (2019), who claims the “not just right” feeling in OCD is central to experience of agency in the disorder, which appears to originate from action monitoring dysfunction in the disorder. Accordingly, Szalai (2019) distinguishes between sense of agency in OCD and schizophrenia, with OCD being specifically related to goal-orientated outcomes.

An investigation by van Oudheusden et al. (2018) has highly relevant implications to the comparison between free will and Compulsive and Repetitive Traits in OCD. In this investigation, the researchers performed factor analysis on perceptions of free will in a large number (*n* = 419) of adults with OCD. The researchers identified three factors, consisting of: alternative possibilities; intentionality; and ownership. Scores on the alternative possibilities factor indicated patients with OCD “experience very little freedom to pursue a different course of action when faced with their symptoms” (van

Oudheusden et al., 2018; p. 6). Lack of intent was found for obsessional content, whereas compulsions were reported to lie within a domain of goal-oriented decision-making. The final factor has potentially huge implications for at Compulsive and Repetitive Trait frameworks. The researchers reported self-ownership as being central to OCD. Rather than being ego-dystonic, symptoms are considered to become ego-syntonic, whereby they become consumed within self-identity of the patients. This, van Oudheusden et al. (2018) report, may decrease free will as it makes it difficult for them to distance themselves from their symptoms (despite desiring to). This claim in particular has huge implications in the proposed Compulsive and Repetitive Trait framework, with an even more complex interaction between ego-dystonic and ego-syntonic principles.

8.1.2.2. Free Will and Autism.

There appears to be no studies investigating volition in autism (with only one case study investigation in infants with autism). There is, however, some evidence of agency in the disorder.

Some evidence indicates sense of agency may be unimpaired in autism. David et al. (2008), for example, reported a dissociation between agency and mentalizing in the disorder. Although imitation is held to be central to agency, the researchers reported intact sense of agency in autism, despite evidence of deficits in relation to mentalising tasks. However, research is not unequivocal on intact sense of agency in autism. For example, if emotional self-awareness is considered as a function of agency, then deficits can be found in autism (Stout, 2019). Additionally, there may be issues with measuring sense of agency. Carruthers (2010), for example, emphasised how it is

possible an individual is able to indicate the source of agency without understanding the relationship between intentions and actions.

A review by Zalla and Sperduti (2015) on studies of sense of agency in autism indicates why evidence may be mixed. The reviewers argued sense of internal agency is a multi-component model, consisting of prospective mechanisms (proprioceptive and sensorimotor processes which occur before a self-directed event) and retrospective mechanisms (autobiographical cognitive processing of these events). Zalla and Sperduti (2015) claim previous evidence indicates a specific impairment related to the prospective mechanisms related to sense of agency in autism, but intact retrospective mechanisms.

Researchers seeking an experimental paradigm have investigated agency defined by the awareness of voluntary action, measuring stimulation in response to simple tests of behaviour (e.g. Haggard et al., 2002; Libet et al., 1999). The only investigation into free will in autism follows this methodology. In this study, Glazebrook, Elliot and Lyons (2008) required participants to record when they heard a tone, which either occurred when the participant pressed a button or when the two stimuli were temporally distinct. The researchers reported adult autistic participants demonstrated comparable performance to age- and gender-matched neurotypical controls. Regardless of the methodological issues (there were small numbers of participants in each of the conditions), the validity of the results is perhaps disputable. It is highly disputed whether these experimental methods are valid measures of free will at all, just as many researchers have argued likening awareness to act with freedom of will is too big of a leap from the available evidence (e.g. Levy, 2005; Ostrowick, 2007; Zhu, 2003).

8.1.3. Free Will beliefs.

Another line of enquiry related to free will is the measure of beliefs in free will and their impact on other thoughts and behaviours (e.g. Vohs & Schooler, 2008). Feldman's (2017) review demonstrates free will to be a unique aspect of agency, relatively independent from other constructs such as choice, self-control and autonomy. Free will beliefs appear to be a good predictor of behaviour, at least in neurotypical adults (Alquist et al., 2013; Baumeister & Brewer, 2012; Rigoni, Kühn, Gaudino, Sartori, Brass, 2012; Vohs & Schooler, 2008). Whilst conscious and subconscious thoughts both affect behaviour (Baumeister, Masicampo, & Vohs, 2010), free will beliefs appear to mediate this relationship. Many behaviours have been demonstrated to be mediated by free will beliefs. A review by Baumeister and Brewer (2012), for example, indicates: high belief in free will may be linked to pro-social (or more aptly, pro-cultural) behaviours, with more punitive attitudes towards those who break societal rules; and low belief in free will may be related to conforming behaviours as well as selfish attitudes and antisocial behaviours.

However, these beliefs appear to be relatively unstable, susceptible to manipulation under experimental conditions. Lowering beliefs in free will can change moral attitudes. Shariff et al. (2014), for example, reported a negative correlation between free will and attitudes towards retributive punishment of criminal offenders. Other studies have found a similar relationship with moral behaviours. Individuals who report greater beliefs in free will seem to be less likely to conform to the group, as inducing disbelief in free will appears to increase conformity (Alquist et al., 2013). Similarly, Vohs and Schooler (2008) primed disbelief in free will, which resulted in increased cheating, both actively and passively. Rigoni et al. (2012) also demonstrated

an increase in impulsive and antisocial behaviours when participants were encouraged to disbelieve free will. Such evidence, it has been suggested, indicates belief in free will may be positively associated with self-control (Rigoni et al., 2012), although it is claimed that these two constructs may be relatively independent (Baumeister and Brewer, 2012).

Further indication of belief in free will being open to manipulation, Feltz and Cova (2014) identified emotions as a moderating variable. In a meta-analysis, Feltz and Cova (2014) found a small effect size across 30 studies which investigated free will belief and emotional valence. There is a positive association between emotions and belief in free will in relation to moral responsibility. How emotions a moderator between personal responsibility and free will beliefs is not clear, although such a line of investigation may be pertinent to a better understanding of Compulsive and Repetitive Traits.

8.1.4. Relationship between free will and free will beliefs.

The relationship between free will and belief in free will is not clear. They may be, in fact, phenomenologically distinct with free will being the capacity for free action/thought, whilst beliefs in free will being the intellectual understanding about the nature of free action.

8.1.5. Summary

The presented evidence indicates there is room for free will, whether viewed from the perspective of volition, agency or even pathology. Volition, as a mechanism responsible for goal-oriented behaviours, may be significant to the ego-syntonic measures of repetitive traits (van Oudheusden et al., 2018). The evidence by van

Oudheusden et al. (2018) has particularly interesting implications for the Compulsive and Repetitive Trait framework. Rather than simply assuming ego-dystonic properties of OCD, it has been indicated ego-syntonic factors are embedded within the disorder, as patients with OCD become self-defined by their symptoms. This may provide a further context whereby autism and OCD can be compared within a shared symptomology model of Compulsive and Repetitive Traits, as properties of mood/affect may be more relevant properties to distinguish between repetitive traits.

Evidence also strongly suggests belief in free will influences behaviour. However, there appear to be no investigations of free will beliefs in of individuals with OCD or autism. Therefore, it is not known whether beliefs in free will either differs in these disorders, or affects clinical symptoms. This line of enquiry may be of clinical utility in autism and OCD as both have issues of control, a significant principle of free will. In autism, individuals appear to seek control in response to the confusing social world (Baron-Cohen, 2008), typically demonstrated through the need for sameness and resistance to change. In OCD, control is central (Meynen, 2012); individuals are compelled to undertake unwanted actions in response to intrusive thoughts (DSM-5, APA, 2013), with high degrees of interference of and inflexibility to symptoms (Goodman, 1989a). Free will also has implications on therapy. For example, free will, as volition, appears to be crucial in cognitive behavioural therapy, where increased effort from the patient is much more likely to improve outcomes (O'Neill & Schwartz, 2004). If free will is related to clinical symptoms, then understanding free will beliefs may offer insight into symptom presentation and modifying free will beliefs could be used as an effective therapy. It appears pertinent, therefore, to investigate whether self-reported free will can predict clinically relevant behaviour, such as an individual's pattern of Compulsive

and Repetitive Traits (CaRTs). This is tested empirically in the next section, which presents a pilot investigation to compare free will beliefs and CaRTs in adults with autism, adults with OCD and neurotypical peers.

A proposed illustration of the possible relationship between free will beliefs and CARTs is presented in Figure 8.1. Here, autism and OCD have been replaced (from Figure 7.1) with the free will categories of Scientific Determinism and Personal Control, respectively. This is based on the claimed relevance of the physical world in autism (e.g. Baron-Cohen, 2008) and control in OCD (Meynen, 2012; Pickard, 2015).

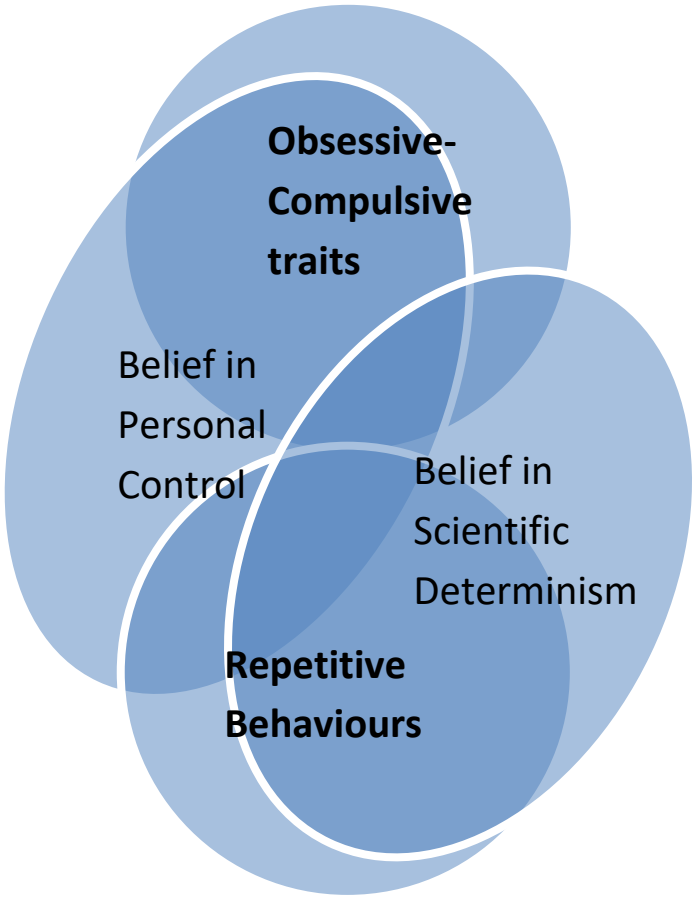

Figure 8.1. The conceptual framework demonstrating the potential relationship between Compulsive and Repetitive Traits and Free Will beliefs (Personal Control and Scientific Determinism).

8.2. Aims and Hypotheses

Following evidence by van Oudheusden et al. (2018) in particular, the overall aim was to test the association between mood, free will and Compulsive and Repetitive Traits. In line with the overall framework presented in Chapters 2 to 7, the aim was to understand shared symptomology issues between OCD and autism. Whilst there is a lack of investigation into free will in autism generally, there is a lack of study of free will beliefs specifically.

Accordingly, this pilot investigation consisted of two main aims: firstly, to understand the effect of disorder (i.e. autism and OCD) on self-reported free will beliefs; and secondly, to assess whether Compulsive and Repetitive Traits are related to self-reported free will beliefs within (and between) any of these groups. In line with the evidence presented in section 8.1, the hypotheses for the free will pilot study are as follows:

H_0 There will be no difference in any free will scores between the three groups.

H_1 The mean Personal Control scores between the groups will differ. The OCD group will demonstrate the lowest Personal Control scores, followed by the autism group, then the control group.

Rationale for hypothesis 1: Philosophical debate has indicated the relevance of control in OCD (e.g. Meynen, 2012; Pickard, 2015); therefore, Personal Control would be expected to be specifically impaired in OCD.

H_0 There will be no difference in Scientific Determinism scores between the three groups.

H_2 The mean Scientific Determinism scores between the groups will differ. The autism group will demonstrate the highest Scientific Determinism scores, followed by the OCD group, then the control group.

Rationale for hypothesis 2: Aspects of the physical world, including science, maths, physics and engineering have been consistently reported to be relevant in autism (e.g. Baron-Cohen, 2008; Wheelright & Baron-Cohen, 1998); therefore, the Scientific Determinism domain would be expected to be significant to autistic participants.

H_0 There will be no relationship between OCD traits and Free Will Personal Control scores.

H_3 OCD scores will be negatively correlated with Free Will Personal Control scores for all groups.

Rationale for hypothesis 3: This null hypothesis is expected to be rejected for the same reasons as the rationale for hypothesis 1.

H_0 There will be no difference between Repetitive Behaviours scores and Free Will Scientific Determinism Scores.

H_4 Repetitive Behaviour scores will be positively correlated with Free Will Scientific Determinism scores for all groups.

Rationale for hypothesis 4: This null hypothesis is expected to be rejected for the same reasons as the rationale for hypothesis 2.

8.3. Method

A pilot study comparing free will beliefs with Compulsive and Repetitive Traits (CaRTs) was undertaken as a final stage of the doctoral thesis investigating the overlap of symptoms between OCD and autism. This pilot study was included within the Compulsive and Repetitive Trait study as reported in chapter 7; all similarities and differences in the methodology is described below.

8.3.1. Measures.

Two questionnaires were used in the pilot study, a subset of questions from an established free will belief measure (the Free Will and Determinism Plus Scale, Paulhus & Carey, 2011) and the Compulsive and Repetitive Trait questionnaire. These two questionnaires – including rationale behind why these particular measures were chosen – are outlined and described below.

The Free Will and Determinism Plus scale (FAD-Plus) is a questionnaire created by Paulhus and Carey (2011) to measure an individual's lay beliefs in free will. Covering a spectrum of free will beliefs, the questionnaire consists of 27 items relating to four relatively independent factors: Personal Control; Scientific Determinism; Fatalistic Determinism; and Unpredictability. The FAD-Plus was chosen for three main reasons. Firstly, it has been designed as a questionnaire, which would easily fit into the general method used in the investigation. Secondly, the questions were designed to be simple enough for the lay person to understand. Finally, it has been validated using

exploratory factors analysis (Paulhus & Carey, 2011). As this validation involved a non-clinical sample, and there was not sufficient time and resources during the thesis to recruit the numbers required for satisfactory validation in the OCD and autism samples, the present investigation could only be exploratory.

The same 5-point Likert scale of "strongly disagree" to "strongly agree" was retained from the original FAD-Plus (Paulhus & Carey, 2011). However, to keep the overall number of items in the current investigation to a minimum, only 14 of the 27 questions from the FAD-Plus were used. These consisted of all the questions within the two constructs of Personal Control and Scientific Determinism (see Appendix 11). As all four constructs were demonstrated to be relatively independent (Paulhus & Carey, 2011), it is likely they could be validly retained in isolation. These two constructs were retained on theoretical grounds. The Personal Control construct may be related to issues relating to OCD and compulsive behaviour, as issues such as inflexibility (Goodman, 1989a) are understood to be central to OCD. However, Scientific Determinism may be specifically relevant to individuals with autism. It has been argued individuals with autism typically demonstrated a more physical understanding of the world around them (Baron-Cohen, 2008), with a preference for non-social information (Wheelright & Baron-Cohen, 1998; Jeste & Nelson, 2009; Wheelwright & Baron-Cohen, 1998). It is worth noting, however, that Paulhus and Carey (2011) emphasised that Personal Control and Scientific Determinism are not necessarily dichotomous concepts.

The full version of the Compulsive and Repetitive Trait questionnaire (see 7.2) was used for this study. This questionnaire combines repetitive traits from both OCD (Yale-

Brown Obsessive Compulsive Scale II; Storch et al., 2010a) and autism (Repetitive Behaviour measures; Moss et al., 2009), along with item-level mood related questions.

8.3.2. Design.

The pilot study employed the same design as outlined in the main Compulsive and Repetitive Trait study (see section 7.4.1).

8.3.3. Sample size and power calculation.

Due to the lack of relevant studies, the same rational for the power calculations was used for this pilot study as in the main Compulsive and Repetitive Trait study (see section 7.4.3)

8.3.4. Participants.

One-hundred and four adults took part in the study. For the two disorder groups (OCD and autism) there was a fairly even distribution of participants across the age ranges, whilst there was a bimodal distribution of age in the control group reducing the likelihood of this group being representative of the wider population (see Figure 8.2). Inclusion and exclusion criteria were identical to the main investigation (see section 7.4.4).

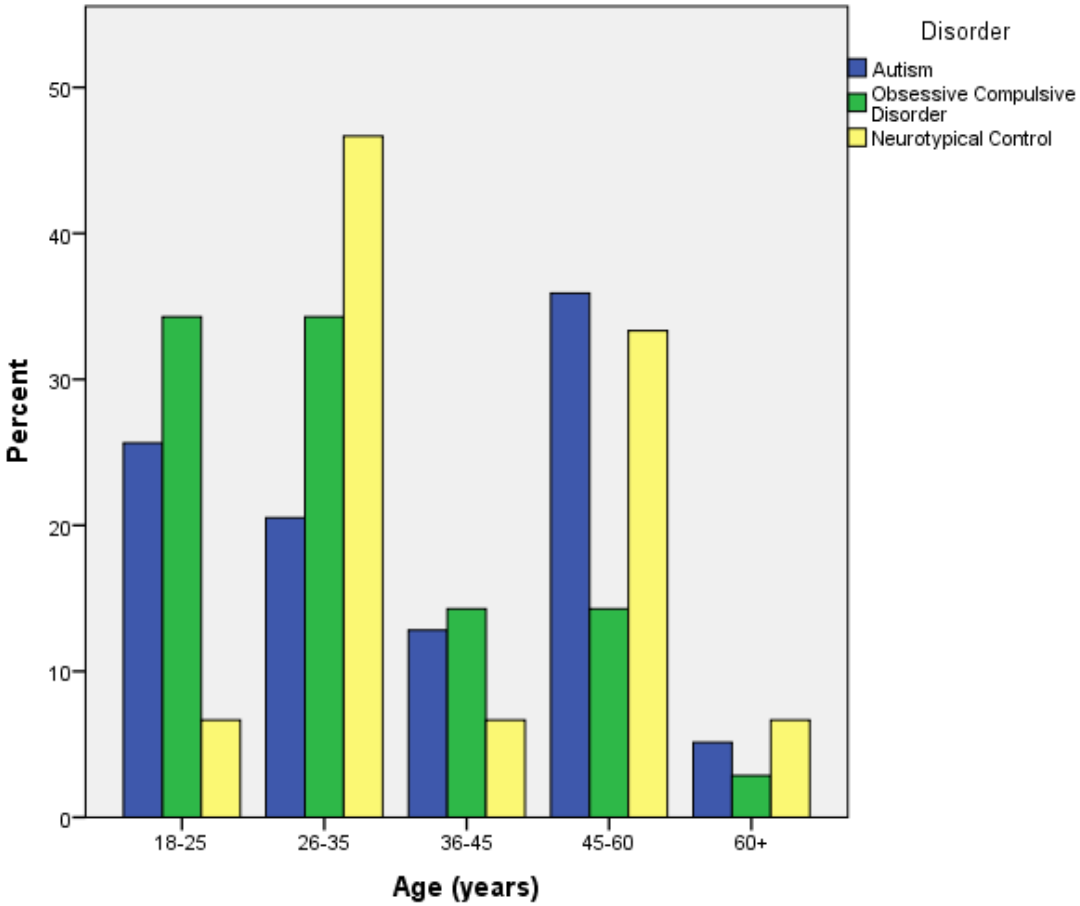

Figure 8.2. Percentage number of participants within each age bracket for each disorder.

Table 8.1. *Frequencies and percentages for demographic variables.*

Demographic variable	Category	Disorder		
		Autism (n = 39)	OCD (n = 35)	Neurotypical (n = 30)
Age	18-25 years	10 (25.6)	12 (34.3)	2 (6.7)
	26-35 years	8 (20.5)	12 (34.3)	14 (46.7)
	36-45 years	5 (12.8)	5 (14.3)	2 (6.7)
	45-60 years	14 (35.9)	5 (14.3)	10 (33.3)
	60+ years	2 (5.1)	1 (2.9)	2 (6.7)
Gender	Male	21 (53.8)	7 (20)	4 (13.3)
	Female	18 (46.2)	28 (80)	26 (86.7)
Country	United Kingdom	35 (89.7)	25 (71.4)	28 (93.3)
	Other	4 (10.3)	20 (28.6)	2 (6.7)
Language	English	37 (94.9)	30 (85.7)	29 (96.7)
	Other	2 (5.1)	5 (14.3)	1 (3.3)
Ethnicity	White (British)	34 (87.2)	25 (71.4)	26 (86.7)
	White (Irish)	0 (0)	0 (0)	1 (3.3)
	Other white	4 (10.3)	6 (17.1)	0 (0)
	Other black	0 (0)	0 (0)	0 (0)
	Indian	0 (0)	1 (2.9)	1 (3.3)
	Chinese	0 (0)	1 (2.9)	2 (6.7)
	Mixed	1 (2.6)	0 (0)	0 (0)
	Other	0 (0)	2 (5.7)	0 (0)

Percentages appears in parentheses.

8.3.5. Setting.

As in the main investigation (see section 7.4.5), participants accessed all elements of the study online, using Online Surveys (JISC, 2018).

8.3.6. Ethical approval.

The same ethical procedure was followed as in the Compulsive and Repetitive Trait questionnaire study (see section 7.4.6).

8.3.7. Recruitment.

Participants with autism and OCD were recruited via email and social media access to autism and OCD services/groups in the United Kingdom (including Facebook and Twitter sites in the UK and the United States), as outlined in the main investigation (see section 7.4.7). Only the neurotypical participants (*n* = 30) recruited in the second stage (see section 7.4.7) were asked to complete the FAD-Plus, as this measure had not been included in the original recruitment of 170 neurotypical controls. All analyses within this chapter refer to the 30 neurotypical control participants. Two of these 30 neurotypical control participants were recruited through the adverts circulated via the autism services. Six were recruited through adverts to either the OCD and/or the autism services. The remaining 22 were recruited via opportunity sample via the lead researcher's personal social media accounts (Facebook and Twitter), as described in section 7.4.7.

8.3.8. Procedure.

Following response to advertisement, participants completed the entire CaRT (see Appendices 1, 2 and 3) and free will questionnaires (see Appendix 11), accessed via Online Surveys (JISC, 2018), following reading a comprehensive participant information sheet (see Appendix 9). As in the main investigation, participants were required to self-report all information, including diagnosis.

8.3.9. Statistical analysis.

Data was analysed using the statistical software IBM SPSS 22.0 for windows. No data was excluded due to the strict design of the questionnaire (using Online Surveys). As a novel investigation, the Free Will scores were analysed for reliability. Cronbach's alpha

was used to analyse internal consistency of the free will questionnaire items, i.e. how closely the items within the groups (either Scientific Determinism or Personal Control) are related to each other. To analyse the relationship between the measures of Compulsive and Repetitive Traits and free will scores, Spearman's rho (non-parametric test) calculations were performed, due to ordinal data being analysed. Finally, to identify whether there were significant differences in the distribution of the demographic data, Fisher's exact tests were run to compare the differences between observed and expected frequencies of gender, age, ethnicity, language and country of residence between the OCD and autism groups.

8.4. Results

This section provides the results of the comparative analyses between the free will beliefs and Compulsive and Repetitive Traits between the two clinical groups (autism and OCD) and the neurotypical control group. Following this results section, the implications of these results are presented in the discussion (see section 8.5), in context of the Compulsive and Repetitive Trait framework. First, definitions of key terms are outlined to provide meaning to the various datasets collected within this investigation.

Definition of key terms

In addition to the terms defined in section 7.5:

Free will count: This overall total relates to the number of overall "disagree" counts subtracted from the overall "agree" counts for each of the two free will belief constructs (i.e. Scientific Determinism and Personal Control).

Weighted free will scores: Paulhus and Carey (2011) fail to report how they applied weighting to their 5-point Likert Scale of "strongly disagree" to "strongly agree". The decision was based on the most logical weighting items rated as either "strongly agree" or "strongly disagree". Each of these counts were multiplied by a factor of two. The new weighted score for "strongly agree" was added to "agree" scores: the new weighted score for "strongly disagree" was added to "disagree" scores. Finally, the total new weighted score for disagree was subtracted from the new weighted total score for agree.

8.4.1. Sample characteristics.

Due to small sample sizes within cells, Fischer's exact test of independence was performed to examine the relationship between demographic characteristics and disorder. This relationship was significant for: gender (X^2 (2, 104) = 15.3, $p < .001$); age (X^2 (2, 104) = 15.0, $p = .044$); or country of residence (X^2 (2, 104) = 6.43, $p = .033$). There was no significant association between ethnicity and disorder (X^2 (2, 104) = 14.0, $p > .05$) or first language and disorder (X^2 (2, 104) = 2.79, $p = .24$). Within the autism sample, 54% ($n = 21$) of the participants were male, whereas 20% ($n = 7$) and 13% ($n = 4$) of the OCD and control sample, respectively, were male. As Figure 8.2 illustrates, relatively few control participants ($n = 1$) were between 18- to 25-years, compared to the OCD and autism groups ($n = 12$ and $n = 10$, respectively). However, there was no association between disorder and age below 35 years (X^2 (2, 104) = 3.85, $p > .05$), which indicates that participants were comparable in age across groups when considering young and old adults (with 35 years as a cut-off). Whilst relatively few participants with autism and controls resided outside of the United Kingdom (10% and 7%, respectively), a much larger proportion of the participants with OCD resided

outside of the United Kingdom (*n* = 10; 29%). Within the OCD group, these additional countries of residence consist of: United States (n = 5); Taiwan; Canada; Italy; Ukraine; and India. There was a similar trend for first language, with relatively few participants with autism and controls speaking a language other than English (*n* = 2 and *n* = 1; 5% and 3%), whereas a larger proportion of participants with OCD spoke an alternative first language (*n* = 5; 14%).

8.4.2. Free Will scores.

A mean count of Free Will scores for both Personal Control and Scientific Determinism can range from -7 (indicating the participant disagrees with the construct) to 7 (indicating the participant agrees with the construct). As shown in Table 8.1, all mean Free Will scores were negative, indicating the participants generally disagreed with both Personal Control and Scientific Determinism beliefs. However, it is notable the standard deviations across all these results are very large, indicating a large variance in Free Will beliefs within each group (see Figures 8.3 and 8.4).

Table 8.2*. Means and standard deviations for Compulsive and Repetitive Traits scores.*

Measure	Variable	Disorder		
		Autism (n = 39)	OCD (n = 35)	Neurotypical (n = 30)
Free will Personal Control	Mean count	-.28 (3.83)	-.71 (3.18)	-2.1 (3.76)
	Weighted score	-0.077 (4.52)	-1.00 (3.77)	-2.43 (4.10)
Free will Scientific Determinism	Mean count	-1.15 (3.00)	-1.29 (3.37)	-1.90 (3.26)
	Weighted score	-2.05 (4.06)	-2.09 (4.60)	-2.43 (3.69)

Standard deviation appears in parentheses.

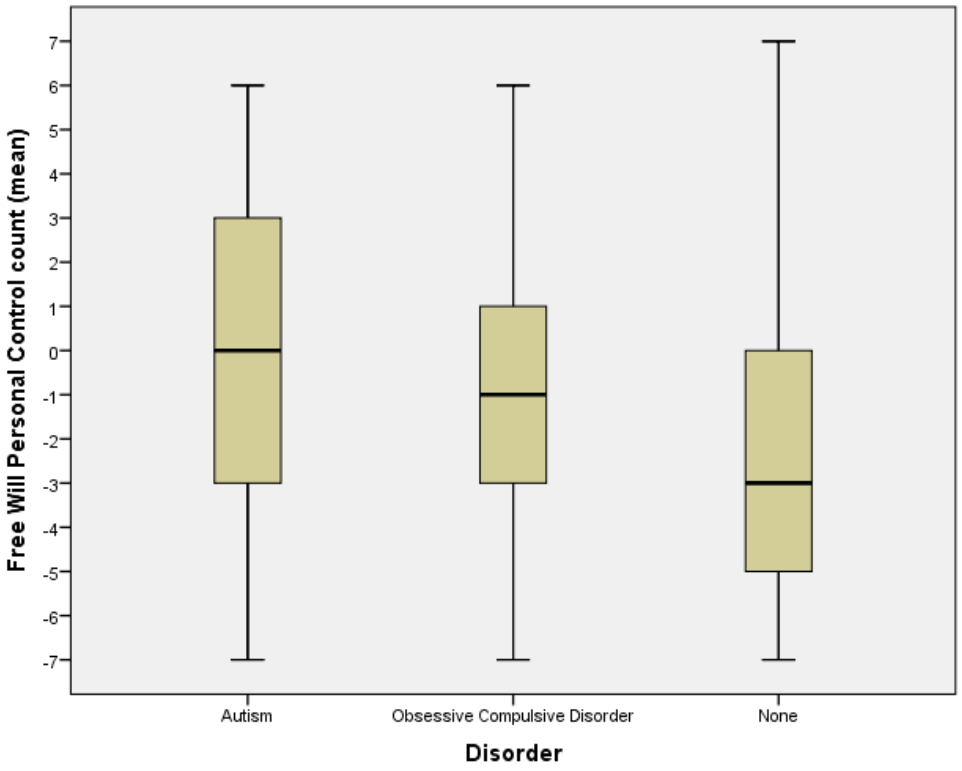

Figure 8.3. Simple boxplot of mean self-reported Free Will Personal Control count by disorder.

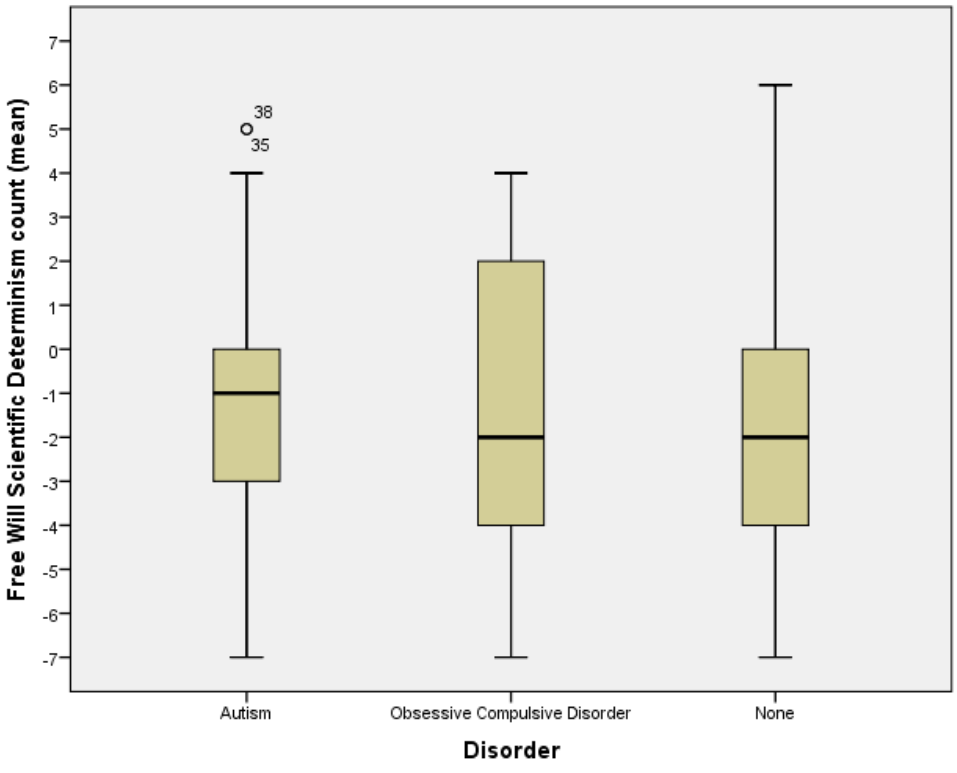

Figure 8.4. Simple boxplot of mean self-reported Free Will Scientific Determinism count by disorder.

8.4.3. Main effects for free will traits.

Cronbach's alpha was calculated to test for internal consistency for the free will items across all participants. For the seven Personal Control items, the internal consistency was moderate (α = .71) and for the seven Scientific Determinism items the internal consistency was acceptable (α = .66). Whilst these scores are around the questionable range (Cho and Kim, 2015), they were comparable to the alpha score of .66 reported for both constructs by Paulhus and Carey (2011). The analysis indicated all the items contributed sufficiently well to the overall consistency: the only item on the Personal Control subscale which increased Cronbach's alpha if deleted was "people have complete control over the decisions they make" (α = .726); and the only item on the Scientific Determinism subscale which increased Cronbach's alpha if deleted was "people's biological makeup determines their talents and personality" (α = .667).

One-way analyses of variance (ANOVA) were performed to compare differences in the means between the number, frequency and mood of Repetitive Behaviours between the three groups. There were found to be no significant group differences for any measures of free will: mean Personal Control counts ($F(2, 101) = 2.36$, $p = .099$, $\eta_{p2} = .045$); mean Scientific Determinism counts ($F(2, 101) = 0.50$, $p = .61$, $\eta_{p2} = .010$); Scientific Determinism weighted ($F(2, 101) = 0.084$, $p = .92$, $\eta_{p2} = .002$). Despite approaching significance, the Personal Control weighted scores was also non-significant ($F(2, 101) = 2.73$, $p = .070$, $\eta_{p2} = .051$).

8.4.4. Compulsive and Repetitive Trait and free will correlations.

Bivariate correlations were calculated between the free will scores and both the Repetitive Behaviour and OCD trait scores. Univariate correlations (Spearman's rho)

indicated, across all groups, there were moderate correlations between Personal Control counts and Scientific Determinism counts ($r(104) = .38$, $p < .001$) and between the weighted Personal Control and Scientific Determinism scores ($r(104) = .39$, $p < .001$).

However, across all the groups there were no significant correlations between Personal Control scores with any Compulsive or Repetitive Trait (CaRT): mood associated with Repetitive Behaviours ($r(104) = .12$, $p = .91$); total number of OCD traits ($r(104) = -.011$, $p = .92$); or mood associated with OCD traits ($r(104) = .077$, $p = .44$); frequency of OCD traits ($r(104) = -.039$, $p = .70$). Again, despite approaching significant the correlation with number of Repetitive Behaviours ($r(104) = .18$, $p = .065$) and Repetitive Behaviour frequency ($r(104) = 0.17$, $p = .078$) were non-significant.

Similarly, there were also no significant correlations between Scientific Determinism scores with any CaRT: mood associated with Repetitive Behaviours ($r(104) = .15$, $p = .12$); Repetitive Behaviour frequency ($r(104) = .16$, $p = .10$); total number of OCD traits ($r(104) = .12$, $p = .22$); or mood associated with OCD traits ($r(104) = .048$, $p = .63$); frequency of OCD traits ($r(104) = .11$, $p = .27$); although again the correlation with number of Repetitive Behaviours approached significance ($r(104) = .18$, $p = .065$);

Bivariate correlations (Spearman's rho) were also analysed within each condition to test for the association between free will beliefs and repetitive traits, for each disorder. For the autism group, these analyses indicated the two free will scales were not significantly correlated ($r(38) = .09$, $p = .60$). As Table 8.2 illustrates, there were only two significant correlations in the autism group between any CaRT and measure

of free will: moderate significant correlations were identified between Scientific Determinism and both total number of Repetitive Behaviours ($r(38) = .34$, $p = .033$) and frequency of Repetitive Behaviours ($r(38) = .32$, $p = .48$). However, it is perhaps notable all significant results disappeared when using the weighted Scientific Determinism scores, neither was there a significant finding for current Repetitive Behaviours and this free will measure ($r(38) = .24$, $p = .14$).

For the OCD group, there were no significant correlations between any measure of free will and CaRT, although for this clinical group there was found to be a large significant correlation between the two measures of free will ($r(33) = .64$, $p < .001$) (see Table 8.3).

For the control group there was a medium significant correlation between the Personal Control and Scientific Determinism items ($r(30) = .41$, $p = .025$). There was only found to be one significant correlation between any CaRT measure and any measure of free will (see Table 8.4): a moderate correlation was identified between the weighted Personal Control score and overall mood associated with OCD traits ($r(30) = .37$, $p = .047$). The correlation between the weighted Personal Control score and the weighted OCD mood score approached significance ($r(38) = .36$, $p = .051$).

Implications of these findings, in line with the research hypotheses and the wider context of how free will beliefs may affect clinical symptoms, are discussed in the following section.

Table 8.3. *Spearman's rho correlations between free will scores and Compulsive and Repetitive Trait scores for the autism group.*

	Total number of Repetitive Behaviours	Overall mood associated with Repetitive Behaviours	Repetitive Behaviour frequency (weighted)	Current Repetitive Behaviour total	Repetitive Behaviour mood (weighted)	Total number of OCD traits	Overall mood associated with OCD traits	OCD trait frequency (weighted)	OCD trait mood (weighted)	Free Will Personal Control count	Free Will Scientific Determinism count	Free Will Personal Control total (weighted)
Free Will Personal Control count	0.14	-0.07	0.12	0.21	-0.08	-0.24	0.17	-0.20	0.20			
Free Will Scientific Determinism count	0.34*	0.13	0.32*	0.24	0.15	0.22	-0.07	0.19	-0.02	0.09		
Free Will Personal Control total (weighted)	0.07	-0.11	0.04	0.14	-0.12	-0.25	0.14	-0.22	0.16	0.98**	0.06	
Free Will Scientific Determinism total (weighted)	0.28	0.11	0.25	0.24	0.12	0.18	-0.13	0.15	-0.02	0.11	0.97**	0.11

Note: $N = 39$, * $p < .05$, ** $p < .01$ (two-tailed)

Table 8.4. *Spearman's rho correlations between free will scores and Compulsive and Repetitive Trait scores for the OCD group.*

	Total number of Repetitive Behaviours	Overall mood associated with Repetitive Behaviours	Repetitive Behaviour frequency (weighted)	Current Repetitive Behaviour total	Repetitive Behaviour mood (weighted)	Total number of OCD traits	Overall mood associated with OCD traits	OCD trait frequency (weighted)	OCD trait mood (weighted)	Free Will Personal Control count	Free Will Scientific Determinism count	Free Will Personal Control total (weighted)
Free Will Personal Control count	-0.15	-0.09	-0.18	-0.17	-0.09	-0.23	0.12	-0.16	0.16			
Free Will Scientific Determinism count	-0.16	0.09	-0.15	-0.16	0.07	-0.04	0.07	0.02	0.02	0.64**		
Free Will Personal Control total (weighted)	-0.18	-0.08	-0.21	-0.20	-0.09	-0.25	0.12	-0.19	0.15	0.99**	0.63**	
Free Will Scientific Determinism total (weighted)	-0.11	0.09	-0.10	-0.11	0.08	-0.07	0.09	0.003	0.02	0.65**	0.97**	0.64**

Note: $N = 35$, ** $p < .01$ (two-tailed)

Table 8.5. Spearman's rho correlations between free will scores and Compulsive and Repetitive Trait scores for the control group.

	Total number of Repetitive Behaviours	Overall mood associated with Repetitive Behaviours	Repetitive Behaviour frequency (weighted)	Current Repetitive Behaviour total	Repetitive Behaviour mood (weighted)	Total number of OCD traits	Overall mood associated with OCD traits	OCD trait frequency (weighted)	OCD trait mood (weighted)	Free Will Personal Control count	Free Will Scientific Determinism count	Free Will Personal Control total (weighted)
Free Will Personal Control count	0.30	0.02	0.32	0.31	0.03	0.13	0.26	0.15	0.27			
Free Will Scientific Determinism count	0.27	0.25	0.27	0.28	0.22	0.20	0.24	0.17	0.26	0.41*		
Free Will Personal Control total (weighted)	0.17	-0.11	0.19	0.19	-0.09	0.02	0.37*	0.03	0.36	0.96*	0.38*	
Free Will Scientific Determinism total (weighted)	0.19	0.13	0.19	0.20	0.10	0.11	0.32	0.07	0.32	0.41*	0.96**	0.42*

Note: $N = 30$, * $p < .05$, ** $p < .01$ (two-tailed)

8.5. Discussion

Overall, there was little evidence to indicate differences between the autism, OCD and neurotypical samples in self-reported free will scores. With regards to hypotheses 1 and 2, there were found to be no significant differences in self-reported free will between the three groups. This suggests beliefs in free will may not differ between adults with autism, OCD and neurotypical peers. Whilst there was huge variation of reported free will beliefs, the variance was not significantly different between either clinical group or the neurotypical controls, which suggests free will beliefs are heterogenous and unrelated to disorder. However, as this appears to be the first direct investigation of free will beliefs in these clinical groups, these results need replicating.

Whilst there were no statistical differences in any free will scores between the groups, the Personal Control and Scientific Determinism scores were strongly correlated in the OCD group, moderately correlated in the control group, but were not significantly correlated in the autism group. This suggests both types of free will may be independent constructs in autism, but may be related in OCD and in neurotypical individuals. In line with the hypotheses, we tentatively suggest the lack of correlation between the two free will constructs in autism may be due to high relevance of the Scientific Determinism category (e.g. Baron-Cohen, 2008), but low relevance of the Personal Control construct, as self-reflection can be an issue (e.g. Baron-Cohen, 1989).

Although beliefs in free will may not differ between the groups, hypotheses 3 and 4 were designed to determine if belief in free will affect the presentation of Compulsive and Repetitive Traits (CaRTs) in autism and OCD. Whilst there were no correlations

between free will and CaRTs within the OCD group, for the autistic participants there were found to be significant correlations between the Scientific Determinism construct and both the total number (i.e. past and current) and frequency of Repetitive Behaviours. This finding is partly in line with the hypothesis. Scientific Determinism was expected to be specifically relevant in autism due to increased preference for processing physical aspects of the world (Baron-Cohen, 2008). Repetitive Behaviours are also likely to be specifically related to autism: this measure arose from research in autism and intellectual disabilities (e.g. Bodfish et al., 2000; Leekam et al., 2007; Moss et al., 2009). This result again needs replicating, particularly as the large number of correlations increased the likelihood of type-I errors (i.e. false positives).

However, two findings challenge this evidence. The significant result for Repetitive Behaviours and Scientific Determinism in the autistic group disappeared when using the weighted free will scores (accounting for the strength of endorsement). This may suggest the finding is not entirely valid, or the method for weighing the scores is not correct. Additionally, the correlation between Scientific Determinism and Repetitive Behaviours in autism is true only for all (past and current) Repetitive Behaviours; for current Repetitive Behaviours the correlation was non-significant. As this is more difficult to explain, it would be logical to assume the significant correlations in autism between Scientific Determinism and Repetitive Behaviours were false positives.

For the control group, a correlation between Personal Control and mood associated with OCD traits was identified. This may indicate subclinical OCD traits may be related to this measure of free will belief. However, since no other measure of OCD traits were

found to be associated with free will in the control group, and because OCD traits were relatively low in the control group, it is possible this result is also a false positive.

In summary, free will beliefs in autism, OCD and neurotypical controls appeared to be comparable. The overall evidence indicates clinical behaviour (both disorder and repetitive traits) may be relatively independent of free will beliefs, which is interesting considering the strong evidence demonstrating the effect of free will beliefs on behaviours in the general population (e.g. Baumeister & Brewer, 2012). It is possible, therefore, the relationship between free will beliefs and behaviour may be limited to moral behaviours, as opposed to individual symptomology. Whilst significant correlations between beliefs in Scientific Determinism and aspects of Repetitive Behaviour indicate an autism-specific association between free will and behaviour, replication is needed to determine the results is not a false positive. This is all the more important since the internal consistency for the two free will constructs were within the questionable range (Cho and Kim, 2015), suggesting these items may not be a consistent measure of both Scientific Determinism and Personal Control beliefs.

8.6. Research Limitations and Strengths

The present investigation appears to be the first study to investigate self-reported free will in autism. This approach is arguably more valid than experimental designs (e.g. Glazebrook et al., 2008), enabling the researcher to assess, more directly, free will in participants. The internal consistency of the free will items was found to be questionable, or at the very best acceptable (Cho and Kim, 2015). It is notable the FAD-Plus has been used in few investigations and has not been validated in clinical populations. The limited size of this pilot investigation means the results need

replicating in larger samples to assess the validity of this measure in samples of adults with autism and OCD. This is particularly important as considerably fewer participants were recruited than generated by the sample size calculation. There was a reluctance (from an ethical position) to keep continuing to recruit neurotypical participants to compensate for the omitted items (as described in section 7.4.7). Also, there became a cap on the number of clinical participants willing to take part despite widespread advertising (see section 7.4.7), possibly because of the lengthy time the study demanded for this group. For the control group, whilst t-test calculations indicated comparable results between this subset of (*n* = 30) neurotypical controls and the larger sample of (*n* = 170) controls, no data on free will was collected on the larger neurotypical group, therefore it is not known if these free will scores are representative of the wider population.

As in the main investigation (see Chapter 7), a major limitation is the unrepresentative sample across all groups. In particular, the proportion of females was particularly skewed, which may have affected the analyses. There is evidence, for example, to suggest that repetitive traits may be presented differently in females with autism (Van Wijngaarden-Cremers et al., 2014). Although the methods for administering the study on the internet may have increased ecological validity in autism (Benford, 2008; Ozonoff, 1995), overall this procedure represented a sampling bias, therefore replication would be needed using more comprehensive recruitment methods. This is particularly significant for the neurotypical group, who mostly responded to advertisement on personal social media (see section 7.4.7). More importantly, it is possible this pilot study was underpowered. The original power calculation used (see section 7.4.3) indicated a minimum of 159 participants, split equally across all three

groups. However, only 104 participants were recruited, unevenly split between the autism (*n* = 39), OCD (*n* = 35) and neurotypical control groups (*n* = 30). Whilst this calculation was based on a more modest figure, to account for the lack of previous relevant research, it is possible the lack of numbers has led to unreliable data and analyses.

In summary, whilst the overall results indicate little effect of free will beliefs on clinical symptoms, due to the described limitations this conclusion is at best very tentative. The hypotheses were originally based on extremely limited evidence and the large number of calculations undertaken to scope as part of this pilot study greatly increased the possibility of Type I errors i.e. incorrectly rejecting a true false hypothesis.

Chapter 9. Critical Appraisal

This appraisal will identify some of the practical and theoretical issues encountered during the current investigation. It will reflect on the main methodological issues, mainly with respect to the recruitment of participants, as well as the challenges encountered when attempting to narrowly define what is a broad area of research (i.e. repetitive traits in autism).

9.1. Origins of the study and motivation for the research

The theoretical perspective behind the research was designed during time working as a practitioner in a service for children with autism. Being personally interested in free will, the original goal was to attempt to understand whether free will has an impact on clinical behaviour (specifically compulsive and repetitive traits). Being novel, this was always going highly challenging – due to the contentious philosophical arguments behind free will. Before being able to realise the research on free will, however, it became initially necessary to summarise the breadth of research on Compulsive and Repetitive Traits (CaRTs). Unfortunately, it was identified early on that establishing this CaRT framework would take most (if not all) of the time needed during the thesis. This meant that the investigation of free will was unlikely to be undertaken. It was originally planned for an experimental approach to the investigation of free will. This was deemed highly relevant as there have been serious criticisms levelled at the validity of the types of design in Glazebook (2015) and Libet et al., (1983). However, to be able to investigate free will at all (in the perspective of a CaRT approach), a compromise was made to include the free will investigation as a cross-sectional questionnaire and as a pilot study. There was insufficient time – within the boundaries of a part-time PhD – to

be able to create what would need to be a ground-breaking experimental approach to free will.

9.2. Methodological Considerations

9.2.1. Ethical considerations.

The risk within the investigation was deemed to be low. The only ethical consideration related to the highly sensitive nature of some of the questions (e.g. aggressive sexual activity) and the possibility of someone becoming distressed through awareness of clinical issues (OCD traits). The former issue was somewhat resolved by making the nature of the questions explicit in the Participant Information Sheet. The latter issue was minimised through providing participants with resources for support at the beginning and the end of the study.

9.2.2. Recruitment of clinical participants.

Recruitment proved to be more difficult than first envisaged. As recruitment was initially slow, a £50 cash prize was advertised to motivate more participants to take part. Even so, the return rate appeared to be low. Following the incentive, 47 participants completed the study, whilst around 1205 accessed the questionnaire. Even though it is likely some of these "visits" were for the same people, the return rate of 3.9% is extremely low and, along with making the study online-only, is likely to reflect a sampling bias.

9.2.3. Statistical Analyses.

As described in section 7.4.7, an insufficient number of participants was recruited compared to that identified by the power calculation. Furthermore, the lack of

relevant studies was a major limitation in producing this calculation. Additionally, due to the exploratory nature of the pilot study, which being a novel experiment was conducted without precedence, the high number of statistical analyses increases the risk of a type-I error (obtaining a false positive result).

9.2.4. Selection of measurement tools.

There are various issues with the measurement tools. Firstly, for the Y-BOCS-II-SR items, although studies have been recently showing a trend for item-level analysis, it is notable the Y-BOCS was created to assess severity of OCD traits on a global level, rather than on the item-level. The self-report version of the Y-BOCS has not been validated in adults with autism, although research over the past decade has consistently demonstrated that OCD appears to be more prevalent in autism than in the general population. The Repetitive Behaviour tool has also not been validated in adults with autism, although Moss et al. (2009) demonstrate good concurrent and content validity between the Repetitive Behaviour Questionnaire and the Autism Spectrum Quotient in a large and varied sample.

Finally, it is likely that, due to the large number of items used in the study (90 items), participants may have suffered from order effects such as fatigue or boredom. However, the design of the questionnaire attempted to minimise this possibility (e.g. an option to finish later on each page and allowing the participant to be able to complete the study to suit themselves). The data appears to show that participants were motivated to answer positively throughout the study, with no clear indication of random answering.

9.2.5. Other considerations.

Various potentially confounding variables was not investigated during the study. Therefore, it is not possible to rule out any of the results because of factors such as depression, or medication use. Whilst the recruitment was very wide, the sample size was not sufficiently large enough to provide confidence these factors were unlikely to play a part in the outcomes.

9.3. Conclusions

In summary, whilst there were methodological issues throughout the investigation (not least the difficulties in recruiting participants), compounded by the challenges of completing the thesis on a part-time basis, the small number of significant results which were identified add to what is limited in a variety of areas of research, i.e.: understanding repetitive traits in adults with autism; comparison between Repetitive Behaviours and OCD traits; and the study of free will in autism and OCD. Despite modest results, it is hoped this investigation may encourage others to continue to compare the difference between ego-dystonic and ego-syntonic traits in autism as well as the effect of free will beliefs, in OCD and other neurological disorders.

Chapter 10. Conclusions

The current research investigated a Compulsive and Repetitive Trait framework. This framework is based on links between symptomology in autism and OCD (e.g. Anholt et al., 2010; Bejerot et al., 2001; Hutton et al., 2008; Hollander et al., 2003; Ivarsson, 2008; Russell et al., 2005), in addition to increasing evidence of repetitive traits in autism not just being ego-syntonic (Baron-Cohen, 1989; Rice, 2009), but also potentially ego-dystonic (Barber, 2015; Saddington, 2013). The evidence presented appeared to generally support the relevance of a combined Compulsive and Repetitive Trait framework and its ability to distinguish subtle differences in symptomology between disorders such as OCD and autism. Furthermore, it also demonstrates the need to understand symptoms at the item-level, rather than making global-level assumptions. These findings would indicate the necessity to replicate the study, testing the CaRT framework using a larger and more representative sample of participants with OCD and autism (including subgroups).

There is some indication the novel measure of Compulsive and Repetitive Traits (CaRTs) may offer some insight into the similarities and differences between symptoms in OCD and autism. OCD traits and Repetitive Behaviours do not appear to form opposites on a positive/negative mood or ego-dystonic/ego-syntonic dichotomy. OCD traits seem to be elevated in autism, even if not as extreme in OCD-only individuals. Presence of repetitive traits may be a feature of clinical disorder generally: there were no significant differences in the number of reported Repetitive Behaviours between the two clinical groups. However, these traits were distinguished by more positive mood in the autism group, whilst the mood associated with Repetitive Behaviours

were comparable between the OCD and control group. Taken together, a Compulsive and Repetitive Trait framework is likely to be complex.

The evidence of an association between free will beliefs and Compulsive and Repetitive Traits is unclear. In general, it can be tentatively stated there appear to be no differences in self-reported free will between adults with autism, OCD or neurotypical individuals. Free will beliefs appear to be very varied across all participants, irrespective of clinical group. Additionally, beliefs in free will do not appear to predict Compulsive and Repetitive Traits, indicating philosophical beliefs in free will may be independent of clinical (repetitive) behavioural measures.

10.1. Future Directions for Research

The findings suggest there may be a considerable overlap between Compulsive and Repetitive Traits. This investigation started with an initial premise of identifying OCD-traits as ego-dystonic (unwanted) and, thereby associated with measures of distress. Measures of Repetitive Behaviour, originating from studies of autistic or learning disabilities samples (e.g. Moss et al., 2009) were used as a potential dichotomy to ego-dystonic traits. These supposedly ego-syntonic traits would be expected to be associated with positive (or at least neutral) mood. The results only partially supported this theory (e.g. participants reported OCD-traits associated with positive mood and Repetitive Behaviours associated with negative mood). Therefore, it is plausible there are many other moderating and mediating variables involved. It is certainly likely the measure of mood involved in the Compulsive and Repetitive Trait questionnaire is overly simplistic. Future studies would need to firstly measure ego-dystonic and ego-syntonic properties using questions designed to test how the individual feels about the

traits (e.g. Buckby, 1999; Rice, 2009; Saddington, 2013) and, crucially, to test the psychometric properties of these measures to identify their validity in participants with autism and with OCD.

Additionally, there may be more valid measurement tools available. At the time of investigation there were no validation studies on self-report measures of Repetitive Behaviours in adults with autism. However, Barrett et al. (2015) has since demonstrated the validity in use of another measure in such a sample. The measurement tools utilised by Barrett et al. (2015) – the Adult Repetitive Behaviours Questionnaire-2 – may be a better measure of higher- and lower-order Repetitive Behaviours. Similarly, the free will measure used is only one part of an overall understanding of free will. There is a wealth of empirical research indicating the relevance of concepts such as volition, choice, intention, desire, control, agency, power and self-awareness (Baumeister & Brewer, 2012; Swineburne, 2012). It may be useful to compare self-reported free will with these cognitive components, with the end-goal being to identify a comprehensive pathway between neuropsychology and free will. Cross-sectional questionnaire methods can be empirically sound, but designs testing practical elements of free will in participants (e.g. Glazebrook, 2008) may also be necessary.

One of the major limitations of the studies relates to the recruitment of the participants. In addition to the insufficient number to satisfy the power calculation, the samples were not entirely representative of the wider population groups. Future investigations should seek to address this balance; recruiting more males may increase Repetitive Behaviours variables (Van Wijngaarden-Cremers et al., 2014). Similarly,

identifying and analysing sub-group differences for both OCD and autism samples (e.g. washers/checkers in OCD; High Functioning Autism and Asperger's Syndrome in autism) may help to delineate the heterogeneous nature of both disorders. Recruiting sufficient numbers of participants within each relevant subgroup may, for example, clarify why there was such variance within groups. Recruiting more representative samples may also confirm the tentative evidence of two autism-specific effects in the free will pilot investigation: a lack of correlation between the two free will constructs; and correlations between Scientific Determinism and some measures of Repetitive Behaviours.

A strong correlation between OCD traits and Repetitive Behaviours implies they are interconnected. Future investigations may help to clarify this relationship. As the simple relationship identified between mood and CaRTs is likely to be only part of a much more complex pathway related to the function of repetitive traits, a functional analytical approach is needed to identify all the variables involved. This approach may instead indicate clusters of ego-dystonic (e.g. dislike, frustration, aggravation) and ego-syntonic (e.g. want, pleasure, stimulation) traits.

Overall, the results somewhat indicate future investigations may benefit from more precise item-level measures of mood (rather than global mood, across all traits). Fundamentally, the only valid way of assessing ego-dystonic and ego-syntonic traits may be directly, asking the participants to what extent each Compulsive and Repetitive Trait (CaRT) is wanted or unwanted (e.g. Rice, 2009).

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Appendix 1. Constructs and questions for CaRT questionnaire.

When has this happened?		
Never/ Hardly ever	This has happened in the past, but not in the last 7 days	This has happened in the past 7 days

How often do it usually occur?			
Sometimes (less than 1 hour per day)	Frequently (between 1 and 3 hours per day)	Often (more than 3 and up to 8 hours per day)	Very often (over 8 hours per day)

When is this most likely to happen? (Tick as many as relevant)					
When unhappy/ Stressed	When bored	When busy	When happy	When alone	Around people

How do you usually feel:				
When you are doing this?				
Very good	Good	Neither good or bad	Bad	Very bad
If you are not able to do this?				
Very good	Good	Neither good or bad	Bad	Very bad

Appendix 2. Self-report Yale Brown Obsessive-Compulsive Scale items (Baer, 1991)

1. I fear I will blurt out obscenities.
2. I fear doing something embarrassing.
3. I fear I might harm myself.
4. I have violent or horrific images in my mind.
5. I fear I might harm other people.
6. I fear I will act on an unwanted impulse.
7. I fear I will steal things.
8. I fear that I'll harm others because I'm not careful enough.
9. I fear I'll be responsible for something else terrible happening.
10. I am concerned or disgusted with bodily waste or secretions.
11. I am concerned with dirt or germs.
12. I am excessively concerned with environmental contaminants.
13. I am excessively concerned with certain household cleansers.
14. I am excessively concerned with animals.
15. I am bothered by sticky substances or residues.
16. I am concerned that I will get ill because of contamination.
17. I am concerned that I will contaminate others.
18. I have forbidden or perverse sexual thoughts, images, or impulses.
19. I have sexual obsessions that involve children or incest.
20. I have obsessions about homosexuality.
21. I have obsessions about aggressive sexual behaviour toward other people.
22. I have obsessions about hoarding or saving things.
23. I am concerned with sacrilege and blasphemy.
24. I am excessively concerned with morality.
25. I have obsessions about symmetry or exactness.
26. I feel that I need to know or remember certain things.
27. I fear saying certain things.
28. I fear not saying just the right thing.
29. I fear losing things.
30. I am bothered by intrusive (netural) mental images.
31. I am bothered by intrusive mental (in my head) nonsense sounds, words or music.
32. I am bothered by certain sounds or noises.
33. I have lucky and unlucky numbers.
34. Certain colours have a special significance to me.
35. I have superstitious fears.
36. I am concerned with illness or disease.
37. I am excessively concerned with a part of my body or an aspect of my appearance.
38. I wash my hands excessively or in a ritualised way.
39. I have excessive or ritualised showering, bathing, tooth brushing, grooming, or toilet routines.
40. I have compulsions that involve cleaning household items or other inanimate objects.
41. I do things to prevent or remove contact with contaminants.
42. I check that I did not harm others.

43. I check that I did not harm myself.
44. I check that nothing terrible happened.
45. I check that I did not make a mistake.
46. I check some aspect of my physical condition tied to my obsessions about my body.
47. I reread or rewrite things.
48. I need to repeat routine activities.
49. I have counting compulsions.
50. I have ordering or arranging compulsions.
51. I have compulsions to hoard or collect things.
52. I have mental rituals (other than checking/counting).
53. I need to tell, ask, or confess.
54. I need to touch, tap, or rub things.
55. I take measures (other than checking) to prevent harm or terrible consequences to myself or family.
56. I have ritualised eating behaviours.
57. I have superstitious behaviours.
58. I pull my hair out.

Appendix 3. Repetitive Behaviour Questionnaire (Moss et al., 2009)

I repetitively fiddle with toys or other items.
I repetitively move my whole body, or parts of my body (other than my hands).
I make repetitive hand and/or finger movements.
I need to tidy away any objects that have been left out, whether or not they are put in the place they are "supposed to go".
I organise objects into categories according to various characteristics such as colour, size, or function.
I continually need to see, speak to or contact a particular "favourite" person.
I ask specific questions over and over.
I have a strong preference for a particular functional object to be present at all times.
I have a strong preference for a particular non-functional object to be present at all times.
I repeat particular sounds, phrases or signs (that are unrelated to the situation) over and over.
I carry out a sequence of specific actions/rituals before, during or after a task, a sequence that is always carried out when performing this task and always occurs in the same way.
I repeatedly talk about specific, unusual topics in great detail.
I repeat speech that I have either just heard or heard more than a minute earlier.
I insist on having the same routine or schedule everyday.
I insist that objects always remain in the same place.
I insist on having objects or activities 'complete' or 'whole'.
I repetitively remove small pieces of lint, fluff, crumbs or dirt from surfaces, clothes and objects.
I organise objects into categories according to various characteristics such as colour, size, or function.
I line up or arrange objects.

Appendix 4. Sample size calculation for main investigation.

F tests - ANOVA: Fixed effects, omnibus, one-way

Analysis:	A priori: Compute required sample size			
Input:	Effect size f	=	0.25	
	α err prob	=	0.05	
	Power (1-β err prob)	=	0.8	
	Number of groups	=	3	
Output:	Noncentrality parameter λ	=		9.9375000
	Critical F	=	3.0540042	
	Numerator df	=	2	
	Denominator df	=	156	
	Total sample size	=	159	
	Actual power	=	0.8048873	

Appendix 5. Advertisement sent to recruit neurotypical participants.

Call for participants

Please could you spare 10 to 20 minutes of your time to complete a study which aims to improve knowledge of Obsessive Compulsive Disorder, particularly for people with autism.

As a part-time self-funded PhD student who is working full time to support people with learning disabilities, I need support from as many participants as possible to validate a new measure which will be later used to understand individuals with OCD and with autism.

The study is all online and can be found at http://www.survey.hull.ac.uk/carts. All further information you may need is on the site.

Thanks for your time,

Sam Chegwin

Appendix 6. Advertisement sent to OCD agencies to recruit participants.

APPROX 1 IN 100 CHANCE TO WIN £50

Do you have OCD (or autism) are you over 18 and can you spare some time to complete a questionnaire? **Or do you know someone who has OCD**, is over 18 and might be able to spare some time to complete a study?

The similarities and differences in Repetitive Behaviours in people with OCD and people with autism are still not very well understood. Neither is the rate of OCD in people with autism. Due to my personal interests in both OCD and autism, I am self-funding a PhD which hopes to address both of these issues.

We require as many adults with a **diagnosis of OCD** (or autism, but not both) as possible to complete a new online study by following the link https://hull.onlinesurveys.ac.uk/cart-and-free-will-survey-ocd.

The study can take as little as 20 minutes but more likely around an hour for people who show a lot of repetitive/OCD behaviours. You don't have to complete the study in one attempt as there is an option to "finish later" on each question.

I would greatly appreciate your help. In addition to helping to develop a better understanding of OCD and Repetitive Behaviours in people with autism, after accurate completion of the study you can also enter into a free prize draw for a maximum **1 in 100 chance to win a £50 cash prize**. Further information is on the site or email s.t.chegwin@2012.hull.ac.uk.

Thank you for your time,
Sam Chegwin

If you have any further questions please contact the researcher, Sam Chegwin at:
S.T.Chegwin@2012.hull.ac.uk
(Sam Chegwin, PhD student)

Supervised by:
Dr Tim Alexander, Research Co-ordinator
The Department of Psychological Health and Wellbeing
Aire Building, The University of Hull
Cottingham Road
Hull
HU6 7RX
Email: T.Alexander@hull.ac.uk

Appendix 7. Advertisement sent to autism agencies to recruit participants.

APPROX 1 IN 100 CHANCE TO WIN £50

Do you have autism are you over 18 and can you spare some time to complete a questionnaire? This will help to develop a new assessment tool comparing Repetitive Behaviours in people with OCD and people with autism. **Or do you know someone who has autism**, is over 18 and might be able to spare some time to complete a study?

The rate of OCD in autism is still not very well understood. Neither is the difference between Repetitive Behaviours and OCD in people with autism. Due to my personal interests in autism, I am self-funding a PhD which hopes to address both of these issues.

We require as many adults with a **diagnosis of autism** (or OCD, but not both) as possible to complete a new online study by following the link https://hull.onlinesurveys.ac.uk/cart-and-free-will-survey-asd.

The study can take as little as 20 minutes but more likely around an hour for people who show a lot of repetitive/OCD behaviours. You don't have to complete the study in one attempt as there is an option to "finish later" on each question.

I would greatly appreciate your help. In addition to helping to develop a better understanding of OCD and Repetitive Behaviours in people with autism, after accurate completion of the study you can also enter into a free prize draw for an approximate **1 in 100 chance to win a £50 cash prize**. Further information is on the site or email s.t.chegwin@2012.hull.ac.uk

Kind regards,
Sam Chegwin

If you have any further questions please contact:
Email: S.T.Chegwin@2012.hull.ac.uk (**Sam Chegwin, PhD student)**

Supervised by:
Dr Tim Alexander, Research Co-ordinator
The Department of Psychological Health and Wellbeing
Aire Building, The University of Hull
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Appendix 8. University of Hull Faculty of Health and Social Care Research Ethics Committee approval letter.

University of Hull

PRIVATE AND CONFIDENTIAL

Mr Samuel Chegwin

Faculty of Health and Social Care

University of Hull

HU6 7RX

Faculty of Health and Social Care

Research Ethics Committee

E Rebecca.Straughan@hull.ac.uk

T 01482 464602

REF 187

06th July 2015

Dear Samuel,

RE: Compulsions in autism: theoretical revision and practical applications.

Thank you for your response letter dated 27th June 2015. Following from your amendments to the areas of the proposal which were queried by the committee it is felt that your responses adequately address the points raised.

On the basis of your amendments I am now able to give Chair's approval for your study and wish you every success with your research.

Yours Sincerely

Miss Rebecca Straughan

FHSC Research Ethics Committee

Appendix 9. Participant Information Sheet.

What is the aim of the research? To begin to understand the difference in obsessive, compulsive and Repetitive Behaviours between different people. We also hope to see if beliefs in free will are related to these behaviours.

Why have I been chosen? We are developing a new assessment tool to hopefully better identify Obsessive-compulsive disorder (OCD) in people with autism, therefore we need to recruit people who have OCD, people who have autism and people without either. For the analysis to be accurate, please do not take part if you have: a diagnosis of **both** OCD and autism; a diagnosed psychiatric disorder (e.g. psychosis); current substance misuse; a diagnosis of a learning disability (IQ below 70); a diagnosis of Fragile X; structural brain abnormalities; tuberous Sclerosis complex; or Smith-Lemli-Opitz syndrome.

What will I be asked to do if I took part? You will complete an **anonymous** online questionnaire containing 14 questions on free will followed by 72 OCD questions and 18 Repetitive Behaviour Questions.

It should take from 20 minutes to about an hour to complete (depending on the amount of Repetitive Behaviours you have). There is an option at each stage to "finish later" if you don't have time to finish.

It is important for the investigation that you answer carefully and honestly - please remember your answers are **anonymous**. Due to the nature of the study, it is also necessary that you have a good understanding of the English language to complete the questionnaire.

What happens to the data collected? It will be kept confidentially and used to analyse obsessive-compulsive behaviour in the general population.

How is confidentiality maintained? As the survey is anonymous, all of the information you provide will be completely confidential and you will not be able to be identified in any part of the collection of data or write-up. Cookies (personal data stored by your Web browser) are not collected in this survey.

What happens if I do not want to take part or if I change my mind? You can withdraw at any time by discontinuing the questionnaire. Consent for the study will be given by completion of the questionnaire and once you have submitted your responses your data cannot be withdrawn as the survey is anonymous.

Potential Risks Some people may become distressed when completing this survey as the information requires you to consider whether you have any Obsessive Compulsive Disorder-like thoughts or behaviours. You might want to be aware that a few questions later on in the survey are about deviant sexual thoughts and behaviour. If you wish, you can discontinue at anytime and your data will not be stored or used in the study. Near the beginning and at the end of the survey a screen will be presented containing helpful resources, websites and contact numbers if you feel you need some support or further information.

Will I be paid for participating in the research? Unfortunately you will not be paid or reimbursed for taking part in the study but it is hoped that your answers could help therapeutic intervention in the future.

Where will the research be conducted? Questionnaires will be completed online.

Will the outcomes of the research be published? The aim is for the research to be published in a scientific journal or at a conference.

Who has reviewed the research project? All research is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. The Faculty of Health and Social Care Ethics Committee at the University of Hull has given a favourable review of the

study. This study has also been peer reviewed by the research team at the Department of Psychological Health and Wellbeing at the University of Hull.

Contact for further informationS.T.Chegwin@2012.hull.ac.uk -- researcher, Sam Chegwin. T.Alexander@hull.ac.uk (01482 464030) -- Research Supervisor, Dr. Tim Alexander.

Appendix 10. Entire free responses by participants reporting difficulty in answering.

Disorder	Reason	Response
Autism	Major Difficulty with items	there is mostly not an anser that fits, so often 'other was selected
Autism	Major Difficulty with items	Avoiding Repetitive Behaviours
Autism	Major Difficulty with items	Some of the questions go from asking how you feel if you fear doing something to asking how you when, not if you have done it, ie fearing harming yourself or others to actually harming, implying that you have performed these actions.
Autism	Major Difficulty with items	When I do some behaviours, I don't feel anything but just have to do them
Autism	Major Difficulty with items	Some of the situations cause me a great deal of stress and anxiety so for the purposes of the Study I have interpreted these as bad or very bad. It was difficult converting the emotions into a scale.
Autism	Major difficulty for personal reasons	Explaining feelings is very hard for me
Autism	Major difficulty for personal reasons	This one is the toughest.
Autism	Major difficulty for personal reasons	Some difficulty, as some of my behaviours I am not aware of, eg i have only been diagnosed ASD recently, so for example, I have only just been told that I touch things and people, I was not even aware of this
Autism	Major difficulty for personal reasons	Sometimes I didn't understand the questions.
OCD	Major Difficulty with items	the end boxes that ask how you feel if you do these things. a lot of the time i don't do most of them, just fear doing them, but there was no option not to fill the section in so i just had to answer very bad
OCD	Major Difficulty with items	I found it difficult to answer the questions about how you feel if you accidentally harm people/yourself, as it made me think 'well I am admitting to having done this if I check any of the boxes'
OCD	Major Difficulty with items	The biggest difficulty was trigger questions and words, but that is part of my ERP. One other difficulty was having to answer some follow up questions about how I felt thinking about something and then how I felt if I did that something. In a few cases, I hadn't actually done the thing I think obsessively about.
OCD	Major Difficulty with items	hard putting a timescale to obsessions and compulsions
OCD	Major difficulty for personal reasons	There are some heavy questions in here. I would try and emphasise in the opening page the confidentiality of the survey. I'm beginning to think about it myself now I have been through and I am unsure what the policy was.
OCD	Major difficulty for personal reasons	sometimes i am unaware of when ocd tendancies occur and what triggers them

None	Major Difficulty with items	The idea of assigning a time frequency to when a compulsive or obsessive behaviour occurs is odd in some scenarios. For instance hand washing, it may be ritualised, but only to the degree that the ritual occurs with visits to the toilet. So in that instance, anyone who says they wash ritually every 3 hours is either really concerned about their hands being clean, or has a small bladder and must toilet every 3 hours, the format of the question has no way of discerning the two.
None	Major Difficulty with items	The one about how do you feel - sometimes the feeling is not 'good' or 'bad' but other emotions so for those I put in the middle.
None	Major Difficulty with items	Deciding on the frequency, in terms of how many hours a day.
None	Major Difficulty with items	Deciding whether something is in the bounds of normal. food rituals time scales done apply as you will only eat 3 X daily usually not for 8 hours a day but at each main meal I have to arrange my food but this may still be only 1 hour a day.
None	Major Difficulty with items	I felt it was odd to describe the frequency of an action in hours, rather than referring to the number of times.
None	Major Difficulty with items	I found the questions that I needed to provide more information for slightly difficult to answer. When asked when this happens... for example when stressed, happy, sad etc. I am not of a certain mood when undertaking these actions. They only happen when the specific activities are undertaken.
None	Major Difficulty with items	Because there was no alternative I have had to put an answer which is not completely accurate in order to continue . For instance, I do like to be tidy and put things where they belong, but not obsessively. But simply because everyone in the house knows the keys are kept blah blah. The raw meat is always on the bottom shelf of our fridge because it can drip onto cooked foodstuff. So for basic hygiene reasons but had I said I worry about it this would have given totally the wrong impression. Similar incidences throughout your questionnaire could have given the wrong interpretation and impression hence why I kept putting never etc because it is not through obsessive compulsive behaviour my family ensure we keep basic hygienic habits.
None	Major Difficulty with items	Some of the questions were repetitive. The format was often unconducive to providing a defined answer i.e. measuring frequency of behaviour in hours.

Appendix 11. Free will questions (Paulhus and Carey, 2011).

Personal control construct:

People have complete control over the decisions they make.
People must take full responsibility for any bad choices they make.
People can overcome any obstacles if they truly want to.
Criminals are totally responsible for the bad things they do.
People have complete free will.
People are always at fault for their bad behaviour.
Strength of mind can always overcome the body's desires.

Scientific Determinism construct:

People's biological makeup determines their talents and personality.
Psychologists and psychiatrists will eventually figure out all human behaviour.
Your genes determine your future.
Science has shown how your past environment created your current intelligence and personality.
As with other animals, human behaviour always follows the laws of nature.
Parents' character will determine the character of their children.
Childhood environment will determine your success as an adult.

Appendix 12. Screen prints of the Repetitive Behaviour Questionnaire (Moss et al., 2009) as used in investigation – with permissions from the authors.

Table 3 Repetitive Behaviour Questionnaire items and subscales

	Repetitive Behaviour Questionnaire item:	Subscale
1.	**Object stereotypy:** repetitive, seemingly purposeless movement of objects in an unusual way *E.g. twirling or twiddling objects, twisting or shaking objects, banging or slapping objects.*	Stereotyped behaviour
2.	**Body stereotypy:** repetitive, seemingly purposeless movement of whole body or part of body (other than hands) in an unusual way. *E.g. body rocking, or swaying, or spinning, bouncing, head shaking, body posturing.* Does not include self-injurious behaviour.	
3.	**Hand stereotypy:** repetitive, seemingly purposeless movement of hands in an unusual way. *E.g. finger twiddling, hand flapping, wigging or flicking fingers, hand posturing.* Does not include self-injurious behaviour.	
4.	**Cleaning:** Excessive cleaning, washing or polishing of objects or parts of the body *E.g. polishes windows and surfaces excessively, washes hands and face excessively,*	Compulsive behaviour
5.	**Tidying up:** Tidying away any objects that have been left out. This may occur in situations when it is inappropriate to put the objects away. Objects may be put away into inappropriate places. *E.g. putting cutlery left out for dinner in the bin, removes all objects from surfaces.*	
6.	**Hoarding:** Collecting, storing or hiding objects to excess, including rubbish, bits of paper, and pieces of string or any other unusual items.	
7.	**Organising objects:** Organising objects into categories according to various characteristics such as colour, size, or function. *E.g. ordering magazines according to size, ordering toy cars according to colour, ordering books according to topic.*	
12.	**Rituals:** carrying out a sequence of unusual or bizarre actions before, during or after a task. The sequence will always be carried out when performing this task and will always occur in the same way. *E.g. turning round three times before sitting down, turning lights on and off twice before leaving a room, tapping door frame twice when passing through it.*	
16.	**Lining up or arranging objects:** Arrangement of objects into lines or patterns *E.g. placing toy cars in a symmetrical pattern, precisely lining up story books,*	
18.	**Completing behaviour:** Insists on having objects or activities 'complete' or 'whole' *E.g. Must have doors open or closed not in between, story must be read from beginning to end, not left halfway through.*	
19.	**Spotless behaviour:** Removing small, almost unnoticeable pieces of lint, fluff, crumbs or dirt from surfaces, clothes and objects. *E.g. Picking fluff off a jumper, removing crumbs from the kitchen table.*	
8.	**Attachment to particular people:** Continually asking to see, speak or contact a particular 'favourite' person. *E.g. continually asks to see or speak to particular friend, carer, babysitter or schoolteacher.*	Restricted preferences
10.	**Attachment to objects:** Strong preference for a particular object to be present at all times. *E.g. Carrying a particular piece of string everywhere, taking a particular red toy car everywhere, attachment to soft toy or particular blanket.*	
13.	**Restricted conversation:** Repeatedly talks about specific, unusual topics in great detail. *E.g. conversation restricted to: trains, buses, dinosaurs, particular film, country, or sport.*	
9.	**Repetitive questions**: Asking specific questions over and over. *E.g. always asking people what their favourite colour is, asking who is taking them to school the next day over and over*	Repetitive speech
11.	**Repetitive phrases/signing:** Repeating particular sounds, phrases or signs that are unrelated to the situation over and over. *E.g. repeatedly signing the word 'telephone'.*	
14.	**Echolalia:** Repetition of speech that has either just been heard or has been heard more than a minute earlier. *E.g.: Mum:' Jack don't do that' Jack: 'Jack don't do that'.*	
15.	**Preference for routine:** Insist on having the same household, school or work schedule everyday. *E.g. likes to have the same activities on the same day at the same time each week, prefers to eat lunch at exactly the same time every day, wearing the same jumper everyday.*	Insistence on sameness
17.	**Just right behaviour:** Strong insistence that objects, furniture and toys always remain in the same place. *E.g. all chairs, pictures and toys have a very specific place that cannot be changed.*	

Appendix 13. Image files of the Repetitive Behaviour Questionnaire

These images were taken from the Appendix: Self-Rating Forms in, Baer, L. (1992). *Getting Control: Overcoming Your Obsessions, Compulsions and OCD.* Constable & Robinson: Edinburgh, but have been removed from this public copy of the thesis due to copyright.