

**Use of Q-methodology to identify clinical psychologists' attitudes towards
genetic research affecting people with intellectual and developmental
disabilities**

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Abstract

Title: Use of Q-methodology to identify clinical psychologists' attitudes towards genetic research affecting people with intellectual and developmental disabilities

Background: Advances in molecular genetics are having a growing influence on people with intellectual and developmental disabilities (IDDs) in terms of increased knowledge of genetic causes of disability and new diagnostic technologies. Awareness and consideration of these influences varies among professionals in the field with those involved in direct clinical work putting more emphasis on presenting behaviours rather than underlying etiology (Hodapp & Dykens, 1994). Different professional cultures appear to affect awareness and application of genetic technologies. Considering the potential of this technology to influence the lives of people with IDDs it is important to understand the beliefs and attitudes held towards them by key professional groups in clinical services. Clinical psychologists are one such group and the aim of the current study was to delineate their views on aspects of the "New Genetics." Method: A Q-methodology design explored the subjective opinions of 16 trainee and 15 qualified clinical psychologists towards relevant genetic research. Participants Q-sorted 81 statements reflective of the research topic according to their level of agreement/disagreement with them. Results: Principal component factor analysis with a varimax rotation showed that participants primarily loaded on to three factors [1] a willingness to integrate medical and social models of disability, [2] a preference for a social model of disability and [3] an appreciation of genetic technologies but with need for caution when applying them with people with IDDs. Both amount and type of professional experience affected factor loadings. Conclusions: The varying attitudes of clinical psychologists towards the "New Genetics" and the identified influences affecting them should be considered in the practical application of developments from the field.

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Declaration

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Chapter 1. Introduction

1.1 Study Overview

Since Watson and Crick's seminal discovery of the double helix structure of deoxyribonucleic acid (DNA) in 1953, the complex picture of gene-behaviour influences and interactions has begun to be unravelled. This discovery heralded the birth of molecular genetics (Watson, 1980). Concerned with the structure, function and interaction of genes at a biological level, molecular genetics advanced the science from the traditional study of Mendelian inheritance patterns (Dykens, Hodapp & Finucane, 2000). It was the marriage of biology and genetics. Understanding genetic material gave impetus and the potential to discover the biological underpinnings of the human condition, including human behaviour.

Molecular genetics has brought with it a host of new techniques and technologies to study the human genome. These technologies have had rapid success culminating with the early completion of the Human Genome Project in 2003, which mapped over 20,000 genes in the human genome (U.S. Department of Energy Genome Program's Biological and Environmental Research Information System [BERIS], 2013). In 1980, Dr. David Comings described this ability to study genes directly at the molecular level rather than the consequence level as the "New Genetics." Currently, genetic tests or DNA-based tests exist for more than 1000 genetic disorders, including many intellectual and developmental disabilities (IDDs) such as the relatively common fragile X syndrome and the rarer Emanuel syndrome (BERIS, 2010).

The technologies of the "New Genetics" have resulted in a better understanding of the genetic causes of behavioural syndromes associated with IDDs

(Muir, 2000) and have contributed to the science of behavioural phenotyping, which seeks to profile syndromic characteristics of genetic disorders (Dykens et al., 2000). Prior to molecular technologies, traditional genetic research methods such as family studies and linkage analysis were capable of identifying and describing many behavioural syndromes, however, they were unable to make definitive gene-behaviour links (Muir, 2000). Currently, an estimated 85% of IDD's have a genetic cause (Curry et al., 1997) and with continual application of new molecular technologies, further genetic associations with IDD's are expected in the future (Ellison, Rosenfeld & Shaffer, 2013). Thus, people with an intellectual and/or developmental disability (IDD) have greater access to diagnostic technologies, which means that the proportion of unknown causes of IDD is steadily diminishing. Having an identified behavioural syndrome enables people with an IDD and their family to access a wealth of syndrome-specific information and health and educational professionals to apply this knowledge in treatments and interventions (Harris, 2010).

Despite the recognised advantages of molecular genetics to IDD, there is concern that clinical practitioners working in the area do not adopt and apply this research (Hodapp & Dykens, 1994). Hodapp and Dykens (1994) concluded that the application of genetic developments were hindered in the clinical field due to several misconceptions about etiology-based approaches such as the view that behavioural syndromes occur at a such a low prevalence as to be insignificant to the general treatment of people with IDD. They argued that genetic etiology is not considered as important as presenting behaviour to clinical professionals, such as psychologists, and that in effect two cultures have grown up in the intellectual disability research field; one consisting of geneticists and medical professionals who adhere to an etiology-based approach and conduct single etiology studies and one consisting of

psychologists and behaviourists who have developed advanced measures of behaviour using mixed etiology groups. Since making their initial claim, Hodapp and Dykens (2012) have stated that this gap between the two cultures has reduced through the progress of research in behavioural phenotypes, however, caution in the application of genetic research by professionals in services for people with IDD continues to be acknowledged (Kuna, 2001; Lopez-Rangel, Mickelson & Lewis, 2008).

The relative rapidity of the developments of the “New Genetics” has led some to speculate that the ethical and moral frameworks applying to these developments have not kept up with the science (Baker, Raymond & Bass, 2012; Bessa, Lopes & Maciel, 2012; Muir, 2000). Several researchers in the field of IDD have also highlighted the scepticism towards genetics and genetic research due to the traumatic history it shares with people with IDDs in the guise of the pseudoscience eugenics (Holland & Clare, 2003). Genetic developments associated with an aim of “curing” disability hinders the separation of the “new” and the “old” genetics and affects how people with IDDs and those working in the field engage with them.

The current study seeks to investigate the use and application of ideas and models from the “New Genetics” and the associated field of developmental and behavioural phenotypes by clinical psychologists working with people with IDDs. As well as examining the influence and spread of such ideas (or lack thereof), the study will also examine whether there are barriers, both personal and institutional, to their widespread adoption in services. The study uses a Q-methodology design to investigate attitudes of participants.

This chapter outlines the genetic advances that affect people with IDDs and explores factors such as historical influences and professional identity hypothesised

to form the context of the attitudes of clinical psychologists towards the “New Genetics”. This chapter will also outline Q-methodology and the rationale for its use in this study. Firstly, the rationale for using the term intellectual and developmental disability is outlined.

1.2 A Note on Terminology

In the United Kingdom, the term “learning disability” came into popular usage towards the end of the 20th century to describe a disability that affects intellectual and adaptive functioning. In the 2001 White Paper on the health and social care of people with learning disabilities, “Valuing People” (2001, 2009), learning disability was defined as the presence of a significantly reduced ability to understand new or complex information, to learn new skills (impaired intelligence) and a reduced ability to cope independently (impaired social functioning), which begins before adulthood, with a lasting effect on development. This term replaced the term “mental handicap” which in turn had replaced terms such as “mental deficiency” in England and “mental subnormality” in Scotland.

In other parts of the world, there have been changes in the characterisation and definition of “learning disability”. The United States has replaced the term “mental retardation” with the term “intellectual disability” and this is defined by the American Association on Intellectual and Developmental Disability (n.d.) as a disability characterised by significant limitations both in intellectual functioning and in adaptive behaviour that originates before the age of 18. Other Anglo-phone countries such as Canada, Australia and Ireland have also adopted this terminology (Canadian Association for Community Living, n.d.; Hudson & Radler, 2005; National Disability Authority, 2011).

Thus, whilst definitions of what constitutes “learning disability” appear similar across Anglo-phone countries, worldwide consensus on a term to describe it is more variable. Currently, both the World Health Organization’s (1992) International Classification of Disease (ICD-10) and the American Psychiatric Association’s (1994) Diagnostic and Statistical Manual of Disorders (DSM-IV) use the term “mental retardation”; however, in future versions of these classification systems the terms intellectual disability (ICD-11) and intellectual developmental disorder (DSM-V) will replace the term mental retardation.

The term developmental disability describes lifelong disabilities attributable to mental or physical impairments that manifest prior to adulthood. Developmental disability includes intellectual disabilities as well as pervasive developmental disorders such as autistic spectrum disorders. This thesis adopts the term intellectual and developmental disability to be inclusive of people with intellectual disabilities who may or may not also have a developmental disability such as autism. In this thesis, the terms intellectual and developmental disability and intellectual and developmental disabilities are abbreviated by the acronyms IDD and IDD’s.

1.3 Genetic Advances

Advances in molecular genetics or the “New Genetics” have resulted in a plethora of new understandings of behavioural syndromes associated with IDD (Ellison et al., 2013). These advances have determined the biological underpinnings of syndromes previously grouped according to physical and behavioural characteristics. Many readily identifiable conditions such as Down syndrome, which was described by Langdon Down in 1866, were identified in this way but it was not until the inception of molecular genetics that the biogenetic basis for these conditions

were understood, such as the identification of a third chromosome on chromosome 21 in Down syndrome (Lejeune, Gautier & Turpin, 1959). This section will describe the main molecular genetic technologies and explore in more detail and with reference to the literature, ways in which genetic advances are influencing the lives of people with IDD.

1.3.1 Molecular genetic technology and concepts. Traditional familial studies in the Mendelian sense have given way to advances in molecular cytogenetics, which can reveal sub-microscopic chromosomal readjustments of the karyotype. Cytogenetics itself has advanced within the “New Genetics” history from providing a rudimentary picture of the karyotype, which could identify large chromosomal anomalies such as trisomy 21, to revealing more specific mutations and deletions (Muir, 2000). Prader-Willi syndrome was the first syndrome to have had its genetic etiology determined by a new high-resolution banding; fluorescent in situ hybridisation (FISH), when in the 1980s a microdeletion on chromosome 15q11-q13 was discovered to underpin the condition. Such techniques have continued to develop and currently a new technique of microarray analysis, comparative genomic hybridization, is proving successful in identifying genetic syndromes (Malan et al., 2009; Shaw-Smith, Redon, Rickman, Rio, Willatt, Fiegler et al., 2004; Slavotinek, 2008). Additionally, the use of transgenic animal models transformed the study of gene mutations enabling more sophisticated examination of the pathways between the phenotype and genotype (Branchi, Bichler, Berger-Sweeney & Ricceri, 2003).

As a consequence of advances in molecular technology a range of new concepts, such as genomic imprinting, expanding mutations and uniparental disomy have emerged to explain genetic etiology (Muir, 2000). Genomic imprinting, where one inherited gene from either parent in a gene pair is repressed or inactive through

an epigenetic mechanism has been found to underpin syndromes such as Prader-Willi syndrome and Angelman syndrome (Nicholls, 2005). In the former, a microdeletion on chromosome 15 occurs on the paternally inherited gene in the gene pair, while in the latter a microdeletion on the same chromosome pair but on the maternally inherited gene occurs.

Dynamic or expanding mutations are mutations of a particular gene sequence which when passed from a neuro-typical¹ individual to their offspring continue to expand and excessively repeat to become a full mutation exemplifying a genetic condition (Chiurazzi & Oostra, 2006). This concept was first identified in fragile X syndrome in the early 1990s (Verkerk et al., 1991) and has since been connected to many conditions, for example, Friedreich's ataxia (Sakamoto, Ohshima, Montermini, Pandolfo & Wells, 2001). The concept of uniparental disomy, which is when both chromosomes in the pair have come from the same parent and the absence of the other parent's chromosome results in an overall chromosomal abnormality, has been implicated in several syndromes including Prader-Willi syndrome (Nicholls, Knoll, Butler, Karam & Lalande, 1989) and Angelman syndrome (Malcolm et al., 1991).

Laboratory studies testing genetic DNA (deoxyribonucleic acid) is another new development in molecular genetics that has revolutionised the way inherited conditions are diagnosed (BERIS, 2010). New molecular probes are used to test specific DNA sequences for abnormalities and thus identify the genetic etiology. Genetic testing of DNA in body tissues can then determine whether a person has that condition.

¹The term neuro-typical is recommended by The National Autistic Society (2011) to describe people without an autistic spectrum disorder whose neurological development is perceived by society as normal. It has also been extended to differentiate other developmental conditions such as ADHD.

As well as confirming a biological basis for existing syndromes, genetic technology also allows for the identification and elucidation of many new syndromes, for example, X linked syndromes (Muir, 2000). The percentage of unknown causes of IDD is reducing each year as more genetic links are being identified. Genetic causal links are also being found for IDD's classified as mild and moderate, which were traditionally thought to have an environmental cause (Gostason, Wahlstrom, Johannisson & Holmqvist, 1991; Matalainen, Airaksinen, Mononen, Launiala & Kaarianen, 1995; Rutter, Simonoff & Plomin, 1996). Also, *de novo* mutations, where neither parent possesses or transmits the mutation are being found to cause IDD (Vissers et al., 2010) and continued application of new technologies is expected to underscore a high proportion of causality of IDD for such mutations (Ellison et al., 2013; Veltman & Brunner, 2012).

1.3.2 Behavioural phenotypes and diagnosis. Behavioural phenotypes are syndromes with a known genetic etiology (Skuse, 2002) and relate to the pathways between the genotype and phenotype (Dykens et al., 2000). Dr. William Nyhan first used the term behavioural phenotype in his description of stereotypic hand wringing in people with Lesch-Nyhan syndrome (Nyhan, 1972). Since then the term has broadened to describe distinctive physical, behavioural and cognitive profiles of genetic syndromes (O'Brien, Barnard, Pearson & Rippon, 2002).

According to Flint and Yule (1994), to be considered phenotypic, a behaviour must occur in all cases of the syndrome, for example, hyperphagia in Prader-Willi syndrome. Dykens (1995) has suggested a more probabilistic view in that a behaviour reliably occurring in most cases of a syndrome can be considered part of the phenotype. These two views, termed total and partial specificity (Hodapp, 1997), have led to some confusion regarding the concept. Currently, the consensus among

professionals appears to centre on the latter viewpoint (Dykens et al., 2000) and the wealth of evidence indicating the variability of expression of phenotypic behaviours both between and within syndromes supports this view (Basile, Villa, Selicorni & Molteni, 2007; Hodapp, 1997; Levy & Ebstein, 2009; Lewis et al., 2006; Sinnema et al., 2011; Varela, Kok, Otto & Koiffmann, 2004).

The science of behavioural phenotyping has grown exponentially with the growth of the genetic technologies described in the previous section (Oliver & Hagerman, 2007) and better understanding of gene-behaviour associations can inform the work of professionals in IDD services. Behavioural phenotypes are information giving devices; they explain why a syndrome has occurred, how it will affect development and explain associated problematic behaviours. A review by Tunncliffe and Oliver (2011) showed that syndromic behaviour is influenced by the environment and that phenotypic behaviour can be subject to change via environmental adjustments. They argue that syndromic behaviour is operantly reinforced by the environment and that early intervention, including provision of information to services regarding the phenotype, can improve quality of life and challenge deterministic views of diagnoses. Understanding behavioural phenotypes also facilitates the development of tailored interventions (Courtenay, Soni, Strydom & Turk 2009). Two well documented examples of this are self-injurious behaviour in people with IDDs (Oliver & Richards, 2010) and early-onset dementia in people with Down syndrome (Kozma, 2008). For professionals working with people with IDDs, engaging with a known behavioural phenotype has the potential to inform the design and planning of interventions and thus genetic research may have significant potential to affect the lives of people with IDDs for the good.

People with IDD, their parents and wider family members welcome greater understanding of the genetics of the diagnosed behavioural syndrome (Costain, Chow, Ray & Bassett, 2012; Lenhard, Breitenbach, Ebert, Schindelbauer-Deutscher & Henn, 2005; Statham, Ponder, Richards, Hallowell & Raymond, 2010; Trottier et al., 2013). Participating in genetic research that affords opportunities to determine a diagnosis or to obtain a greater understanding of an existing diagnosis is also welcomed by parents (Statham et al., 2010; Trottier et al., 2013). Indeed, parent support groups are considered a key driving force in behavioural phenotyping research (Harris, 2010) and an important connecting link between the two previously described professional cultures in IDD (Finucane, Haas-Givler & Simon, 2003). A diagnosis is thought to result in greater understanding and perceived control by providing a probable trajectory for the condition and to facilitate access to services and better managed care (Klein-Tasman, Gallo, Phillips, Kristin & Fine, 2008; Lopez-Rangel et al., 2008; Costain et al, 2012; Trottier et al., 2013). A diagnosis of a behavioural syndrome can also help to alleviate parent guilt and for an adult with an IDD, a diagnosis can be validating of difficulties experienced due to the diagnosis (Costain et al, 2012; Trottier et al., 2013). There is also an altruistic element in receiving a diagnosis, in that people with IDDs and their families report a willingness to be involved in further research to increase understanding of and reduce any stigma related to the diagnosis (Costain et al, 2012; Trottier et al., 2013). This altruism is akin to Titmuss's (1970) "gift relationship" which suggests that people are motivated to enhance the public good for no personal gain.

However, behavioural phenotyping has signalled to some a re-emergence of the medical model of disability². Reflecting Hodapp and Dykens's (1994) two cultures, this perception affects the integration of research findings into clinical practice (Kuna, 2001). Many professional groups working with individuals with IDD, including clinical psychologists, adhere to and have been trained within a culture of a social or bio-psycho-social understanding of disability. The social model of disability³ also informs more recent service provision and policies for people with IDD (Valuing People 2001, 2009); therefore, integrating knowledge regarding behavioural phenotypes into such a culture faces many obstacles.

The relative rarity of individual genetic syndromes may make implementing specific phenotypic information cumbersome to staff trained in taking a more holistic view of the person. However, collectively, genetic syndromes affect an estimated half a million people in the UK (Oliver & Hagerman, 2007), thus knowledge about their phenotypic differences and commonalities should be

² The medical model of disability views disability as physically or biologically caused. In this view, if a diagnosis is untreatable by medical means it will stay fixed and unchangeable. Its focus is on treating a diagnosis to cure the disability.

³ Oliver (1981) coined the term "social model of disability" and described it as such:

"This new paradigm involves nothing more or less fundamental than a switch away from focusing on the physical limitations of particular individuals to the way the physical and social environment impose limitations upon certain categories of people" (Oliver, 1981: 28). According to Barnes (2013), Oliver's term reflected the shifting political, academic and general societal views about disability. These changing attitudes had been affected by several factors, such as social constructivist views, changing population demographics and human rights initiatives. In contrast to the medical model, the social model disentangles disability from a physical or biological impairment and deems it to have a social or environmental cause.

considered in services to facilitate person-centred planning. In their review of the value of genetic investigations for adults with IDD, Baker et al. (2012) outline several examples of how a diagnosis can positively affect health and behavioural outcomes. In addition, the importance that a diagnosis can have for people with IDD and their families, as reflected in the many syndrome specific support groups established by families, underpins a need for professionals to be knowledgeable and open to applying syndrome-specific information in clinical work.

1.3.3 Genetic screening and testing. A further way in which genetic advances affect people with IDD is through genetic screening and testing⁴. An outcome of the Human Genome Project is the development of genetic screens and tests for several physical diseases and IDD (BERIS, 2010). The use of such screens and tests have an impact on people with IDD in advance of birth if parents decide to use them, or indeed through routine use as in the case of a screen for Down syndrome which is used prenatally worldwide (Buckley & Buckley, 2008). Also, parents of children with IDD and adults with IDD can obtain a genetic test during their lives to help inform treatments and prognosis (Baker et al., 2012). A UK study found that public attitudes towards non-invasive genetic testing were generally positive when looking from the perspective of the individual but less so when

⁴ The terms genetic screening/screens and genetic testing/tests are used interchangeably in this thesis, however, it is important to note that there is a difference in when they are used. A genetic screen is used at a population level to identify individuals who may have, or be susceptible to, or be a carrier of a serious genetic anomaly, whereas a genetic test is used on case-by-case basis when there is pre-existing evidence (e.g. a family history) that a genetic anomaly may be present (Nuffield Council on Bioethics, 1993). An example of the former is the screening of all newborn children for phenylketonuria (PKU) and an example of the latter is testing for the Huntington's disease gene in individuals known to be at high risk of developing the condition.

looking from a public health perspective due to concern regarding possible eugenic practice (Kelly & Farrimond, 2012). Therefore, genetic testing can create a complicated bioethical discourse, particularly in relation to prenatal testing. A full discussion of this discourse is beyond the scope of this thesis (see Diesfeld, 1999, for a detailed bioethical literature regarding genetics and people with IDD); however, several salient points of the discourse and debate follow.

One view of genetic tests is that their aim to “cure” disability underscores an inherent devaluation of the lives of people with IDDs. Edwards (2003) argues that any justification for genetic screening programmes is dubious as it is based on an assumption that there is such a thing as a “good life”. Indeed research shows health care providers often view quality of life as synonymous with good health and thus a disabling condition equates to poor quality of life (Leplege & Hunt, 1997). For example, Ormond and colleagues (Ormond, Gill, Semik & Kirschner, 2003) found support for this among medical and genetic counselling trainees. This view shows the negative identity that can be ascribed to a person with an IDD. In relation to prenatal testing, Stainton (2003) argues that to terminate a pregnancy due to IDD is saying that this negative identity is correct and not questioned. Shakespeare (1998) contends that such social discrimination needs to be addressed via the education of the public and professionals regarding disability and highlights a need for genetic counselling to help parents make informed decisions about the social implications of having a child with an IDD rather than the medical facts. Another perspective on genetic research is that a focus on the search for genetic causes of IDDs is detrimental to the study of other contributing factors such as social, environmental, economic and political factors, which can serve to further discrimination (Gooding, 1996; Wilkinson, 1996). Arguments for the advantages of genetic screening include

the ability to make better-informed reproductive decisions, (Carmichael, 2003). In addition, as discussed in relation to behavioural phenotypes, search for a genetic cause is considered important due to genotype-phenotype interactions in determining behavioural and other outcomes (Thapar, Gottesman, Owen, O'Donovan & McGuffin, 1994).

1.3.4 Genetic advances: section summary. This section has outlined the advances in molecular genetics affecting people with IDD. The proposed advantages of genetic research in terms of consequent advances in the behavioural phenotyping and diagnostic genetic technologies have been outlined and arguments for and against their advancement highlighted. In the context of rapid genetic developments, resistance may be noted in professionals working in the area of IDD, who view this development as an unwelcome negative emphasis on a medical model of disability. The next section will outline the historical relationship of people with IDDs and the medical model of disability. It will show how cultural knowledge of this history may affect attitudes of those working with people with IDDs towards the “New Genetics.”

1.4 History of Intellectual and Developmental Disabilities

This section will describe the history of IDD and relate it to its relationship with the medical model of disability in the form of eugenic practice.

1.4.1 People with IDDs in history. Historical descriptions and characterisations of people with IDDs and actions towards them have reflected the prevailing attitudes of the time. The terminology IDD was preceded by terms such as “idiot”, “imbecile”, “feeble-minded” and “mental defective”, each considered appropriate in their epoch (Atkinson, Jackson & Walmsley, 1997). Throughout

history, stigmatising characterisations of people with IDD have been made, ranging from the contemptuous “less than human” to the more benign “eternal children” and “holy innocents” (Wolfensberger, 1972). Given these descriptions and characterisations, the actions towards people with IDDs by society have at varying times and in varying ways served to segregate them from wider society, undermine their human rights and mask their individuality. For people with IDDs, this has meant a continual challenge to fight against the stigma created by *social constructions* of disability.

From the 1970’s onwards, the social construction of IDD has evolved alongside changing socioeconomic and political systems. Currently people with IDDs are recognised and valued for themselves and societal structures reflect this (UNESCO, 1994; Valuing People 2001, 2009). Society is now adhering to the concept of inclusion, whereby people with IDDs are respected, given opportunities and above all, are not excluded (Culham & Nind, 2003). The origin of this current view has its roots in the 1960’s Disability Rights Movement (Hirst, 2000). Since this time people with IDDs have experienced several changes that have positively affected their lives, namely, the introduction of the principles of normalisation (Bank-Mikkelsen, 1969), the process of deinstitutionalisation (Beadle-Brown, Mansell & Kozma, 2007) and legally sanctioned access to education, for example, the passing of the Education (Handicapped Children) Act, 1970 in the United Kingdom.

Prior to this people with IDDs lived through a much darker time influenced by the pseudoscience of eugenics. To understand how this pseudoscience became popular, its origins in history, through pertinent societal changes, will first be traced.

1.4.2 Societal changes and the impact on people with IDD. Called the Age of Reason or Enlightenment, the 18th century was characterised as a period of great scientific thought and development. Prior to this, religious teachings had influenced social and political life and within this discourse people with IDDs were generally seen as innocently benign and allowed to live as part of society and hence the “disability” of intellectual disability was almost invisible in society. People with IDDs either had place in their home, were allowed to drift between villages or found refuge in the existent institutions such as lunatic asylums (Atkinson et al., 1997). In the 18th and 19th centuries, the development of scientific thinking and a search for rational explanations for phenomena led to more emphasis on medical explanations of IDD and less on divine explanations (Brigham, 2000). In 1866, John Langdon Down described the physical characteristics of what later became known as Down syndrome, an early example of an attempt to classify an intellectual disability, the medical cause of which would not be determined for almost another century. It was at this time also that people with IDDs were recognised as different to people with mental illness and terms such as “idiot” and “imbecile” came into use. Despite their derogatory meaning today, these terms had a medical and legal meaning when first introduced (Atkinson et al., 1997). Another facet of scientific development that affected people with IDDs was the Industrial Revolution (Barnes, 1991). With the increased industrialisation came changes in work structures with mass movement away from the land to factory settings. In this environment, people with IDDs were not accommodated and increasingly they were seen as a burden and a problem. Changing times meant a changing attitude.

Through these changes, people with IDDs became associated with social ills such as crime, immoral behaviour and rising costs of schools, hospitals and prisons,

which engendered further stigma (Bachrach, 2004; Kliwer & Drake, 1998). The State began to see a responsibility to support citizens to overcome external barriers to self-improvement and development of their moral character and as a result of this, the perceived threat of people with IDD was progressively addressed via legislation and eugenic policies (Stainton, 2000). This led to the development of classification systems for people with IDD and their increased social segregation (Brigham, 2000). The 1886 Idiots Act made the legal distinction between “idiots” and “imbeciles”. The start of the 20th century also witnessed continual pressure in parliament to pass legislation to control the perceived threat of people with IDD, for example, the work of Mary Dendy (Cruickshank, 1976). Through these legal developments, institutional care for people with IDD was initiated. In 1913, the Mental Deficiency Act was passed with little opposition with the aim of providing institutional care for the “feeble-minded” and “moral defectives” (Gilbert, 2009, para. 27). It could be argued that institutionalisation of people with IDD had begun as far back as the Middle Ages, for example, Foucault (1964) observed that people with IDD and “mad” people were confined to Poor Law institutions previously housed by the ill and infirm. However, such institutionalisation did not have a compulsory and statutory requirement. The new, early twentieth century institutions, designed for the care and management of “idiots”, “moral imbeciles and “feeble-minded persons”, ultimately served to separate people with IDD from society. The segregation of males and females within institutions was also common with the intention of reducing reproduction by people with IDD. The 1913 Mental Deficiency Act remained on the statute book until it was replaced in 1959 by the Mental Health Act.

1.4.3 History of IDD: section summary. It was during the social climate of the late 19th and early 20th century that the eugenics movement was born of scientific developments and raised on the prevailing attitudes towards people with IDD in a seemingly mutual relationship. One perhaps could not have thrived without the other. Miller and Levine (2013) describe the “historic trauma” of people with IDD as being rooted in eugenics. The next section will outline and discuss the development of eugenics and its impact on the lives of people with IDD. Understanding this perspective may in part explain contemporary attitudes to current genetic advances and the caution towards them.

1.5 History of Eugenics

In 1859, Charles Darwin published “*Origin of the Species.*” Expounding the theory of natural selection as the key mechanism of evolution, Darwin’s theory was to have a profound impact on the scientific world and beyond. In terms of people with IDD, this impact began with Darwin’s cousin, Sir Francis Galton. A man of notable influence in the scientific community himself, Galton was inspired by Darwin’s theory and in particular in its application to the selective breeding of animals. Transferring this reasoning to humans, Galton considered society’s morals, which sought to protect the weak and vulnerable, as an interference with human betterment through the process of evolution by natural selection. He promoted selective breeding of the genetically “superior” to better the human race, or positive eugenics, as it later became known. In 1883 Galton coined the term “eugenics” from the Greek, *eugenes*, meaning good in stock to describe this burgeoning ‘science’ and defined it as:

“the study of the agencies under social control that may improve or impair the racial qualities of future generations, either physically or mentally.” Forrest (1974).

Eugenics thus implied that some people’s genetic make-up was inferior causing them to be a reproductive threat, weakening the gene pool.

Understanding human evolution as a process of non-random selection of adaptive traits began to influence and shape the thinking towards the causes of difference among people, specifically the differences seen in people with IDD. This emerging philosophy, or Social Darwinism as it became known, caused people to consider whether care for “mental defectives” was warranted as it was seen to defy the “law” of natural selection causing a degeneration of society. As well as being seen as a burden on society, people with IDDs were also viewed as a threat to social progress.

As discussed previously, social changes were highly conducive to the adoption of eugenic ideas and Galton’s teachings were hugely influential on the development of eugenics movements in both Europe and America. Whilst Europe’s evolving industrialisation influenced views of IDD, post-Civil War America was managing the pressure of immigration and the onset of an economic depression in 1873. In this climate people with IDDs were viewed as a hindrance and again, segregation via institutionalisation was seen as a solution to this particular problem (Kliewer & Drake, 1998). Earlier attempts by French immigrant, Edouard Séguin, to educate people with IDDs by emphasising the importance of developing skills of independence and self-reliance (Blatt, 1987), were subsumed beneath the eugenics movement and such positive initiatives largely remained dormant throughout the eugenic era.

The popularity of eugenics led to the creation of several international eugenic societies whose purposes were to promote eugenic ideas. In Britain in 1907, supporters of eugenics formed the Eugenics Education Society, now called the Galton Institute since 1989. The Society aimed to promote the biological improvement of the nation by mitigating against the reproductive influence of those considered genetically unfit. Social organisations campaigned for segregation and sterilisation and marriage restrictions for people with IDD to prevent the “degeneration” of the population. These societies attracted support from many influential figures, among them many left-wing thinkers such as George Bernard Shaw, William Beveridge and Bertrand Russell. In 1912, the first International Eugenics Conference was organised by the British Eugenics Education Society with 400 delegates in attendance including Winston Churchill. With this type of support, it is not difficult to understand how the eugenics movement was able to influence the politics of the day. This statement by Henry Goddard, Director of the Research Laboratory of the Training School at Vineland, New Jersey, for Feeble-minded Girls and Boys, (as cited in Kliewer & Drake, 1998) sums up how popular the movement was:

“The large share of attention which has been given to the new science of eugenics, or race betterment, shows conclusively that society is intensely interested in this problem of the improvement of the race...The feeble-minded person is not desirable, he [sic] is a social encumbrance, often a burden to himself. In short it were better both for him and for society had he never been born. Should we not then, in our attempt to improve the race, begin by preventing the birth of more feeble-minded? (Goddard, 1914, p. 558.)”

Eugenic practices are described as *positive* eugenic practices, when reproduction by people with “good genes” is promoted and as *negative* eugenic practices when reproduction by people with “bad genes” is prevented (Shakespeare, 1998). Galton’s original aim was to develop positive eugenic practices through selective breeding. History, however, has shown how the popularity of eugenic principles led to the widespread adoption of more negative eugenic practices, with the segregation, sterilisation and in Nazi Germany, murder of people with IDD in an attempt to eradicate their “disability” (275,000 people with disabilities were murdered in the Holocaust, United States Holocaust Memorial Museum, n.d.).

1.5.1 Legacy of eugenics. Eugenics shrouded itself in scientific terms to justify its actions, which were ultimately based on the moral reasoning and societal attitudes of the time (Kevles, 1995). Kliewer and Drake (1998) argue that a legacy of the eugenics movement is so-called “scientism” being used to hide an ideology of segregated control of people with IDDs. Although attitudes towards disability have changed, history has imbued the application of genetic science to IDD with a degree of mistrust. Shakespeare (1998) has described the capabilities of the “New Genetics” as “weak eugenics” in that new technologies such as prenatal screens influence the reproductive choices of the individual. This is opposed to “strong eugenics” of the past where population-wide strategies attempted to eliminate disability. Shakespeare (1998) described the motivation of the latter as social stigma surrounding IDD whereas a medical judgement that people with IDDs have poorer quality lives motivates the former. Although many may disagree with Shakespeare’s view, it does highlight the argument described elsewhere in this thesis regarding the implicit motivation of the medical model to “cure” disability. It seems that whatever guise this “cure” takes, some people may always perceive a eugenic undercurrent.

Shakespeare (1998) suggested that an alternative to curing the faulty gene is to cure the faulty society that discriminates against the disability and in doing so making life with a genetic difference easier to live. This view is what underpins the social model of disability.

Hence, the on-going legacy of the eugenics movement may mean that scepticism may always permeate genetic advances. Smith (1994) suggested that following the eugenic era professionals working with people with IDD's sought to create distance between their work and eugenic practices, which ultimately led to a caution towards the medical model and genetic services. This legacy may serve a protective function in acting as a watchdog over professional practices, but it may also serve to thwart potentially beneficial advances for people with IDD's and their families. The focus of this thesis is that the weight of this historical knowledge currently influences the attitudes of clinical psychologists towards applications of modern genetic developments. In their role, they may consider themselves to have a responsibility to monitor and question genetic developments that may in turn act as a barrier to their application of them. The following section examines more closely the profession of clinical psychology and its history with IDD.

1.6 Clinical Psychology and Intellectual and Developmental Disabilities

This section will trace the history of the profession of clinical psychology in relation to people with IDD's. It will argue that the work of clinical psychologists in IDD has most successfully been associated with a social model of disability. It will show how initially their relationship grew through an association with psychiatry. This association may make embracing a medical model more complicated. Current cohorts of clinical psychologists have trained and worked through the culture of the

social model of disability. As well as the legacy of eugenics, clinical psychologists professional identify will influence their attitudes towards genetics. This section considers factors that have shaped the profession and its working relationship with people with IDD. A brief history of clinical psychology's origins is included to place this in a context, followed by a description of clinical psychologists working history with people with IDDs.

1.6.1 Origin of clinical psychology. Clinical psychology as a professional discipline is relatively new with its notional origin towards the end of the 19th century when Lightner Witmer established the first psychology clinic in the US in 1896 and coined the term "clinical psychology" as "the study of individuals, by observation or experimentation, with the intention of promoting change" (Compas & Gotlib, 2002).

Witmer was the first to attempt to apply psychology to promote change, but such interventions were overshadowed at that time by the dominance of assessment methods in the field. James McKeen Cattell, a former mentor of Witmer at the University of Pennsylvania had developed the first "mental test" of intelligence based on the work of Galton (Benjamin, 2005). Although the test floundered due to a lack of association between its results and scholastic attainment (as assessed by the newly developed correlation statistic, Sokal, 1982), newer, more valid intelligence tests soon emerged.

The emphasis on assessment in clinical psychology at this time was no happenstance. Like anything else, the discipline did not evolve in a vacuum and was influenced by the wider context. Psychiatry was the dominant profession in the field of mental health and was reluctant to share this platform with other professions and treatment of the severely mentally ill or any other clinical populations was not a

domain clinical psychologists were welcomed in, however, nor was this of prominent interest to those in this newly developing field (Benjamin, 2005). Clinical psychologists were more interested in individual differences and behavioural phenomena. Influenced by Darwinism and statistics, psychologists armed themselves with instruments purporting to measure individual differences (Benjamin, 2005).

Differences in human intellectual ability were the first phenomena of interest, again, influenced by social pressures at the time with policy makers being in need of science to justify their concerns about the social and moral danger of people considered of inferior intellect. Witmer, the person credited with establishing clinical psychology as a discipline was vehemently opposed to intelligence testing, as he believed all children were capable of learning (Thomas, 2009) but such testing was the bedrock upon which the discipline developed.

It could be said that clinical psychology, as a profession, gained prominence through its work in assessment of IDD. Clinical psychologists gained further prominence through intellectual and personality assessments in World Wars I and II and between the wars clinical psychology firmly established a foothold in the United States through subsequent work in child development (Benjamin, 2005).

1.6.2 Clinical psychology and IDD. As clinical psychology developed as a profession in the 20th century, its focus began to shift towards its aim of promoting behavioural change. Within psychology behaviourism developed as a major school of thought. Behaviourism, concerned with how external, observable behaviour could be shaped by the environment (Skinner, 1984), represented a separation of psychology from psychiatry. Psychological assessment instruments had been developed in conjunction with a medical model of innate fixed difference, but

behaviourism was rooted in environmental and social influences and hence was concerned with learning and change rather than a deterministic disease model.

After the culmination of the original eugenic movement in the Nazi Holocaust, the change of focus to behavioural approaches was to have a welcome impact on people with IDD. Just as Séguin had attempted to show the educability of people with IDD a century before and as Witmer had asserted that all children were capable of learning, the behaviourists intended to demonstrate behavioural modification through reinforcement and operant conditioning. In 1949, Fuller conducted the first behavioural research with a person with an IDD, "*Operant conditioning of a vegetative human organism*," which showed that a person with severe IDD was capable of learning. By the 1960s there was a proliferation of behaviourist approaches. The principles of Applied Behaviour Analysis with people with IDD developed and were promoted by prominent psychologists such as Jack Tizard (Kappel, Dufresne & Mayer, 2012). This culminated in 1965 with the publication of Ullman and Krasner's (1965) "*Case Studies in Behaviour Modification*." The Journal of Applied Behavior Analysis (ABA) was created in 1968 to provide professionals with access to the research being conducted (Society for the Experimental Analysis of Behavior, 2013). Other notable behavioural works in the UK included the work of the Hester Adrian Research Centre in Manchester, which aimed to "*promote, sustain and carry through research into the learning processes in mentally handicapped children and adults*" (Pulan & Abendstern, 2004, p. 34). Using behavioural techniques proved effective in changing the quality of life for people with IDD and was in direct conflict to the medical model, whose focus on an underlying biological cause was considered counterproductive (Ullmann & Krasner, 1965).

By 1970 ABA was deemed an appropriate treatment to improve social behaviours and psychologists were considered to be working unethically if they did not adhere to behaviourist principles (Ullman & Krasner, 1965). However, there was growing criticism of punitive behaviourist approaches such as restriction, physical interventions and electric shock treatment. These criticisms occurred in the context of social changes affecting people with IDD, in particular the normalisation movement (Nirje, 1970; Wolfensberger, 1972) and the emergence of the social model of disability (Oliver, 1981). The main principle of normalisation was that the environment should change, not the person, which was regarded as conflicting with the aims of behavioural modification (McGill & Emerson, 1992). Proponents of normalisation argued that behavioural interventions that artificially manipulated contingencies did not take into account how people with IDD are affected by social attitudes and imposed norms (Remington, 1998). The ideology of the social model of disability, whereby disability is viewed as being caused by social environments that do not accommodate people's impairments, also underscored a need for psychologists to take account of how their behavioural approaches were being used. Thus, normalisation and the social model of disability called into question and highlighted limitations of ABA interventions for people with IDD (Wolfensberger, 1989).

Clinical psychologists, many of whom had grown disgruntled with traditional behaviour modification approaches, have been receptive to these criticisms and since the 1980s psychologists working with people with IDD have evolved and developed positive behavioural approaches using the principles of inclusion. Such approaches serve to determine the idiosyncratic communicative functions of behaviours for individuals and adapt environments to better meet their needs.

1.6.3. Clinical psychology and IDD: section summary. This section has described clinical psychology's history of working with people with IDD. It has shown how the profession gained a foothold through its success with assessments of intellectual functioning that initially served to further the segregation of people with IDD. It has further shown how, via subsequent application of behaviourism, clinical psychologists developed a more positive relationship with people with IDD and are currently perhaps a professional group that has earned the trust and respect of this group of people. Clinical psychology in the last century progressed from its origins in its relationship with psychiatry in the medical model to a more defined professional identity that has advanced alongside a social model of disability. Understanding clinical psychologists' current position underlines the professional issues they may have in returning to more biologically based models via application of genetic research.

If genetic developments are to be effectively applied they need the support of clinical professionals. Clinical psychologists are a key professional group in services for people with IDD and are well placed to ensure the promotion and application of relevant genetic advances. Gaining insight into attitudes they hold on this subject is an important step towards understanding the current state of Dykens and Hodapp's "two culture" view of professionals in the IDD field.

The next section will describe Q-methodology, the methodology used in this study to explore clinical psychologists' attitudes to the "New Genetics." A rationale for the use of this method is also detailed followed by a description of the study aims.

1.7 Q methodology

Q methodology is the systematic study of subjective opinions, attitudes and beliefs (Stephenson, 1935; Brown, 1993; McKeown & Thomas, 1988). The initial stage of a Q methodology study is to define the topic of interest and gather the existent views held about it; in Q terminology this is termed the Q-concourse. The Q-concourse then forms the Q-set or Q-sample, which are a list of statements representing all the varying viewpoints. Participants whose views are sought regarding the topic of interest (the P-set or P-sample) are asked to sort the statements according to a specific condition of instruction in a process called Q-sorting. A Q-sort entails the participant sorting and ranking the statements according to their level of agreement or disagreement with them (Brown, 1996). The individual participant Q-sorts are correlated and factor analysed and the emerging factors interpreted to understand the overall group's views as well as their similarities and differences (van Exel & de Graaf, 2005).

1.7.1 Background of Q methodology. William Stephenson, a British physicist-psychologist, introduced Q methodology in 1935. Stephenson developed the method in response to limitations he saw in the traditional factor analytic research in psychology, which he called R methodology (Brown, 1996). He argued that in R methodology, predefined categories were imposed on participants via tests and measures rather than obtaining participants' subjective viewpoints. Stephenson disagreed with this as a measure of subjectivity as he believed this was removed when the participant was reduced to traits and characteristics (Robbins & Krueger, 2000). He also disagreed with the prevailing view among psychometricians that subjective opinion represented pure mental experience and was therefore immeasurable (Robbins & Krueger, 2000). In contrast, Stephenson viewed

subjectivity as the internal frame of reference one calls upon to make sense of the world around them i.e. a person's behaviour makes apparent their internal frame of reference. According to Stephenson, R methods remove the internal frame of reference to measure opinion and thus do not uncover subjectivity. Q methodology, however, permits people to give self-referent accounts by looking at life from the standpoint of the person living it (Brown, 1996). Through the process of Q-sorting a participant's subjective viewpoint or personal profile emerges (Smith, 2001; Brouwer, 1999). McKeown and Thomas (1988) have further described subjectivity as being operant in that categories are not imposed on participants but are instead imposed by participants through the structure they give to their data in the Q-sort.

To study his interpretation of subjectivity, Stephenson used an inverted R methodology by correlating persons instead of tests. Instead of measuring people with tests he measured tests (Q-sample) with people (P-set). Q methodology was met with some hostility when first described by Stephenson. Positivist views were central to psychology and leading psychometricians, Burt and Cattell, criticised the method as a return to a less objective, introspective science (Kitzinger, 1986). Stephenson's insistence that subjectivity was measurable and communicable was in conflict with their view that only observable, objective behaviour was of relevance to scientific query. Burt and Stephenson in their 1939 paper, "*Alternative views on Correlations Between Persons,*" outline the similarities and points of departure between their views on the theoretical implications of correlating persons rather than tests in factor analysis. This paper highlights Stephenson's dedicated focus on using Q methodology to systematically obtain and understand subjectivity in all types of situations. Despite continued disagreements about the use of the method, Stephenson's theory persuaded new generations of scientists who would go on to

further develop the field and Q methodology is now increasingly used in psychology (Combes, Hardy & Buchan, 2004; Dick, Gleeson, Johnstone & Weston, 2010; Eccleston, De Williams & Stainton Rogers, 1997; Shinebourne & Adams, 2007; Stunner & Marshall, 1995; Walker, 2009; Westbrook, McIntosh, Sheldrick, Surr & Hare, 2012).

1.7.2 Characteristics of Q methodology. To understand Q methodology, Cordingley, Webb and Hillier (1997) suggest you need to understand how it is different to normative R methodology. In R methodology the measure or scale is considered to access the respondent's view objectively. However, the respondent is confined by the strictures of the measure and the view they give can only be a view contained in the measure (Brown, 1980). The assumptions of the researcher cannot be separated from the measures they use. In Q methodology the respondent's Q-sort can contain any configuration and the view contained therein represents their subjectivity.

Another difference between Q and R is that in the latter meaning is applied a priori to the categories of interest; however, in the former the researcher can make no assumptions about how the Q-sorts will be configured (Stainton Rogers, 1995), as it is the participants who impose structure on data and not the researcher (Kitzinger & Stainton Rogers, 1985). Meaning is thus only attributed to items through the act of doing a Q-sort (McKeown & Thomas, 1988).

1.7.3 Rationale for use of Q methodology. When little is known about a topic an exploratory research method is indicated. The topic of this study, clinical psychologists' attitudes to research developments in genetics is little understood with no previous research available. Q methodology is recommended for exploratory research and research that seeks to understand subjectivity (Thomas & Watson,

2002; Watts & Stenner, 2005; Wigger & Mrtek, 1994). The decision to use Q methodology over more traditional exploratory research approaches such as interviews and subsequent qualitative analysis was based on the advantages of the methodology that were deemed to better suit the research aim of obtaining the viewpoints of clinical psychologists regarding genetic and associated research affecting people with IDD. This study expects different, varying views to exist on the topic and it was important to use a method that allows for this variability. Through the Q-sort process this variability in attitude is allowed for and the method is sensitive to differences in views too.

Although Q methodology could be described as being less naturalistic than an interview (Stenner, Watts & Worrell, 2008), the use of pre-generated statements and comparisons between individual Q-sorts enables the general configuration and structure of views on the topic to emerge. More participants and hence views can be included in Q studies (Smith, 2001) than would likely be feasible in an interview design. This is advantageous when little is known on the topic and there is an interest in obtaining several perspectives (Stainton Rogers, 1995). In addition, Q methodology often provokes thinking about topics that would not necessarily have been introduced in an interview. This thinking was of interest as the study topic may not have been of manifest concern to the participant prior to the process, for example, participants may have never considered what they thought about behavioural phenotyping. Participating in interviews is usually undertaken by participants to whom the interview topic is already of intrinsic interest. When a topic is of a potentially controversial nature, Q methodology lets participants communicate their perspective in a confidential manner (McKeown & Thomas, 1988). Genetic research and its application, as has been documented in this chapter, is controversial

and attitudes towards it need to be studied with care. Clinical psychologists may be working in services that may have a contradictory ethos to their own views and may be hesitant to express the latter in an interview.

Cross (2005) suggests that Q methodology is a more robust measure than alternative methods for the study of attitudes such as Likert scales and semantic differentials. She refers to identified advantages of the method such as fewer uncertain responses and missing data (Dennis, 1986) and more focus on participant subjectivity than on estimating population statistics (Kitzinger, 1987). It is also argued (Donner, 2001; Stenner et al., 2008) that the active role taken by the participant in Q-sorting means that they have the freedom to understand the statements from their subjective, “gestalt” point of view rather than be confined by a scale or measure, statements are sorted in relation to all other statements. This aspect was important in the current study in order to understand the relative importance or emphasis clinical psychologists place on aspects of genetic advances. Q methodology has been used to successfully assess staff attitudes (Dick et al., 2010; Wastell, 2010; Westbrook et al., 2012). It has also been recommended by the National Institute of Health and Clinical Excellence (NICE, 2004) to assess staff attitudes in relation to certain topics.

Q methodology is also suitable for constructivist accounts of social and natural reality (Robbins & Krueger, 2000), as no categories are imposed that may distort reality and a priori assumptions are not made (Brown, 2002). The focus in Q methodology is on different views rather than extreme views, which makes it an optimum approach in exploring uniquely developed attitudes. Furthermore, as this approach allows for the detection of multiple viewpoints, it can identify alternative or less dominant perspectives (Farrimond, Joffe & Stenner, 2010). Ensuring the

discovery of all views was again of importance in the current study as only anecdotal knowledge of clinical psychologists views on the topic previously existed.

The rationale for the use of Q methodology in this study is thus that it appears to have more strengths in being able to answer the research question than other comparable methodologies.

1.8 Purpose of the Current Study

The application of potentially beneficial genetic research in clinical and health services for people with IDD is poorly understood. It is argued that the research is too far removed from the services and that many clinicians and their model of training, the social model of disability, is in conflict with the medical model in which this research originates. As well as professional influences, broader historical influences in relation to people with IDD may also impinge on the application of genetic research. This research study explores clinical psychologists' attitudes towards genetic influences on people with IDD. It also considers factors that influence these attitudes. This study will further understanding of how well genetic research is welcomed and applied in services by clinical psychologists. In doing so, it will also highlight factors which either enhance or act as barriers to their application.

Therefore, the main aim of the present study is to understand clinical psychologists' views of research in the "New Genetics" that has clinical implications for people with IDD (Finucane et al, 2003). Exploratory analysis through Q methodology will be conducted to meet the aims of the study.

Chapter 2. Method

2.1 Method Overview

This method section outlines all the steps of the Q methodology process from development of a Q-set reflective of the study topic, to the recruitment of a P-set and the collection and analysis of participant Q-sorts. It begins by detailing the study's progression from ethical approval onwards.

2.2 Ethical Approval

A study proposal was peer-reviewed by the Division of Clinical Psychology Research Sub-Committee at the University of Manchester. An application (ref. ethics/12307) to obtain ethical approval for the study was then submitted to an ethics committee at the University of Manchester. The study was granted ethical approval on the 20th December 2012.

2.3 Study Design

The study used a Q methodology design. The rationale for using this method to explore clinical psychologists' attitudes to aspects of the "New Genetics" is outlined in the previous chapter. Q methodology reveals subjectivity through the structured process of Q-sorting. Through this process, participants give meaning and structure to the data (Brown, 1980). Interpretation of this structure is made through factor analytic techniques, which assume proper Q-study design and Q-sorting administration, and aims at elucidation of patterns of responses among the participants (Thomas & Watson, 2002). This methodology entails several stages and the next sections of this chapter will outline these in relation to the current study.

2.4 Exploration of the Q-concourse

The Q-concourse or “communication concourse” is described as “the flow of communicability surrounding any topic” in “the ordinary conversation, commentary, and discourse of everyday life” (Brown, 1993). The discourse includes opinions, beliefs and attitudes about the topic under review. According to Brown (1993), the level of discourse dictates the sophistication of the Q-concourse. The Q-concourse gives life to the Q-set or Q-sample, which contains statements reflective of the concourse.

In accessing the discourse, several strategies may be applied, such as interviews with relevant stakeholders or extraction of statements from existing sources, for example, items from standardised scales (Brown, 1996; Dennis, 1986; Wigger & Mrtek, 1994). McKeown and Thomas (1988) describe the former as an example of a “naturalistic” method and the latter as an example of a “ready-made” method. A combination or “hybrid” of these methods is also described by McKeown and Thomas (1988) who argue that the selection of methods used depends on what best suits the research at hand. Directly obtaining viewpoints from relevant stakeholders has the inherent advantage of making sure the meanings of stakeholder viewpoints are accurately understood while also mirroring the viewpoints of similar others completing the Q-sorts; however, extensive interviewing may not be practically feasible and in such cases use of other methods is warranted (McKeown & Thomas, 1988). Although it is acknowledged that the full concourse may never be known, Stainton Rogers (1995) emphasised the need to carefully review the written and verbal discourse around the topic.

In this study, the Q-concourse was developed using a hybrid method of interviews and statements drawn from academic literature including books and journal articles. A similar method has been used by others (Stainton Rogers, 1991).

2.4.1 Themes from the literature. A review of books proffering opinions, ideas and research findings relating to behavioural genetics and phenotyping, genetic screening and books relating these themes to intellectual and developmental disabilities were reviewed. All books were written in the past 30 years with the majority being written in the past ten years. Journals pertaining to developmental and intellectual disabilities were also reviewed to identify studies that reflected the following themes: the impact of genetics on development; the impact of the environment on development; the use of genetic screening or testing; the impact of receiving a behavioural diagnosis; the use of behavioural phenotyping, service developments in IDD services. The researcher (C. V.) extracted material from these sources that could be construed as opinion statements about research findings and ideas on the identified themes. A list of sources accessed can be viewed in Appendix 1.

2.4.2 Interviews. Four informal interviews with experienced clinical psychologists working in services for people with IDDs and/or researchers working in the IDD field were conducted. The interviews discussed the aforementioned themes. In an effort to obtain non-leading responses these themes were introduced to the interviewee in an open-ended style, for example, “What are your thoughts and views on genetic screening?” The researcher (C. V.) made notes during the course of the interviews to aid the task of identifying relevant opinion statements.

The interviewees (2 female, 2 male) were selected due to their extensive experience of working with people with IDD. Three worked clinically in services for people with IDDs and were each affiliated with a clinical psychology university training programme. One of these participants (male) was also involved in active research with people with IDDs. The fourth interviewee (female) was affiliated with a university and conducted research in the IDD field. After accepting an invitation to participate, interviewees received, by email, a participant information sheet and a study consent form that they signed and returned to the researcher (C. V.) prior to their interview. Appendix 2 contains the study information sheet and consent form for the interview participants. Three of the interviews took place at the interviewees' places of work. Due to distance one interview was conducted by telephone. The interviews took between 30-45 minutes to complete.

2.5 Development of the Q-set

After the discourse was deemed exhausted a Q-set reflecting all the viewpoints on the topic was developed. In developing a Q-set it is imperative that a broad range of items is selected in order that all viewpoints on the topic can emerge in the Q-sorts and a relative preference can be indicated (Barbosa, Willoughby, Rosenberg & Mrtek, 1998; Brouwer, 1999; Cordingley et al., 1997). McKeown and Thomas (1988) outline two techniques for the selection of statements, structured and unstructured sampling. Structured sampling takes into account theoretical considerations and ensures that statements are included to address all relevant hypotheses. Unstructured sampling entails selecting items presumed to be of relevance to the topic due to their emergence in the discourse; however, it is less concerned with addressing every sub topic. This method is considered more

appropriate for researchers assuming a social constructionist approach (Cordingley et al., 1997). Due to the exploratory nature of the study unstructured sampling of statements was used.

Van Exel and de Graaf (2005) state that it is the responsibility of the researcher to draw a representative sample from the concourse. The researcher (C. V.) and a study supervisor (D. J. H.) continuously reviewed the extracted statements under six emerging themes to ensure the representation of all viewpoints. These themes were given the following names: Intelligence/intellectual functioning; Genes (nature) versus environment (nurture); Diagnosis of IDD's arguments for and against; Diagnosis and service provision/intervention for people with IDD's; Services and implementation of research; Socio-political influences on spread of genetics research.

When a final list of 124 statements had been extracted they were subject to a final review. Each statement was printed onto cards and laid out under their initial themes. The researcher (C. V.) and her supervisor (D. J. H.) reviewed each of the statements and the themes and deemed them appropriate. Following suggestions by Donner (2001), statements were selected that had distinctive meanings and were varied enough to avoid overt repetition. Statements were also selected according to Donner's (2001) recommendation that statements are plausible competitors with one another, meaning that some participants may find them of interest whereas others may not be inclined to choose them. Statements were also phrased in a similar style to enhance the cohesion of the Q-set (Donner, 2001). This process resulted in several statements being removed or amalgamated with similar others and the final Q-set consisted of 81 statements grouped under the six broad themes described previously. This was in accordance with a recommendation that a Q-set should

contain between 40 and 90 statements related to the research topic (Dennis, 1986; Kerlinger, 1986).

2.5.1 Pilot of the Q-set. Many researchers recommend conducting a pilot phase in Q methodology. Stainton Rogers (1991) suggested that an important function of this process is that it allows consideration of whether a variety of viewpoints have been obtained.

The 81 statements were piloted with four third year trainee clinical psychologists (3 female, 1 male) from two university training programmes in the north west of England. The pilot participants were independent of the study, had varying degrees of knowledge of Q methodology and had completed a clinical psychology training placement in a service for people with IDD's not more than 12 months previous.

The pilot study's purpose was to assess both the statements and the Q-sort process. Specifically the pilot study aimed to assess the readability and clarity of the statements and identify any ambiguity; to gain feedback about the statements' ability to reflect the study area and identify any omitted relevant areas; to ensure that the statements were balanced across the "*agree*", "*disagree*" and "*neutral*" categories; to assess the time taken to complete the Q-sorting process and to ensure that the verbal instructions and study materials were clear and sufficient.

Following the pilot, the 81 statements were retained (Appendix 3); however, 16 statements were re-worded or re-phrased due to ambiguity, lack of clarity and potential for misunderstanding or confusion. Certain terms used frequently throughout the Q-set were identified by participants as being open to misinterpretation or different interpretations. To account for this it was considered

appropriate to include a written explanation of terms (Appendix 4) for participants to read prior to their Q-sort.

The final Q-set was considered to have captured important elements of the concourse though by no means was exhaustive of it. The number of statements was comparable to other Q methodology studies that allowed for meaningful statistical results to be generated (Stainton Rodgers, 1995; Walker, 2009).

In terms of practicalities, it took an average of 45 minutes for the pilot participants to complete the Q-sorting process. The verbal instructions were considered sufficient; however, to ensure that standardised instructions were used with the study participants the researcher (C. V.) drafted a narrative instruction script (Appendix 5).

2.5.2 Personal reflection on the development of the Q-set. In developing the Q-set, the study researcher (C. V.) endeavoured to be aware of factors that might affect her focus and selection of statement items. Factors such as the researcher's (C. V.) varying personal interest in aspects of the topic, professional clinical experiences, including teaching experiences as well as broader moral and ethical beliefs were all brought to bear on this aspect of the research. To reduce any potential bias related to these factors the researcher (C. V.) openly shared and discussed the Q-concourse with others, including a study supervisor and colleagues on her clinical psychology training programme. The researcher (C. V.) also obtained feedback on statement selections to achieve a balanced Q-set. Time was also used to give perspective to the process, whereby statements and themes were viewed afresh several weeks after their initial selection. The researcher (C. V.) reflected however that due to the idiosyncratic, unstructured nature of the process it was impossible to conclude that personal experiences were not implicated in the development of the Q-

set. That being said, it is thought that the open way in which the Q-set was developed resulted in a sufficiently broad range of views on the topic area being reflected and that although it is also unlikely that another researcher's work would result in the exact same Q-set, it is likely the themes surrounding them would be similar in nature.

2.6 Development of the Q-sort Task Materials

The Q-sort materials consisted of the 81 statement Q-set, a score sheet based on a quasi-normal distribution grid (Figure 1), a guide bar indicating the “*agree*”, “*disagree*” and “*neutral*” categories (Appendix 6), the narrative instruction script and the written explanation of terms.

2.6.1 The Q-set. The statements in the Q-set were randomly assigned a number between 1 and 81. Each statement was then printed on white paper that was then laminated and the statements cut into 100mm x 20mm laminated cards. The number of each statement was printed on the back of the card prior to laminating.

2.6.2 Guide bar. The guide bar was included to help participants structure the sort according to the pre-determined quasi-normal distribution required.

2.6.3 Score sheet. Q-sort ranking usually follows a normal distribution with fewer statements allowed in the extremes and the majority towards the centre (Dennis, 1986; Prasad, 2001). This forced-choice method is commonly used and recommended in Q studies (Brown, 1980). There are usually an odd number of columns in the distribution so that a central, neutral column can be included. It is the study researcher who decides the number of columns to include and the number of statements allowed in each (Kitzinger, 1987). According to Brown (1980) a topic that evokes strong polarised opinions would suit a flatter distribution. Identified

advantages of the forced distribution include the view that it leads to greater participant reflection on the statements' meanings (Prasad, 2001) and that the number of ambiguous responses is reduced in comparison to other methods such as surveys (Cross, 2005).

The quasi-normal distribution score sheet included nine columns ranging from -4 to 4 (Figure 1). The number of statements allowed in each column is indicated by the number in brackets beneath each column. A relatively wide distribution was chosen by the researcher (C. V.) to enable more nuanced views of the study topic to emerge.

2.6.4 Narrative instruction script. The narrative instruction script was designed to standardise the Q-sort process for participants. Further details of these instructions are included in the section 2.8.4 of this chapter, which describes the study procedure.

2.6.5 Written explanation of terms. The list of terms explained included, behavioural phenotyping, behavioural syndromes/genetic disorders, genetic screening and genetic testing, and intellectual and developmental disabilities. Descriptions and definitions of these terms were sourced from resources used in identifying themes from the literature (see section 2.4.1). These terms were explained due to confusion about their meaning experienced by participants in the pilot study. The explanations were checked for readability and accuracy by a study supervisor and a subset of the pilot participants.

2.7 Identification and Recruitment of Participants (P-set)

2.7.1 Inclusion criteria. Participation in the study was open to both trainee and qualified clinical psychologists, as the researcher (C. V.) was interested to understand whether cohort effects such as stage of career and training experience would affect the results. Trainee clinical psychologists are likely to draw their opinions on genetic research affecting people with IDD from knowledge they are acquiring through teaching and training experiences, whereas qualified clinical psychologists are likely to draw their opinions from actual clinical experience. As well as providing insight into what may affect attitudes to the topic, differences and similarities in the opinions of trainee and qualified clinical psychologists can show how attitudes to genetic research in relation to people with IDD are evolving among this professional group.

To participate meaningfully in the study it was deemed appropriate for participants to have had recent involvement in a service for people with IDD. Relevant services included, but were not limited to, adult community learning disability teams and child and adolescent learning disability services. Qualified clinical psychologists working in a relevant service and trainee clinical psychologists completing or who had completed a learning disability training placement no more than 12 months prior were eligible to participate in the study.

Participation in the study was open to both trainee and qualified clinical psychologists, as the researcher (C. V.) was interested to understand whether cohort effects such as stage of career and training experience would affect the results. Trainee clinical psychologists are likely to draw their opinions on genetic research affecting people with IDD from knowledge they are acquiring through teaching and training experiences, whereas qualified clinical psychologists are likely to draw their

opinions from actual clinical experience. As well as providing insight into what may affect attitudes to the topic, differences and similarities in the opinions of trainee and qualified clinical psychologists can show how attitudes to genetic research in relation to people with IDDS are evolving among this professional group.

2.7.2 Exclusion criteria. Clinical psychologists currently working in other areas were excluded from the study. Trainee clinical psychologists who had yet to begin or complete a learning disability training placement or who had completed this placement more than 12 months before were also excluded.

2.7.3 Sample size. As Q methodology studies relate to participant views, rather than comparing the representativeness of participant character traits to a specific population, a large sample is not required (Mrtek, Tafesse & Wigger, 1996; Smith, 2001). Similar to other qualitative research methods, participants are selected based on their relevance to the study's aims rather than on whether they are representative of a wider population (Chinnis, Debra & Stephen, 2001; Cordingley et al., 1997).

The number of participants (P-set) is usually smaller than the number of statements in the Q-set (Brouwer, 1999; Smith 2001). As a general guide a ratio of one participant for every three statements is recommended (Webler, Danielson & Tuler, 2007). Another guide is that four or five participants are included to define each viewpoint in the concourse (Brown, 1980) and according to Brouwer (1999) there are usually only three or four viewpoints in a concourse and rarely more than six. In accordance with these recommendations, 27-30 participants were considered sufficient for this study containing a Q-set of 81 statements.

2.7.4 Recruitment. The researcher (C. V.) attended meetings of the North West Special Interest Group for Learning Disabilities and invited attendees, who included both qualified and trainee clinical psychologists, to participate in the study. The participant information sheet and study consent form (Appendix 7) were also circulated to the members of this group via email. Three university clinical psychology programmes in the north west of England agreed to distribute an e-mail to second and third year trainee clinical psychologists. The e-mail contained an invite to participate along with the participant information sheet and study consent form. A sampling method called snowball sampling (Noy, 2008) was also used, whereby participants were asked to recommend other potential participants for the study.

2.8 Conducting the Q-sorts

2.8.1 Venues and environmental conditions. The study researcher (C. V.) arranged individual meetings with participants to complete the Q-sorts. Each participant completed their Q-sort at a time and place convenient to them. Meeting locations included university buildings and places of work. All Q-sorts were completed in quiet areas with no external distractions and with sufficient desk or floor space for the quasi-normal distribution grid. It took a mean of 66.5' (range = 35-105') for participants to complete all aspects of the process outlined in section 2.8.

2.8.2 Obtaining informed consent. The researcher (C. V.) reminded participants of the purpose and aims of the study and asked them to re-read the participant information sheet. Participants gave their signed consent to the study's

aims and agreed that they understood what participation would involve, including anonymity and confidentiality issues.

2.8.3 Initiating the Q-sort process. Before beginning the Q-sort process participant queries were addressed. The written explanation of terms, the 81 statement Q-set, the guide bar and the quasi-normal distribution grid score sheet were then presented to participants. The Q-set statement cards were shuffled anew for each participant.

2.8.4 Completing the Q-sort process. The narrative instruction script was used to guide the completion of each participant's Q-sort. This paragraph outlines the procedures contained in this script and relevant explanatory details for their inclusion. Figure 2 also details the main procedural steps for completion of this study.

When completing a Q-sort participants are guided by a specific condition of instruction (Cordingley et al., 1997). In this study participants were instructed to sort the statements according to the following condition of instruction:

“To what extent do you agree or disagree with the viewpoint expressed in each statement”

The instruction reminded participants to carefully read each statement and to use the guide bar to complete the initial ranking by placing the statements in the “agree”, “disagree” and “neutral” categories. The neutral category was described as being for statements that the participant felt neutral, uncertain or ambivalent towards. This initial ranking step is considered useful in aiding the sorting of statements along the wider continuum (Stainton Rogers, 1995). This process also familiarised participants with the statements prior to performing the main Q-sort.

The number of statements in each category were counted and noted on the score sheet. This showed the initial distribution of statements for each participant across the three categories. Participants were then instructed to take the statements from their “*agree*” pile, and working from the outermost column in, the +4 column (Figure 1), to place the statements according to their level of agreement with them. Participants continued to do this until all statements in their “*agree*” pile had been placed. The reverse was then instructed for the statements in the “*disagree*” pile, with statements being placed from the -4 column inwards until all statements in the pile were sorted. The “*neutral*” pile was the last to be sorted. If applicable, surplus statements from either the “*agree*” or “*disagree*” piles were added to the “*neutral*” pile or excess neutral statements were placed towards the lower ranges of the agree and disagree panels. Participants were restricted to four statement choices for columns +4 and -4, six statement choices for columns +3 and -3, 10 statement choices for columns +2 and -2, 13 statement choices for columns +1 and -1 and 15 statement choices for column 0, the most neutral column. The instructions informed participants that ranking of statements within columns of the score grid was unnecessary. The Q-sort process came to an end once the correct number of statements were placed in each column of the score grid and the participant was satisfied that the sort represented their views. Each statement’s position in the score grid was recorded using the number it had been pre-assigned (1-81). Upon completion participants were asked to briefly reflect on their experience of the Q-sort process. They were also asked to give a reason for each statement choice made in the -4 and +4 columns. Thus, participants gave qualitative feedback for the eight statement choices that represented their lowest and highest levels of agreement.

2.8.5 Demographic details. After completing all aspects of the Q-sort process participants were asked to state the amount of time, in years, they had accumulated working with people with an IDD in any capacity. Qualified clinical psychologist participants were also asked to state how much of this time had been accumulated post qualification as a clinical psychologist.

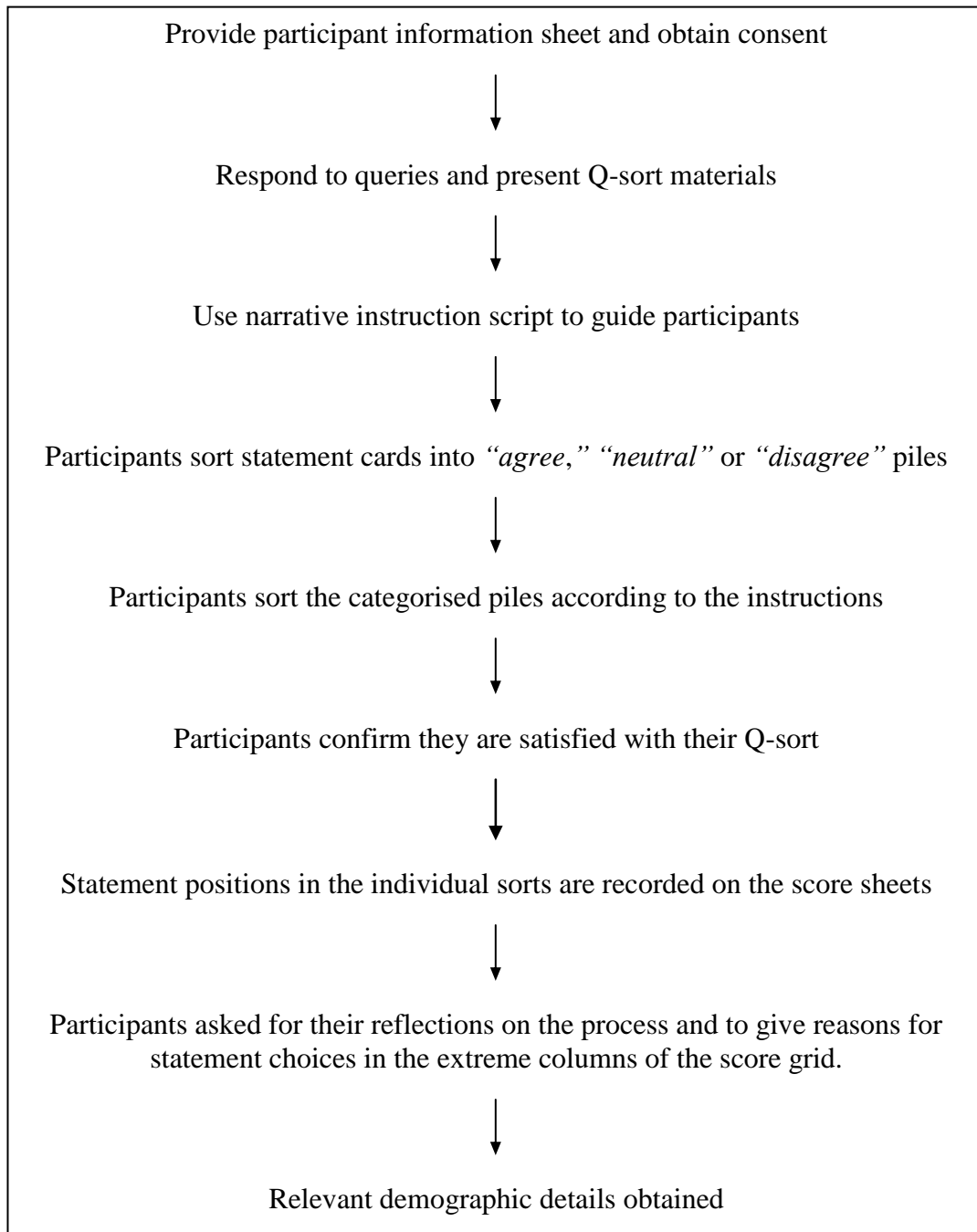


Figure 2. Procedures in Completing the Q-Sort

2.9 Data Handling and Confidentiality

The guidelines of the Data Protection Act 1998 for handling, processing, storage and destruction of information were adhered to during this study. To ensure confidentiality all quasi-normal distribution grid score sheets and paper sheets containing qualitative and demographic information were anonymous, coded only with an assigned study number. The researcher (C. V.) was the only person able to differentiate between study numbers of participants in the qualified clinical psychologist group and participants in the trainee clinical psychologist group. The anonymised data were entered into a password protected computer. Participant identifiable information included a list of participant e-mail addresses for contact purposes and the signed consent forms. The former was contained in an encrypted document on a password-protected computer to which only the researcher (C. V.) had access. The latter were stored in a locked filing cabinet at the researcher's (C. V.) university base. Any information from participants regarding their professional work experiences with people with IDD was obtained to meet the needs of the study only. No unnecessary information was recorded and any spontaneous information offered by participants, for example, experiences of a personal nature, were not recorded unless specific consent to do so was obtained from the individual participant. Email addresses were the only personal details recorded. These were not recorded on any study data and the encrypted computer file on which they were recorded was kept separate to computer files containing study data. No hardcopy version of this information was kept. Qualitative feedback in the form of direct quotes are cited in this thesis, however, no personal references are made within these and the participant study numbers have been omitted, therefore, there is no connection between the feedback and the participants involved.

2.10 Data Analysis

PQMethod 2.33 (Schmolck & Atkinson, 2012), a statistical software package designed for handling Q data was used to analyse the data in this study. PQMethod inverts traditional factor analysis by representing participants in columns and representing the items (statements) in rows. A correlation matrix is produced that correlates each participant's Q-sort with all the other Q-sorts available in the study. The correlation matrix shows how similar or dissimilar pairs of participant Q-sorts are. Rotated factor analysis then groups together Q-sorts that appear similar and according to Dennis (1986), more than one but less than seven factors typically emerge. PQMethod also incorporates varimax rotation of factors, however, within the package factors can also be rotated manually. Use of PQMethod's inbuilt varimax rotation procedure is recommended and manual or theoretical rotation of factors recommended for specific circumstances only (Coogan & Herrington, 2011). In this study, the data was subject to principal components analysis with a varimax rotation. The extracted factors were then interpreted in conjunction with 'exemplar' participants' qualitative feedback regarding the statements placed in the extremes of their quasi-normal distribution score grid. Exemplar participants are those whose Q-sorts load strongly on to a factor. Theory, research and culture are influences thought to complement the interpretation of factors (Stainton Rogers, 1995) and were incorporated in the interpretation of factors in this study. Demographic information can also aid interpretation of factors (Eccleston et al., 1997). Consequently, participants' clinical psychology status (i.e. trainee or qualified) and amount of work experience with people with IDD were considered in the analysis of the emerging factors.

Chapter 3. Results

3.1 Participant Information

Participants were 15 qualified clinical psychologists (11 female, 4 male) and 16 trainee clinical psychologists (14 female, 2 male). The qualified clinical psychologist participants were recruited from a Learning Disability Special Interest Group in the north west of England, the area in which they all worked. The trainee clinical psychologist participants were recruited via three university clinical psychology departments in the north west of England and were either in their second or third year of training. Table 1 outlines the number of years work experience participants had with people with IDD.

Table 1.

Work Experience in Years with People with IDDs

Work Experience	Overall group Mean (range)	Trainee Mean (range)	Qualified Mean (range)
In any capacity	6.7 (.5-27)	2.8 (.5-9)	10.9 (1.5-27)
Qualified experience	Not applicable	Not applicable	7.1 (.33-21.5)

3.2 Q-sort Process

Table 2 outlines how the 81 statements in the Q-set were ranked by the overall group of participants, by the trainee clinical psychologist participants and by the qualified clinical psychologist participants. The time taken to complete the Q-sort process is also detailed by group.

Table 2

Initial Participant Q-sorts

Q-sort descriptors	Overall group	Trainee	Qualified
	Mean (range)	Mean (range)	Mean (range)
Number of <i>agree</i> <i>with</i> statements	37.9 (29-47)	38.5 (29-47)	37.3 (31-46)
Number of <i>disagree</i> <i>with</i> statements	23.4 (9-35)	24.8 (15-35)	21.9 (9-29)
Number of <i>neutral</i> statements	19.7 (5-36)	17.7 (5-34)	21.9 (9-36)
Time taken to complete Q-sort	66.5' (35-105')	66.8' (35-105')	66' (35-105')

3.3 Statistical Analysis

A dedicated Q methodology statistical programme, PQMethod 2.33 (Schmolck & Atkinson, 2012), was used. This programme computes inter-correlations among Q-sorts and uses factor analysis to identify relevant factors emerging from the data. Principal components analysis was used in this study to factor analyse the Q-sorts. It was used in conjunction with a varimax procedure to rotate the factors and identify those that should be extracted. This procedure maximises the dispersion of factor loadings within the factors; thus, increasing the sum of variance explained by the extracted factors and allows those sorts that load clearly on to a single factor be identified (Donner, 2001). As Stainton Rogers (1995) identified, factors are interpretable if at least one Q-sort has a majority loading on the factor and minor loadings on the other factors. Participants' loadings on each of the

extracted factors were determined and the exemplary Q-sorts that defined each factor were identified. Analysis of an exemplar Q-sort for each factor was made based on a weighted averages method in which higher loading exemplars are given more weight in the merger (Brown, 1980). To aid interpretation of the data, a factor array was created for each factor by merging all the exemplars.

See Brown (1980) and Stainton Rogers (1995) for more in-depth descriptions of statistical analysis in Q methodology.

3.4 Factor Analysis

Principal components analysis yielded eight initial factors. As recommended by Kaiser (1960), only those factors with an eigenvalue greater than one were extracted. This resulted in a three factor solution (Table 3), which was then subjected to a varimax rotation.

Table 3

Eigenvalues and Explained Variance for a Three Factor Solution

Factor	Eigenvalue	E.V.* (Cumulative E.V.)	Rotated E.V. (Cumulative E.V.)
1	15.2514	49%	32%
2	3.4880	11% (61%)	26% (58%)
3	1.2041	4% (64%)	7% (65%)

Note. * E.V. = explained variance

Factor 1 accounted for 32% of the rotated variance and was the strongest factor to emerge from this analysis. Factors 2 and 3 accounted for 26% and 7% of the rotated variance respectively.

3.4.1 Factor loadings. Table 4 shows the factor loadings for the participants on each of the three factors. More loadings were positive, indicating that participants agreed rather than disagreed with the factors even if they did not load significantly on to them (This table is reproduced in Appendix 8 in an expanded form with categorical descriptions included with loadings).

Table 4
Loadings for Each Participant and Factor

Participant	Factor 1	Factor 2	Factor 3
1	0.0921	0.7522*	-0.0393
2	0.4548	0.6173*	0.1643
3	0.4599	0.5267*	0.2254
4	0.2184	0.7584*	0.1694
5	0.3909	0.6441*	0.1796
6	0.4372	0.1691	0.5464*
7	0.8247*	0.2227	0.1233
8	0.2031	0.6912*	0.3047
9	0.3083	0.7088*	0.2336
10	0.7018*	-0.0040	0.3525
11	0.7620*	0.1635	0.2757
12	0.7965*	0.1451	0.2031
13	0.0617	0.7744*	-0.0098
14	0.7758*	0.3593	-0.1065
15	0.4748	0.4760	0.2685
16	0.2159	0.6483*	-0.4502
17	0.6087*	0.3311	0.2648
18	0.7226*	0.2308	0.2025
19	0.3940	0.4125	0.4794
20	0.6261*	0.4816	0.0525
21	0.0279	0.7655*	0.2498
22	0.7775*	0.3447	0.1892
23	0.6642*	0.5103	0.1806
24	0.3466	0.7745*	0.1254
25	0.8154*	0.0980	0.2578
26	0.6623*	0.3281	0.2532
27	0.5774	0.3686	0.4543
28	0.7808*	0.0317	-0.1611
29	0.2193	0.8081*	0.0507
30	0.4420	0.3139	0.4996
31	0.8263*	0.2716	0.0635

Note. * = Scores significantly loading onto a factor

In line with Cattell (1944) and the systematic analysis of each factor solution, 27 participant responses (marked with a * in Table 4 above) were included, which explained 65% of the variance (Table 3). Fourteen participants loaded on to Factor 1, 12 participants loaded on to Factor 2 and one participant loaded onto Factor 3. The four participants who failed to load significantly on to any factor had mixed loadings (i.e. very similar scores on more than one factor). Participants who were significantly associated with a given factor were assumed to share a viewpoint (McKeown & Thomas, 1988).

3.4.2 Factor demographics. Table 5 shows the number of participants by their participant group (trainee or qualified clinical psychologist) that loaded on to each of the three factors, together with the mean work experience in years each participant had with an IDD population. The years' experience as a qualified clinical psychologist is also indicated for the qualified clinical psychologists. Due to the different ratio of trainee and qualified clinical psychologist participants on each factor, the amount of work experience with people with IDDs varied across factors.

Overall, there was little difference in the amount of work experience for qualified clinical psychologists loading on Factors 1 and 2, but participants loading on Factor 2 had, on average, four times the number of years experience in a qualified role as participants on Factor 1. Participants on both factors had overall work experience means close to the mean for all qualified clinical participants in the P-set (10.9 years). The mean amount of qualified experience for participants on both factors, however, was different to the qualified P-set mean (7.1 years), i.e. less with regard to Factor 1 and slightly higher for Factor 2.

In the trainee clinical psychologist group, the mean amount of work experience of those loading on to Factor 2 was almost double that of their counterparts on Factor 1. The Factor 1 mean was near to the mean for all trainee clinical psychologists in the P-set (2.8) and the Factor 2 mean was 1.6 years more.

The participant who loaded on to Factor 3 had more overall and qualified work experience with an IDD population than the mean overall and qualified experience of qualified clinical psychologists in the P-set.

Table 5

Number of Participants Loading on to Each Factor and Mean Work Experience in Years with People with an IDD

Factor	Overall group		Trainee clinical psychologists		Qualified clinical psychologists	
	No.	Experience	No.	Experience	No.	Experience (Qualified experience)
1	14	5.25	9 (F*)	2.33	5 (3F, 2M)	10.6 (2)
2	12	12.5	4 (3F, 1M)	4.4	8 (6F, 2M)	10.5 (8.33)
3	1	15.5			1 (F)	15.5 (11)

Note. * = F denotes female participants and M denotes male participants

3.4.3 Factor arrays. Table 6 outlines where the statements in the Q-set were ranked on each of the three factors. Each factor has a column within the table detailing its factor array. Rankings range from -4, representing the “*most disagreed with*” statements to factor +4, representing the “*most agreed with*” statements.

Table 6
Q-sort Arrays for all Factors

Statements	Factor Arrays		
	F 1	F 2	F 3
1 Diagnosing different intellectual and developmental disabilities does not lead to greater understanding or change	-4	-1	1
2 The medical model of disability is more helpful than the social model of disability in intervening with people with intellectual and developmental disabilities	-2	-4	-4
3 Sociocultural factors such as socioeconomic status and family background are more important than behavioural phenotyping in explaining behaviour	0	3	-1
4 Growing up in the same family has no discernible or marked effect on the IQs of siblings	-2	-3	-3
5 Intellectual ability is affected more by nature than by nurture	-1	-1	0
6 Humans are not born with innate tendencies; experience in the environment shapes all learning	-3	-1	-3
7 Behavioural syndromes rarely occur	-2	-1	-1
8 The cause of intellectual disability can be of equal or greater importance than the immediate and broader environment as a determinant of well-being	-2	-3	-2
9 Genetically inherited traits are fixed and non-modifiable	-3	-4	-4
10 Genetic screening/testing causes negative attitudes towards disability to pervade and continue	-2	1	0
11 What’s not important is genetic screening but the information it yields and how that is used	3	0	4
12 Awareness of behavioural phenotyping and its developments is generally of a low level among staff	2	1	1
13 The social model of disability is more helpful than the medical model of disability in intervening with people with intellectual and developmental disabilities	1	4	1

Statements	F 1	F 2	F 3
14 Understanding the meaning or function of an idiosyncratic behaviour is more important than understanding the diagnostic label	2	4	-1
15 Understanding the diagnostic label informs idiosyncratic formulations, interventions and treatment plans	4	0	2
16 The pathway to the same behaviour may be different for individuals with different genetic disorders	2	1	2
17 Impact of genetic screening on the individual and society should be considered before it is used	3	1	4
18 Many professionals working with individuals with intellectual and developmental disabilities are unconcerned with why someone has the impairment	1	1	-2
19 Diagnostic labels can serve to deny people access to services	1	2	2
20 Service provision should be provided primarily on the basis of need not diagnosis	3	4	-3
21 Challenging behaviour needs to be considered in the wider context of family and socioeconomic background, it is unethical to work with people with challenging behaviour and ignore this context	4	4	3
22 Focusing on the individual's expressed difficulties is more helpful than looking at the difficulties in the context of a diagnosis or conflicting diagnoses	1	3	-1
23 Associating genetics with people with intellectual and developmental disabilities can undermine the progress of the social model of disability	-1	1	4
24 The environment that a child experiences is as much a consequence of the child's genes as it is of external factors: the child seeks out or creates his or her own environment	-1	-2	0
25 It is difficult to hold both a medical and a social model of intellectual and developmental disability	-4	-2	3
26 Diagnosis specific services presume specialisation which fits with a medical model	-1	1	1
27 Heterogeneity of presentations within diagnostic categories can render the diagnosis meaningless	-1	2	-3
28 Genetic screening can lead to negative social engineering with the creation of 'designer' societies where people with disabilities are undervalued and social/environmental influences on disability undermined	0	1	4
29 Inclusion in society can be enhanced by understanding individual difference that can be traced to a specific genetic disorder	1	-3	-1
30 Behavioural phenotyping can make a positive impact on quality of life for people with intellectual and developmental disabilities if the information is used appropriately	4	0	3

Statements	F1	F2	F3
31 The medical and social models of disability can and should be integrated to enhance the lives of people with intellectual and developmental disabilities	4	2	3
32 Diagnosing intellectual and developmental disabilities/behavioural syndromes can hinder and create barriers to therapeutic work	-3	0	-2
33 The cause of the intellectual and developmental disability is unknown in most cases	1	0	-1
34 Genetic disorders lead to different behavioural outcomes	2	-1	2
35 Families usually want to know the cause of their child's intellectual and developmental disability	2	2	2
36 Health and social care resources should be directed towards understanding and improving social and environmental factors affecting people with intellectual and developmental disabilities	2	3	0
37 Health and social care resources should be directed towards disseminating and applying research, originating from all fields of relevance, affecting people with intellectual and developmental disabilities	2	1	1
38 The environment has less of an impact in an equal society than it does in a more unequal society	1	-1	0
39 Genetic screening may reduce complex behaviour to genes, ignoring the impact of other factors on behaviour	0	3	2
40 A diagnostic label is more helpful to an individual than it is unhelpful	0	-2	3
41 For disorder-specific services to be effective for the individual there must be no doubt in the accuracy of their diagnosis	-2	-1	-1
42 Genetic screening, and its consequences, masks and denies the individuality and opportunity in people with intellectual and developmental disabilities	-2	1	0
43 What happens in the womb influences intelligence more than anything that happens after birth	-2	-4	-4
44 Genetic screening creates a battle between innate social instinct versus human rights	0	0	0
45 Defining services by diagnostic labels means better, more individually tailored services are delivered	0	-3	-2
46 Geneticists have a moral and ethical responsibility to the today not the tomorrow	-1	-1	-2
47 No one knows what the non-genetic causes of individuality are	-2	-3	1
48 Innate abilities allow children to develop typically, absence of such abilities affects development	1	-2	2
49 Genetic testing would not add to the quality of life for a person with an intellectual or developmental disability	-3	0	-3
50 Labelling genetic disorders/behavioural syndromes may negatively affect prognosis	-1	0	-1

Statements	F 1	F 2	F 3
51 Positive behaviour support is more influential in improving the quality of life for a person with an intellectual or developmental disability than the outcomes of behavioural phenotyping and genetic testing	1	3	1
52 Genetic screening and behavioural phenotyping are akin to modern day eugenics	-4	-1	-2
53 The non-shared environment, such as individual school experiences, account for more differences in siblings than genes	0	0	-1
54 Genetic screening/testing should be available to all as people have a right to information about their genetic make-up	0	0	0
55 The shared environment has a greater influence on sibling similarities and differences than genes	0	2	-1
56 Genetically inherited traits/characteristics can be subject to change and adaptation by the environment - Heritability does not mean immutability	2	2	2
57 Degree of intellectual disability (mild, moderate, severe, profound) is a better predictor of behavioural outcomes than behavioural syndrome diagnosis	-3	-2	-2
58 Genetics and behavioural phenotyping represent a shift backwards to the medical model of disability	-3	1	0
59 Genetic aspects of a condition may be viewed as irrelevant or potentially negative by professionals/staff members	0	0	0
60 The science of intelligence testing is flawed	3	2	1
61 Genetic screening, if used in an appropriate, responsible way, has the potential to positively affect lives	3	0	3
62 Genes have a greater influence on sibling similarities and differences than the shared environment	-1	-3	-1
63 Understanding the causal pathway to an individual difference can have a positive influence on well-being	1	-1	2
64 Culture is the product of individual psychological make-up rather than vice versa – a person does not inherit cultural knowledge, they acquire it through experience	1	2	1
65 Genes have a continuing influence on individuals as they develop	2	0	2
66 Focusing on how genetics influences behaviour downplays the role of more important social influences	-1	2	0
67 Diagnosing different intellectual and developmental disabilities/behavioural syndromes provide unnecessary labels and can create stigma	-1	0	-2
68 As people get older their environment has a stronger influence on their behaviour	-1	-2	-2
69 The social environment is the product of individuals innate social instincts	-1	-2	-1
70 The diagnosis is more meaningful than the presenting behaviour	-4	-4	-4
71 Generic services beneficial to all is the ideal but if this is unattainable disorder-specific services should be preferred	0	-2	-1

Statements	F1	F2	F3
72 People with different behavioural syndromes have more similar than dissimilar behaviours	0	-2	1
73 Behavioural phenotyping is beneficial in understanding some syndromes such as Down syndrome but this is an exception, behavioural phenotyping does not generally aid understanding as much as other factors	-2	-1	-3
74 Current knowledge and understanding of different disorders is too limited to make disorder-specific services worthwhile	-1	-1	1
75 There are no direct genes for intelligence but an inherited resistance to stressors e.g. resistance to toxins which then enhances the ability to develop intelligence	0	-2	0
76 Robustness of the conceptualisation of disorders/disabilities needs to be improved e.g. autism, otherwise the science (phenotyping) on which it is based is flawed and any predictions made on this basis are flawed and potentially destructive	1	-1	1
77 The voice of people with intellectual and developmental disabilities in the genetic testing debate is unheard	3	3	-2
78 In clinical practice it is difficult to keep up to date on new research developments due to time and service pressures	1	1	1
79 Research of relevance to people with intellectual and developmental disabilities is often difficult to access by services which affects the application of new research	0	0	0
80 Social instincts may mean it is natural to exclude people with intellectual and developmental disabilities (IDDs) from the group; however, our human side and social responsibility should cause us to fight against this and recognise the value to the world of people with IDDs and diversity in general	2	2	0
81 A partially inherited low IQ might be subject to extensive improvement through education	0	1	0

The individual Q-sort arrays for each factor are reproduced in quasi-normal distribution grids (Figures 3, 4 and 5), which represent the “ideal” Q-sort for each factor. Statements have been entered into the grids according to z-scores, from highest to lowest, beginning with the first statement entered in the +4 column and ending with the last statement entered in the -4 column. The statements placed at the extreme ends of each Q-sort grid, termed the “exemplars,” will be discussed in relation to the specific factor together with reflective comments made by participants regarding the exemplars (See Appendix 9 for a full list of reflective comments for all factors).

3.5 Distinguishing and Consensus Statements

A *distinguishing* or distinctive statement occurs when a statement’s score on two factors exceeds the magnitude of difference between a statement’s score on any two factors that is required for it to be statistically significant (van Exel & de Graaf, 2005). A statement may be distinctive between two factors but is not usually recognised as such unless it distinguishes one factor from all the other factors (van Exel & de Graaf, 2005). A statement that does not distinguish between any of the factors is called a *consensus* statement (van Exel & de Graaf, 2005). Distinguishing and consensus statements illustrate differences and similarities between factors. Such statements will be highlighted in the factor descriptions included in this chapter. Lists of distinguishing statements and consensus statements for each of the factors are detailed in tables in Appendices 10 and 11 respectively.

3.6 Factor Descriptions and Interpretations

In this section, themes relating to the three factors will be described. Factors are given names that best describe the pattern of statements in the given factor (Corr, 2001) and thus the three factors will be named to reflect their main themes. The exemplary statements in the “*most agreed with*” (+4) and the “*most disagreed with*” (-4) columns of each of the respective factor arrays are discussed. These statements are introduced under headings indicating their status as either a positive or a negative exemplary statement. Under these headings, statements are discussed in the order of ranking within the factor arrays, for example, on Factor 1, statement number 21 was the most agreed with statement overall and statement 70 the most disagreed with and they are therefore discussed first and last under their respective headings. The statements will be discussed in conjunction with the relevant reflective comments. Where applicable, participant ratings of other statements in the Q-set will be highlighted to support factor interpretations. These are presented in brackets with the corresponding factor array rating.

3.6.1 Factor 1: Integration of social and medical models of disability.

Factor 1 accounted for 32% of the variance and represented the view of nearly half the participants. The overarching theme that emerged from this factor was the importance of both the medical and social models of disability and that their integration was the most appropriate response when working with people with IDD.

Most Disagree			Neutral			Most Agree		
-4	-3	-2	-1	0	1	2	3	4
1	49	47	24	53	78	16	77	21
52	32	8	69	81	63	14	20	31
25	58	42	5	79	19	37	17	30
70	9	10	74	55	48	12	11	15
	6	41	23	59	22	34	60	
	57	73	50	28	29	80	61	
		43	46	44	76	36		
		7	68	72	38	65		
		4	26	75	51	35		
		2	27	54	13	56		
			66	39	18			
			62	40	33			
			67	71	64			
				3				
				45				

Figure 3. Factor 1 Array

3.6.2 Factor 1: Positive exemplary statements.

Statement 21: Challenging behaviour needs to be considered in the wider context of family and socioeconomic background, it is unethical to work with people with challenging behaviour and ignore this context. Factor 1 participants who agreed with statement 21 thought that understanding the wider context in which a behaviour occurs is necessary to understand the cause, “*Ignore context assumes behaviour is just about person and not a reasonable response to environment,*” “*We don’t exist in a vacuum.*” This was also thought important for comprehensive formulations, “*Can’t formulate on diagnosis alone*” and for intervention, “*Always have to know about family and wider system to effect any change regardless of whether it is a phenotypic behaviour.*” One participant highlighted a nature-nurture interaction, “*I do believe environment shapes behaviour but within confines of what is inherent to the individual,*” whilst another described challenging behaviour as a “*social construction.*” Statement 21 was a consensus statement, meaning that the overall P-set agreed with its premise.

Statement 31: The medical and social models of disability can and should be integrated to enhance the lives of people with intellectual and developmental disabilities. Factor 1 participants agreeing with statement 31 recognised a complementary role for the social and medical models in working with people with IDD, with both judged to be of value, “*Maximum benefit can be derived if both models are taken into account.*” Some participants considered a conflict to exist between these perspectives but one that should be overcome, “*The strengths of the medical and social models should be integrated to benefit the lives of individuals rather than a continued battle between the models.*” One participant commented that

integration of these models was required as a psychologist, *“If you’re not doing this you’re not doing your job.”*

Factor 1 participants also disagreed with statement 23 (-1, Associating genetics with people with intellectual and developmental disabilities can undermine the progress of the social model of disability).

Statement 30: Behavioural phenotyping can make a positive impact on quality of life for people with intellectual and developmental disabilities if the information is used appropriately. The potential benefits of behavioural phenotyping in terms of formulation and intervention were recognised by Factor 1 participants agreeing with statement 30, *“Phenotyping has the potential to enable us to understand the individual needs of clients leading to better informed formulations and intervention plans.”*

Factor 1 participants disagreed with statement 73 (-2, Behavioural phenotyping is beneficial in understanding some syndromes such as Down syndrome but this is an exception, behavioural phenotyping does not generally aid understanding as much as other factors).

Statement 15: Understanding the diagnostic label informs idiosyncratic formulations, interventions and treatment plans. Participants on Factor 1 agreeing with statement 15 considered diagnosis of value in informing the formulation, *“Understanding diagnostic label gives you an idea why somebody behaves or responds in a certain way”* and that it was important to understand a diagnosis when designing interventions as, *“If you don’t you might use an intervention that is doomed to failure.”* Diagnosis seemed to be viewed as a helpful heuristic or adjunct when working with people with IDD’s rather than a dominant factor, *“Key question is inform not dictate.”* One participant also stated that having a diagnosis helped

when working with staff teams indicating the possible dominance of or preference for a medical model among other staff groups.

Factor 1 participants disagreed with statement 32 (-3, Diagnosing intellectual and developmental disabilities/behavioural syndromes can hinder and create barriers to therapeutic work).

3.6.3 Factor 1: Negative exemplary statements.

Statement 1: Diagnosing different intellectual and developmental disabilities does not lead to greater understanding or change. Factor 1 participants disagreeing with this statement thought that a diagnosis offered insight into prognosis, *“If you’re able to identify syndrome then gives you a realistic picture of what you can change”* and that there was evidence to disprove this statement, *“I think there’s evidence to say that’s not true, in forensic and mental health stream, in medical model, in family unit.”* Statement 1 was a distinguishing statement for Factor 1, meaning that there was a significant difference between Factor 1 participants’ views of the premise of this statement and the views of other participants in the P-set.

Factor 1 participants agreed with statement 34 (+2, Genetic disorders lead to different behavioural outcomes).

Statement 52: Genetic screening and behavioural phenotyping are akin to modern day eugenics. Disagreement with this statement centred on Factor 1 participants viewing the aims of genetic screening and eugenics as being different, *“Genetic screening is about information for preparation to ensure person is helped in right way and not trying to reduce social value like eugenics did.”* Time was thought to have changed cultural beliefs for the better, *“Culture has moved to the*

individual, intentions are sound” and that not allowing people access to genetic screening was a “disservice.” One participant however felt that there remained a residual negative threat with genetic screening *“It has potential to be abused.”*

Factor 1 participants disagreed with statement 10 (-2, Genetic screening/testing causes negative attitudes towards disability to pervade and continue), statement 58 (-3, Genetics and behavioural phenotyping represent a shift backwards to the medical model of disability) and statement 49 (-3, Genetic testing would not add to the quality of life for a person with an intellectual or developmental disability). They also agreed with statement 11 (+3, What’s not important is genetic screening but the information it yields and how that is used).

Statement 25: It is difficult to hold both a medical and a social model of intellectual and developmental disability. Comments by Factor 1 participants on this statement related to this being part of a clinical psychologist’s role and not doing this meant that this role was not being fulfilled adequately, *“Clinically neglectful not to hold both in mind,” “That’s formulation and that’s what we’re supposed to do. No reason why two can’t be integrated.”* Statement 25 was another distinguishing statement for Factor 1.

Statement 70: The diagnosis is more meaningful than the presenting behaviour. Some participants disagreeing with this statement appeared to see a more equitable role for diagnosis and presenting behaviour, *“I think the two are equally important. If you ignore presenting behaviour you are not going to get very far.”* Others seemed to consider presenting behaviour as more relevant, for example, *“A diagnosis without a presenting problem would not be seen (in services).”*

Participants seemed to believe that a diagnosis could be helpful but that it was

necessary to look beyond a diagnostic “*label.*” Statement 70 was a consensus statement.

Factor 1 participants agreed with statement 20 (+3, Service provision should be provided primarily on the basis of need not diagnosis) and statement 14 (+2, Understanding the meaning or function of an idiosyncratic behaviour is more important than understanding the diagnostic label). These two statements were in the +4 rank on Factor 2.

3.6.4 Factor 2: Social model of disability is more helpful.

Factor 2 accounted for 26% of the variance and represented views of 12 participants and was therefore another primary factor. The main theme emerging from this factor was that the social model of disability is more helpful and of more relevance in working with people with IDD. Opinions about the utility of a diagnosis were relatively negative among Factor 2 participants. In comparison to Factor 1’s theme of equal importance of medical and social models, Factor 2’s main theme was that presenting behaviour and environmental factors are more important than information yielded from a diagnosis.

Most Disagree			Neutral			Most Agree		
-4	-3	-2	-1	0	1	2	3	4
43	47	48	74	65	28	66	51	14
9	29	57	1	67	37	64	36	20
2	62	75	63	30	17	35	77	13
70	45	68	34	79	23	19	22	21
	8	71	52	15	16	60	39	
	4	24	38	32	10	56	3	
		25	76	11	81	27		
		40	41	50	78	31		
		72	73	54	26	80		
		69	46	61	42	55		
			6	53	12			
			5	44	18			
			7	49	58			
				59				
				33				

Figure 4. Factor 2 Array

3.6.5 Factor 2: Positive exemplary statements.

Statement 14: *Understanding the meaning or function of an idiosyncratic behaviour is more important than understanding the diagnostic label.* Factor 2 participants agreeing with statement 14 considered a diagnostic label to be unhelpful in understanding the personal meaning and the function of behaviour, “*Label tells you nothing about a person,*” “*In some ways diagnosis can be helpful but understanding the function is more meaningful. That’s what you would be looking for without a diagnosis.*” A diagnosis was seen to add little value to clinical work by some participants, “*Whilst I do acknowledge that individual behaviour can have a genetic influence it doesn’t tell you very much regarding what to do,*” while other participants were concerned that a diagnosis was detrimental to therapeutic progress, “*Focusing on diagnosis distracts from supporting people,*” “*Some information is more dangerous than no information if you go of the diagnostic label.*” One participant stated that families often want to know about a diagnosis but considered this misguided as she thought that the most important work was “*not diagnosis related.*” Statement 14 was a distinguishing statement for Factor 2.

Factor 2 participants agreed with statement 27 (+2, Heterogeneity of presentations within diagnostic categories can render the diagnosis meaningless).

Statement 20: *Service provision should be provided primarily on the basis of need not diagnosis.* Participants agreeing with statement 20 considered the presenting needs of a client as more relevant than a diagnosis with regard to accessing services. This was due to a perceived variation of need within diagnostic categories, “*Need to look at individuals when assessing them as there is differentiation within a diagnosis,*” and that sometimes there was a diagnosis in the

absence of a clinical need and vice versa, “*You generate delusions of need based on diagnosis*” and “*People on fringes (of diagnostic criteria) miss out.*”

Overall, participants agreeing with this statement considered individual needs more important than diagnosis but also that having a diagnosis made access to services easier, summed up by the following comments, “*This is a far cry from reality*” and “*You have to know diagnosis to access certain services.*” One participant identified autistic spectrum disorders as a possible exception with need more closely linked to the diagnosis in this instance, “*There are usually some connecting factors.*” Statement 20 was another distinguishing statement for Factor 2.

Factor 2 participants agreed with statement 36 (+3, Health and social care resources should be directed towards understanding and improving social and environmental factors affecting people with intellectual and developmental disabilities).

Statement 13: The social model of disability is more helpful than the medical model of disability in intervening with people with intellectual and developmental disabilities. Factor 2 participants who agreed that the social model was more helpful in intervening with people with IDDAs considered it more useful as it was seen to place more emphasis on wider contextual factors, which were considered of greater relevance than the diagnostic label in working with people with IDDAs, “*You need to understand history and experience. You then have a better formulation, intervention and outcome when treating the diagnosis.*” The view that the social model has broader scope in interventions for people with IDDAs was apparent from comments about perceived negative characteristics of the medical model, for example, one participant called this a “*pharmacological straitjacket*” and another thought that homogenous diagnostic labels were “*unhelpful.*”

Factor 2 participants agreed with statement 23 (+1, Associating genetics with people with intellectual and developmental disabilities can undermine the progress of the social model of disability).

Statement 21: Challenging behaviour needs to be considered in the wider context of family and socioeconomic background, it is unethical to work with people with challenging behaviour and ignore this context. Participants on Factor 2 agreed with both the premise of this statement and that personal experience and other evidence showed an undoubted link between environmental influences and challenging behaviour, “*Much more compelling evidence about the environment*” and “*If we ignore context we are not going to be able to understand function of behaviour or the formulation.*” Statement 21 was a consensus statement.

Factor 2 participants agreed with statement 3 (+3, Sociocultural factors such as socioeconomic status and family background are more important than behavioural phenotyping in explaining behaviour).

3.6.6 Factor 2: Negative exemplary statements.

Statement 43: What happens in the womb influences intelligence more than anything that happens after birth. Factor 2 participants disagreeing with this statement considered intelligence to be affected by much more than prenatal influences, “*Intelligence is influenced by a range of factors, this is life-long*” and thought that acknowledging a biological influence on intelligence only was blaming of mothers, “*It’s a rod to beat women with.*”

Factor 2 participants agreed with statement 39 (+3, Genetic screening may reduce complex behaviour to genes, ignoring the impact of other factors on behaviour).

Statement 9: Genetically inherited traits are fixed and non-modifiable. This statement was disagreed with by participants on Factor 2 who thought that inherited traits are subject to adaptation by environmental factors such as opportunity, “*People can change given opportunities and support*” and “*Social modelling.*” Other participants considered the interaction between genetics and environmental factors and epigenetics to be of importance, “*Traits are highly modifiable with lifestyle.*” Statement 9 was another consensus statement.

Factor 2 participants disagreed with statement 48 (-2, Innate abilities allow children to develop typically, absence of such abilities affects development) and agreed with statement 66 (+2, Focusing on how genetics influences behaviour downplays the role of more important social influences).

Statement 2: The medical model of disability is more helpful than the social model of disability in intervening with people with intellectual and developmental disabilities. Statement 2 is the direct opposite of statement 13, a statement placed in the polar opposite (+4) area of the Factor 2 array. Considering they are opposites of each other, it might have been expected that they would have had an equal number of participants placing them in their respective ranks on the Factor 2 array. However, this was not the case as more participants on Factor 2 placed statement 2 in the -4 column than the number of participants who placed statement 13 in the +4 column (10 versus 4 participants). Thus, whilst the medical model was not seen as more helpful than the social model, the extent to which the latter was seen as more helpful did not match the antipathy towards the medical model. This suggests that some participants loading on Factor 2 took a balanced view with both approaches regarded as useful with the balance tipping in favour of the social model.

Reflective comments about this statement have similar themes to comments made by participants about statement 13. These indicated negative beliefs about the medical model's utility with regard to work with people with IDD, "*Medical model little more than descriptive, it doesn't tell you what to do*", "*Tells you nothing regarding diagnosis, put them in a bin.*" Some participants based their opinions on personal clinical experience or other evidence, "*Clinical experience is indicative that it is pertinent to consider systemic factors*", "*More evidence for social model of disability being relevant and meaningful.*" One participant casually commented on the dominance of the medical model as though this was universally acknowledged "*Medical model is more dominant but not more helpful.*"

Statement 70: *The diagnosis is more meaningful than the presenting behaviour.* All participants on Factor 2 disagreed with this consensus statement at the -4 level. Reasons for disagreement included a perceived inability to understand individual difference within diagnoses, "*Heterogeneity and complexity, hard to capture that accurately in one label*" and the greater value placed on understanding behaviour "*We can understand an individual more effectively by analysing the function of their behaviour.*" Reasons why participants considered the presenting behaviour to be more meaningful were that it was considered more tangible to an individual's life, "*If a person is angry then what's happening now,*" was subject to modification unlike a diagnosis, "*Can't change genetic diagnosis but you can modify the behaviour*" and that it better informed clinical work, "*Looking at meaning behind the behaviour is more important to inform how we work.*"

Participants on Factor 2 expressed negative views about the relevance of a diagnosis and its contribution to clinical work including the derisive comment, "*A load of rubbish, ridiculous statement.*" Another participant commented that a

diagnosis “*Gets in way of work you do*” and another that diagnoses did not have “*Much scientific validity.*” A slightly positive view about diagnoses was held by some participants on Factor 2, for example, participants highlighted an informative aspect of a diagnosis and a comment that in “*rare*” circumstances a diagnosis can help predict behaviour.

Participants on Factor 2 agreed with statement 22 (+3, Focusing on the individual’s expressed difficulties is more helpful than looking at the difficulties in the context of a diagnosis or conflicting diagnoses) and disagreed with statement 40 (-2, A diagnostic label is more helpful to an individual than it is unhelpful). However, they also slightly disagreed with statement 1 (-1, Diagnosing different intellectual and developmental disabilities does not lead to greater understanding or change), intimating that a diagnosis was seen to have some value.

3.6.7 Factor 3: Genetic advances are positive but can create conflict with recognising the value of people with IDD.

Factor 3 accounted for 7% of the rotated variance. The factor represented one participant's views but to account for this proportion of the variance indicated that this pattern of sorting was present in some form in the Q-sorts of other participants. This illustrates how all of the Q-sorts can contribute to the emergence of factors (Stainton Rogers, 1991). Factor 3 therefore represented a secondary viewpoint of those participants who loaded on to other factors.

One of the main themes emerging from this factor was that caution must be used when applying the advances of genetics in the area of IDD so as not to undermine human rights and diminish the inherent value of people with IDDs to society. Another theme was Factor 3's relatively positive views of the medical model of disability in terms of diagnoses and thoughtful genetic testing. This view however was in conflict with a further opinion that the social model of disability was more important than the medical model of disability but risked negative consequences by advances in the latter. To summarise, Factor 3 could be described as nature - proceed with caution.

Most Disagree			Neutral			Most Agree		
-4	-3	-2	-1	0	1	2	3	4
43	27	52	50	36	26	15	21	11
9	73	18	14	38	1	16	25	17
70	20	57	53	24	37	19	30	23
2	4	67	55	42	12	34	31	28
	49	68	41	44	47	35	40	
	6	8	3	5	51	39	61	
		45	62	54	60	48		
		32	29	58	13	56		
		77	22	59	64	63		
		46	69	10	72	65		
			71	66	74			
			7	75	76			
			33	79	78			
				80				
				81				

Figure 5. Factor 3 Array

3.6.8 Factor 3: Positive exemplary statements.

Statement 11: What’s not important is genetic screening but the information it yields and how that is used. The Factor 3 participant’s reflective comments about this statement related to the ethics and use of genetic screening. She stated that the “*Most important thing to grapple with is ethics of genetic screening. (If it is) used to have a baby or not then it is not useful, is immoral. (If it is) used to increase understanding then the better I feel about it.*”

The Factor 3 participant also agreed with statement 61 (+3, Genetic screening, if used in an appropriate, responsible way, has the potential to positively affect lives), a view shared by Factor 1 participants.

Statement 17: Impact of genetic screening on the individual and society should be considered before it is used. The participant again highlighted ethical values in the use of genetic screening, particularly in relation to decisions regarding childbirth, fearing that genetic screening was not being used ethically in all countries. Her comments were, *“(This is) most important thing, should be considered before it is used. Genetic screening used to some degree and it’s not considered, what it means to people, especially in some countries. Every human being has rights. Genetic screening is fine as long as no decisions are made regarding child birth; take unborn child’s life without medical intervention.”*

Statement 23: Associating genetics with people with intellectual and developmental disabilities can undermine the progress of the social model of disability. The participant had strong views regarding the importance of people with IDD to society and regarded the medical model as undermining their value through its focus on the intellectual, *“I think people with learning disabilities have a huge amount to offer society. Society isn’t created by just intelligence. Other things are important – characteristics like being loving, caring, funny, strong, love for gardens etc. carry as much value for society. Social rather than medical/intellectual models.”* Statement 23 was a distinguishing statement for Factor 3.

Statement 28: Genetic screening can lead to negative social engineering with the creation of ‘designer’ societies where people with disabilities are undervalued and social/environmental influences on disability undermined. The participant reflected on how society could be affected if genetic screening were used in this way, *“I think this is so true. ‘Designer’ societies occur if genetic screening goes wrong - used to reduce learning disability rather than understand it. End up*

with a flawed society.” Statement 28 was another distinguishing statement for Factor 3.

The Factor 3 participant disagreed with statement 49 (-3, Genetic testing would not add to the quality of life for a person with an intellectual or developmental disability), indicating their support of genetic testing if used appropriately, as per their reflective comments for statement 11 above.

3.6.9 Factor 3: Negative exemplary statements.

Statement 43: What happens in the womb influences intelligence more than anything that happens after birth. The participant described a personal example to demonstrate why she had placed this statement in the -4 column. Statement 43 was a consensus statement.

Statement 9: Genetically inherited traits are fixed and non-modifiable. The Factor 3 participant disagreed with this consensus statement for the same reason as participants on Factor 2, *“Inherit but environment impacts on life.”*

The Factor 3 participant also slightly agreed with statement 47 (+1, No one knows what the non-genetic causes of individuality are), which was further indicative of their view of the importance of environmental factors in shaping individuality.

Statement 70: The diagnosis is more meaningful than the presenting behaviour. The Factor 3 participant disagreed with this consensus statement because she thought *“Both important, both are meaningful.”*

The Factor 3 participant showed agreement for statement 40 (+3, A diagnostic label is more helpful to an individual than it is unhelpful). They also showed slight disagreement for statements 14 and 22 (-1, Understanding the

meaning or function of an idiosyncratic behaviour is more important than understanding the diagnostic label; -1, Focusing on the individual's expressed difficulties is more helpful than looking at the difficulties in the context of a diagnosis or conflicting diagnoses). These views show the value the Factor 3 participant afforded diagnosis, although they considered presenting behaviour more important. Factor 3 ratings on these statements were different to those of participants on the other two factors, in particular, participants on Factor 2.

The Factor 3 participant also disagreed with statement 20 (-3, Service provision should be provided primarily on the basis of need not diagnosis). Again, participants on the other two factors had contrasting views about this statement and the contrast between Factors 3 and 2 was greater than the contrast between Factors 3 and 1.

Statement 2: The medical model of disability is more helpful than the social model of disability in intervening with people with intellectual and developmental disabilities. As with participants on Factor 2, the participant on Factor 3 disagreed with this statement, commenting, *“Not true – social model is hugely important.”* Whilst the participant considered the social model more important, she also identified some positives of the medical model, *“Some benefit to defining cause – families feel less pressure, less responsibility as they see it as part of phenotype.”* The participant also highlighted a tug of war type dynamic between the medical and social models and gave her view that the medical model was beginning to regain sway, *“What happened in society is that the medical model has existed for 100 years and social model only since 1970s, we're just beginning to have social model but pendulum might swing back to medical model.”*

Unlike participants on Factors 1 and 2, the Factor 3 participant agreed with statement 25 (+3, It is difficult to hold both a medical and a social model of intellectual and developmental disability) but they also strongly agreed with statement 31 (+3, The medical and social models of disability can and should be integrated to enhance the lives of people with intellectual and developmental disabilities).

3.7 Exemplary Statements Shared By Factors

There was some overlap in the positive and negative exemplary statements across the three factors. Statement 70, a consensus statement, was a negative exemplary statement on all factors. Reflective comments regarding this statement were similar for participants on Factors 1 and 3 who thought that overall both a diagnosis and presenting behaviour were important. Factor 2 participants however gave more weight to the presenting behaviour. Statement 21, another consensus statement, was a positive exemplary statement on Factor 1 and Factor 2 and for similar reasons.

Factors 2 and 3 shared all the same negative exemplary statements. In relation to statements 43 and 9 (consensus statements), participants on both factors accorded more relevance to environmental than genetic or biological variables in determining individual differences. Reflective comments explaining why statement 2 was strongly disagreed with showed differences between participants on Factors 2 and 3. The comments by Factor 2 participants showed a greater level of disregard for the medical model in comparison to the social model of disability than those by the Factor 3 participant who identified some clinical benefits of the former.

Chapter 4. Discussion

4.1 Discussion Overview

This chapter will discuss an interpretation of the findings of the Q methodology study. It will also consider the implications of these findings in relation to clinical practice. A discussion of the study strengths and weaknesses is then followed by a consideration of future research in the area.

The study aimed to explore clinical psychologists' attitudes to research emerging from the "New Genetics" that have a bearing on people with IDD. It sought to investigate views on this topic, including issues relating to genetic diagnoses and behavioural phenotyping, genetic screening and testing, aspects of the nature and nurture debate, causes of individual differences and the impact of genetic developments on service provision for people with IDDs, in order to examine the translation and application of such ideas in clinical practice.

4.2 Summary and Interpretation of Findings

In this study, three factors emerged from the Q-set representing the Q-concourse relating to genetic research and people with IDDs, with 27 of the 31 participants loading significantly on to one of these factors.

Factor 1, *Integration of social and medical models of disability*, represented the views of 14 participants, with a majority being trainee clinical psychologists. In addition, the qualified clinical psychologists loading on to this factor had fewer years' qualified experience working with people with IDDs than their counterparts loading on to the other two factors and the total overall group of qualified clinical psychologist participants. Due to its high participant loadings, this was a primary

factor. In working with people with IDD, the integrated use of principles of both the medical and social models of disability were deemed important and necessary. Participant reflections underscored the view that clinical psychologists should be able to work well within both models, drawing on each as appropriate. Consequently, diagnoses of behavioural syndromes were seen to add weight to clinical work by guiding assessments and interventions. If a diagnosis were available, participants on this factor viewed incorporating its implications in clinical work as important. Ultimately, however, the presenting needs of the client were deemed more important, regardless whether a diagnosis existed. Participants on the other two factors shared the view that environmental and contextual variables are more important in explaining behaviour than genetic factors; however, participants on this factor were more likely to endorse nature and nurture interactions. Advances in genetic testing and screening were also seen in a favourable light by Factor 1 participants.

Factor 2, *Social model of disability is more helpful*, represented the views of 12 participants, the majority of whom were relatively experienced qualified clinical psychologists. Several trainee clinical psychologists loaded on to this factor also and they too had more experience working with an IDD population relative to the overall group of trainee clinical psychologists. One of the main themes was the importance afforded to the meaning and function of a behaviour relative to the importance given to a genetic diagnosis. Understanding idiosyncratic behavioural differences in the context of the environment was thought to offer more in working with people with IDD than understanding the implications of a diagnosis. Indeed several negative beliefs regarding the utility of a diagnosis were stated. Overall, approaching clinical work with an IDD population from the perspective of a social model of disability

was vastly more encouraged than adopting the principles of a medical model of disability, which was thought to have little to offer in practical terms. The social model was viewed as offering a more holistic perspective by taking into account all factors that may impinge on behavior. Related to this was the view that environmental variables influence individual differences more than genetic or biological variables. Another aspect that distinguished this factor was the view that service provision should be based on clinical need and not on diagnosis. Points of debate on this issue were mentioned by participants, for example, that lack of a diagnosis may hinder access to services and that the level of differentiation within a diagnosis underscored the importance of looking at actual need.

Factor 3, *Genetic advances are positive but can create conflict with recognising the value of people with IDD's*, represented the view of an experienced qualified clinical psychologist. As it acquired fewer participant loadings than the other two factors, Factor 3 was considered a secondary factor. A main theme that distinguished this factor was an emphasis on genetic screening and testing. There seemed to be a push and pull relationship to this with the potential benefits of it recognised but also an acute awareness of the ethical implications it raised in terms of how it is used. Factor 3 also emphasised the need to protect people with IDD's from negative evaluations about their disability. These evaluations were associated with genetics and the medical model of disability. Despite some negativity associated with the medical model of disability, Factor 3 was actually the factor most in favour of diagnosis. Diagnosis was viewed as meaningful and helpful and there was agreement that service provision based on diagnosis rather than need was beneficial, a view not shared by participants on Factors 1 and 2. Understanding the individuality of people with IDD's as a product of environmental and social variables

and not just genetic make-up was a further theme in this factor. It appeared that this individuality was thought to be given better understanding by the Factor 3 participant within the social model of disability. This emphasis on recognising and respecting individuality seemed to be the reason behind the tension in this factor between acknowledging the potential benefits of genetic advances for people with IDD's and the expressed caution towards the medical model of disability.

The qualitative feedback indicating participant reasons for placing statements in the extremes of the scoring grid, i.e. the statements most agreed and disagreed with, informed the interpretation of factors. Although there was much agreement across factors, the qualitative responses showed subtle differences in the reasons for this agreement. The responses of Factor 1 participants showed their favorability towards integrating medical and social models of disability in working with people with IDD's whereas participants on Factor 2 were more likely to endorse the social model only. Participants on Factors 1 and 3 appeared to agree that the medical model is helpful in working with people with IDD's but reflective comments showed qualitative differences why participants held this opinion. Whilst Factor 1 participants appeared to promote the value of integration of models in therapeutic interventions, the Factor 3 participant highlighted the benefits of the medical model in terms of informing developmental trajectories through diagnoses alongside their firm view that the social model was more important in clinical work. Participants on Factors 2 and 3 strongly disagreed with all of the same statements; however, qualitative feedback showed that although both appeared to have very positive and strong opinions about working with people with IDD's in a holistic way, the views of the Factor 3 participant showed more openness towards the potential influential role played by genetics in behavioural outcomes. Examining the qualitative feedback also

gave insight into the issues of most importance for participants on each of the factors. The genetic testing debate appeared more relevant on Factor 3, whereas Factor 2 was most concerned with ensuring the stability of the social model in working with people with IDD, and for Factor 1, ways of utilizing the benefits of all explanatory models of IDD emerged as the most important issue.

4.3 Clinical and Service Implications for the Study

Understanding clinical psychologists' attitudes towards and beliefs about genetic research can provide insight into how they may incorporate such findings in their clinical work (Finucane et al., 2003). As outlined in the introductory chapter, this research is of increasing importance in services for people with IDDs.

Application of genetic information in the form of genetic screening and behavioural phenotyping has greatly advanced in the past number of decades and more rapidly since the completion of the Human Genome Project, but clinical services appear to have been slow to utilise this information (Hodapp & Dykens, 1994). Hypothesised reasons for this delay have included the low occurrence rate of some syndromes, the association of stigma with diagnostic labels, the inability of a diagnosis to inform practical concerns and the variability within diagnostic categories (Dykens et al., 2000). Greater inter-profession communication in recent years is thought to have increased practitioner application of genetic research (Hodapp & Dykens, 2012). Despite this assertion, the perspective of other researchers and anecdotal evidence suggests that adoption of genetic research remains variable in clinical and educational IDD services (Kuna, 2001; Lopez-Rangel et al., 2008; Reilly, 2012). Whilst behavioural syndromes of well understood phenotypes, such as Lesch-Nyhan and Prader-Willi syndromes are familiar to clinical psychologists and their

phenotypic information applied, the phenotypic information of other less well defined and/or less common syndromes is often overlooked. This study aimed to gauge an understanding of the views of clinical psychologists actively affiliated with services for people with IDD towards relevant genetic research. Several variables appeared to have an impact on participant views. In the next section, these are discussed in relation to their impact on clinical practice.

4.3.1 Variables that influence participant views. The findings of this study suggest that several variables may affect clinical psychologists' views about genetic research. The amount of experience with an IDD population was associated with a different pattern of response to the Q-set. More experienced clinical psychologists, both in terms of level of qualification and number of years' qualified experience, held views that were less favourable towards genetic research. In contrast, trainee and more newly qualified clinical psychologists were more receptive towards such ideas. The former group had a stronger affiliation with a social model of disability that views the broader context in which a disability occurs as more informative than a genetic or syndromal explanation. The latter group were more inclined to promote an integration of models. Several explanations could account for this difference. Firstly, it could be an issue of timing and training whereby more recently trained clinical psychology cohorts have had greater exposure to information about genetic research findings and their interactions with IDD. More experienced psychologists who trained, prior to the Human Genome Project, at a time when normalisation and social role valorisation theories dominated the field, may not endorse genetic research in the same way. Statements in the P-set relating to access to up to date research information indicated that this was hampered

in resource-pressured services, which adds further weight to the argument that information sharing in practice is not carried out with consistency.

Secondly, these differences may reflect experience, in that more experienced clinical psychologists have a greater awareness of what works and is useful and adapt their practice accordingly. Experience could prove that in working with an IDD population, interventions embedded in a social model of disability are more effective than using phenotypic information. This may also relate to the reality that a diagnosis frequently does not exist for an individual (Rauch et al., 2006) and therefore other factors must be found to explain behaviour and shape interventions. If this is the case, it is easier to understand the perspective of Factor 2 participants, as how can clinical psychologists appreciate the benefits of genetic research if they do not have reliable experience of using genetic information in the first place.

The researcher (C. V.) noted that the majority of Factor 1 participants had trained or were training at a specific university. Thus, a third reason that may explain the discrepancy in responses between Factors 1 and 2 could be external to clinical experience and connected to the emphasis in the academic IDD teaching they received. If so, a way of ensuring better use of genetic information by clinical psychologists would be to consider more systematically using training as a way of imparting this information. As accessing current research appears more difficult at a service level sharing it at a training level seems more appropriate.

Personal experience of genetic diagnoses appeared to have been a further factor that affected response patterns. This appeared most clearly on Factor 3 where the participant that defined this factor intimated a history of genetic disorder in her family. Having this experience was associated with a more favourable attitude towards diagnosis. This was congruent with research discussed in the introduction

chapter regarding the value bestowed on a diagnosis by people with IDD and their family members (Costain et al., 2012; Lenhard et al., 2005; Statham et al., 2010; Trottier et al., 2013). This experience also seemed to generate a greater awareness or thoughtfulness about the impact of genetic advances such as genetic testing and screening. Although this was of concern to all participants, it was given greater relevance on Factor 3. This was evidenced by the higher number of statements pertaining to this issue being placed in the extremes of the scoring grid.

The subsequent section provides a discussion of how the differing views of genetic research among clinical psychologists may affect and inform clinical practice.

4.3.2 The impact of participant views on clinical practice. The variables affecting clinical psychologists' views of genetic research likely have an impact on whether this research is adopted and applied in services for people with IDD. A service staffed by Factor 1 participants would likely function differently to one staffed by Factor 2 participants. In this section, a discussion of how these participant views may serve to hinder or support application of genetic research is outlined. Attention is given to genetic diagnoses of behavioural syndromes and genetic screening and testing as these were the two main areas of genetic research highlighted in the introduction chapter as affecting people with IDD.

The pattern of sorting of statements pertaining to the medical and social models of disability was different for participants on Factors 1 and 2. It seemed that Smith's (1994) assertion that professionals working with an IDD population seek to create distance from the medical model was more accurate for the more experienced psychologists on Factor 2. These participants trained in an era with greater reticence towards medical interventions than newer cohorts of clinical psychologists and this

may have had an impact on the views that emerged in this study. Indeed, during the Q-sorting process several participants highlighted the so-called “political correctness” of the clinical psychology profession, as though there was a perception or idea as to how a clinical psychologist should respond, with an implicit presumption that clinical psychologists working in IDD services would take a particular stance with regard to the “medical model”. This may in turn constitute a barrier to the implementation of genetic research but one that may necessarily change as new cohorts, with different values, enter the profession. However, it must be noted that Factor 2 was also comprised of several trainee clinical psychologists, indicating that the steadfastness of negative views of the medical model of disability continue to linger despite progress within the model.

Awareness of the ethical issues surrounding genetic research was evident in the responses of all participants. Whilst this was particularly more prescient for Factor 3, participants on the other two factors also showed this awareness. For example, Factor 2 participants highlighted the importance of contextual variables, which they felt should not be overshadowed by genetics and Factor 1 participants’ engagement with the debates regarding genetics was illustrated by some of their exemplary statements. As suggested in the introduction, there was an apparent need among psychologists to protect people with IDDs from potential sources of discrimination and this seemed to relate to fears regarding genetic advances. This concern is unarguably well founded in relation to the rapid development in genetic screening, which has not been matched in the development of policy, guidance and regulation (Muir, 2000). However, it could be argued that, although well intentioned, this concern is not as appropriate in relation to diagnoses of genetic syndromes. As noted in the introduction and via comments from the present study, such a diagnosis

is generally welcomed by people with IDD and their families. For a clinical psychologist to moderate the importance of a diagnosis may undermine this need. Even if they themselves do not see its clinical relevance, it is perhaps therapeutic to understand this with the client. Taking a stance against a diagnosis may appear quite paternalistic, in that it implies the psychologist knows best.

In relation to the rights to and ethics of genetic screening and testing, there appeared to be some ambiguity towards this among all participants in this study. Evidence for this came from the placement of statements reflecting this issue in the neutral categories of the consensus table (see Appendix 11). This finding is perhaps reflective of wider society in which the advances of genetics are occurring so rapidly that it is difficult to maintain a stable understanding of their ethical implications (Baker et al., 2012; Bessa et al., 2012; Muir, 2000). At a clinical service level, ambiguity about this issue may underscore a need for more discourse and debate among professionals about the impact of genetic screening and testing. Unclear or negative views may act a barrier to people with IDD being directed to relevant genetic tests, which could potentially positively affect their lives. Robertshaw and MacPherson (2006) argued that genetic testing for adults with IDD is underused. Greater discourse and debate about this issue may give professionals opportunity to reflect more on their personal opinions about genetic testing and determine how compatible these are with the services in which they work. Including people with IDD in this debate may also serve to show the personal meaning of a diagnosis and perhaps challenge any perceived stigma attached to a diagnosis by professionals.

Exposure to a referent issue or object can influence attitudes (Allport, 1954), and the more contact with the issue or object, the more positive attitudes become. This has been found in relation to attitudes toward people with IDD (McManus,

Feyes & Saucier, 2010) and the same process may be at work with regard to the attitudes of professionals towards genetic research. As shown in this study, personal experience of a genetic diagnosis was associated with a more positive attitude towards diagnosis. Following this logic, if genetic advances are helpful in working with people with IDD then more frequent exposure to them and experience of their benefits should improve practitioners' attitudes towards them. Services addressing early identification of dementia in Down syndrome are perhaps a good example of how the discovery of a genetic link can be used to define service provision (Janicki & Dalton). In the future, the increasing identification of genetic etiologies of IDDs and their associated phenotypes should be reflected in practice providing more opportunities for clinical psychologists and other professionals to experience the value of genetic information. According to Hodapp and Dykens (2012), this process is already underway. This study has shown some support for this assertion, however, the variability among the P-set in their acceptance of the utility of diagnoses indicates that this is not universal. It is likely that genetic research will need more time to prove its worth before better acceptance of it among professionals aligned to a social model of disability.

Although it could be argued, that research showing the effectiveness of interventions based on behavioural phenotypes should be shared with clinical services as a way to challenge negative views about the utility of diagnoses, there is also a need to understand why these views remain strong among experienced clinicians. For example, are these views simply reflective of a disconnect between research and practice or has application of diagnostic information proven to be ineffective. Alternatively, diagnostic information may be experienced as useful but not as useful relative to other factors or not as useful in all cases. Respect should be

paid to these views to understand more fully potential barriers to the application of genetic research. As well as an affiliation with a social model of disability, this thesis has identified inconsistent sharing of research findings, limited experience of rare behavioural syndromes and a professional responsibility to protect people with IDD from any negative scientific repercussions among potential barriers to the application of genetic research. However, several other barriers are likely to exist and the fact that a diagnosis may have limited clinical utility in many cases should not be ruled out. If this is so, then the views of the more experienced psychologists in this study could be considered realistic relative to the idealistic views of the less experienced psychologists. It may also indicate a need for sensitivity in integrating rather than imposing clinical interventions associated with a medical model of disability in services and with professionals aligned to a different way of working with people with IDD. As Hodapp and Dykens (2012) identified, communication and sharing of research and views between professionals is the optimum way to ensure the application of relevant findings.

4.4 Study Limitations and Strengths

The rationale for choosing Q methodology for this study was that it is a useful tool with which to identify alternative values and viewpoints. As participants are asked to react to statements about the topic rather than articulate themselves as in an interview format, views emerge that may not otherwise have. In addition, in Q methodology, the sorted statements are not analysed as separate items of information but rather in terms of their mutual coherence for the respondent (Brouwer, 1999). These characteristics were congruent with the aim of the present study, which sought opinions on a topic not well researched in the area but likely to generate varying

views. This section outlines the strengths and limitations of the methodology for this study.

4.4.1 Study limitations. During the Q-sort process, several participants reported finding certain statements confusing and ambiguous. Participant comments pertained to the readability and clarity of statements. Negatively worded statements and complicated and ambiguous terminology were reasons given for these comments. Another issue raised by participants was a lack of definition for terms or phrases contained in statements. Although these issues referred to a limited number of statements, participants' interpretations of them may have varied as a result and it is difficult to quantify the impact this had on the study outcome. The pilot study had aimed to address these precise issues and it had resulted in several changes to statement wordings and the creation of an "explanation of terminology" leaflet. This feedback from participants however, indicates that the pilot study missed some important points of confusion. Four trainee clinical psychologists were included in the pilot study. In light of the differences found between trainee and qualified clinical psychologists, perhaps the pilot would have been more comprehensive had it included representatives from the qualified clinical psychologist group also. It is likely that this would have eliminated more sources of confusion in the statements. This underpins the importance of completing a pilot study with representatives from all participant groups in a study.

As outlined in Chapter 2, there were several themes built into the Q-set, which were reflective of a broad study topic. During the sorting process, a number of participants highlighted this and stated that it was difficult to sort the statements as they wanted to agree with many of the matters within these themes. The higher number of statements in the "agree" pile relative to the "disagree" pile in the initial

Q-sorts (see Table 2 in Chapter 3) illustrated this dilemma. The researcher (C. V.) encouraged participants to sort statements according to their strength of feeling about the issues raised by each statement. As this dilemma was raised by a substantial number of participants, this guidance was added to the study protocol but it highlighted some pertinent issues that may have limited the study. First, it showed how the forced-choice sorting method possibly resulted in participants having to give a neutral or negative rating to statements that they had agreed with. It also infers that the breadth of the study topic may have been too wide, which restricted those participants who wanted to agree with more of the themes raised by the topic. A more focused Q-concourse, broken down into its constituent themes such as, diagnosis and behavioural phenotyping etc. and presented as individual Q-sorts might have been more appropriate. Considering these themes holistically within the broad umbrella of “New Genetics” allowed for the level of importance attached to each theme to emerge in the factors, for example, the importance attached to genetic screening and testing on Factor 3 relative to the other factors. As an initial foray into this study area, perhaps obtaining this level of information was an important first step.

A frequently cited limitation of Q methodology is its lack of generalisability; however, as with qualitative methods in general, Q methodology is less concerned with generalising findings and more concerned with describing phenomena (Krefting, 1991). Also, due to the operant nature of study topics, reliability is less of a concern as this only becomes important when a test purports to measure a stable attribute. As Q methodology does not involve such concepts, reliability is therefore not of importance given that the purpose of Q methodology is to provide a continuum of perspectives regarding a specific topic and not for it to be

generalisable (Stenner & Marshall, 1995). However, it would be interesting to determine whether the viewpoints that emerged in this study are common among the wider population of clinical psychologists.

There are several strategies used in the selection of statements for the Q-set. These have been described as being structured, for example, drawing statements from existing measures or unstructured whereby all items thought to be of relevance are added (McKeown & Thomas, 1988). If using an unstructured method McKeown & Thomas (1988) stated that researcher bias may inadvertently be introduced as you cannot conclude that all attitudes on a topic have been elicited. The unstructured method used in this study may thus have served as a limitation to this study. However, Robbins and Krueger (2000) further argued that even if the Q statements are directly obtained from participants this does not mean that the potential for researcher bias is fully eliminated, as it is the researcher that guides participants in the research interview.

4.4.2 Study strengths. An advantage of this study was its ability to engage participants. During Q-sorting the vast majority of participants commented that the process had interested them and that the topic had been thought provoking. This gives justification to the rationale of using this methodology, as it is likely that had an alternative research method been used certain views, prompted by the “thought provoking” statements, would not have been captured.

Although potential researcher bias in terms of statement selection was described as a study limitation in a previous paragraph, in terms of the actual Q-sorting process researcher bias is minimised as the data is generated by participants who have an interest in the topic rather than by the researcher (Barry & Proops, 1999). This is an advantage of Q methodology inherent in its design. Researchers

acknowledge and present the reality of constructions of different individuals without insisting on a superior objective perspective of the researcher's own construction of reality (Kitzinger, 1986). Participants reveal what the topic means to them through their structuring of the data. Thus, this is a strength of this study and Q methodology more generally.

As well as defining the structure of subjectivity, Q methodology examines the relationship between phenomena and subjective interpretation (Robbins & Krueger, 2000). In doing so, Q methodology can explain why things are important as complex divisions and linkages between and among disparate individuals can be exposed. However, it goes beyond describing subjectivity, it evaluates and makes comparisons between different viewpoints. This means it can usefully explore tastes, preferences and attitudes. The nuances in the different viewpoints explored in this study exemplifies this advantage.

Sorting the Q-set is a novel administration method that requires the active participation of the research participant. As a result of this active participation, it is rare to have missing data and undecided responses (Dennis, 1986). Donner (2001) has noted that participants want to see their opinions translated into factors and quantified. Furthermore, the ranking of the statements during the Q-sort requires participants to make fine discriminations they otherwise might not make (Dennis, 1986). In other research methods, such as Likert scales, participants are asked to indicate their levels of agreement on a range of statements. However, an advantage of Q methodology over other methods such as survey methods is that participants have to identify their level of agreement with a statement in relation to all the other statements (Donner, 2001). This allows inferences to be made about the relative importance of different issues in the Q-set as well as allowing the researcher to

clarify the range of constructs present in viewpoints on the research topic (Barbosa et al., 1998).

4.5 Recommendations for Future Research

This study represents a preliminary step in understanding clinical psychologists' attitudes to an extremely complex topic. Extension of this study with a further sample of clinical psychologists is warranted. This would serve to delineate further what influences attitudes; clinical work or clinical training experience or personal circumstances, factors identified as influencing attitudes in this study. Further replication would lend more weight to these findings. Completing the study with psychologists working and training in other parts of the UK would also inform as to whether service cultures differ across geographical areas and how this might influence views about genetic research.

Services for people with IDD are typically staffed by several professional groups, such as nurses, psychiatrists and speech and language therapists. All of these professionals have cause to engage with genetic developments affecting service-users. Running this study with a sample of these professional groups would be useful in determining whether conflicting or concordant attitudes towards genetic developments exist across staff groups, which might provide an insight into service provision for people with IDD.

In completing this study, certain demographic factors such as clinical experience were shown to mediate viewpoints. Through the course of Q-sorting other factors were found to have a bearing on attitudes, for example, personal experience of a genetic diagnosis. As the research did not directly seek to determine the influence of these more happenstance factors, more systematic study of their

influence would usefully inform the study aims. Perhaps, openly asking participants whether they thought any personal or professional factors influenced their Q-sort would allow for this more informed insight into the formation of attitudes and would be worth incorporating in future studies of a similar nature.

4.6 Personal Reflection

In conducting research in the area of IDD, the researcher (C. V.) was conscious of the interest of people with IDDs in being involved in research studies that have implications for them (Ramcharan & Grant, 2001). In addition, with regard to genetic research, Miller and Levine (2013) have argued that genetic advances will suffer without the involvement of people with IDDs. Thus, the researcher (C. V.) was conscious that a lack of input from people with IDDs in the current study was a limitation, as it seemed to go against the principle of participation, widely adopted by disability organisations, “Nothing about us, without us” (Charlton, 1998). Although the attitudes of clinical psychologists towards genetic advances were of interest in this study, on reflection, the researcher wondered whether having input from people with IDDs at some stage in the process might have strengthened the study design and its findings. Although the principle of inclusion is of importance to the researcher (C. V.), she would not have included people with IDDs in the study merely to acknowledge this principle, but to understand the opinions of people with IDDs towards genetic developments affecting their lives. She reflected that interviews with people with IDDs would perhaps have been a useful addition in the development of the Q-concourse.

4.7 Conclusion

Using a Q methodology design, this study identified three viewpoints among clinical psychologists about research from the “New Genetics” that have implications for people with IDD. These viewpoints indicated varying levels of support and acceptance of this research. One strong viewpoint advocated for the integration of this research in clinical practice, another showed more scepticism towards what it could offer in practical terms. A third viewpoint, advocated by fewer participants, showed appreciation for the research but also caution towards its application. Factors associated with this variability were level and amount of clinical psychology experience, training experience and personal experiences in relation to aspects of the study topic. Suggestions for future research include replicating the study with a wider sample of both clinical psychologists and other professionals working in services for people with IDDs to extricate further how personal and professional ideologies may affect clinical practice, and as a consequence the lives of people with IDDs. Consideration of the views of people with IDDs about this topic would also determine the compatibility of attitudes between professionals and service users, which would likely enhance the cohesion of service delivery. This study represents a first step towards understanding the attitudes of a professional group towards the “New Genetics” and its burgeoning influence on people with IDDs.

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Appendix 1: List of Q-concourse Books and Journals

Books

Genome: The autobiography of a species (M. Ridley, 2000)

Lifelines (S. Rose, 2005)

Nature via Nurture: genes, experience and what makes us human (M. Ridley, 2003)

The Blank Slate (S. Pinker, 2002)

The Extended Phenotype (R. Dawkins, 1989)

The Language Instinct (S. Pinker, 1994)

The Language of the Genes (S. Jones, 1993)

The Selfish Gene (R. Dawkins, 1976)

Journals

British Journal of Learning Disabilities

Journal of Applied Research in Intellectual Disabilities

Journal of Intellectual Disability Research

Appendix 2: Participant Information Sheet and Consent Form for Q-concourse Interview Participants

Use of Q-methodology to identify clinical psychologists' attitudes towards genetic research affecting people with intellectual and developmental disabilities

Participant Information Sheet

You are invited to take part in a study aiming to understand clinical psychologists' attitudes towards and beliefs about recent research developments and ideas that impact on people with learning and developmental disabilities.

Please take time to read the following information carefully. You can contact the study researcher if there is anything that is unclear or if you would like further information (please see contact details towards the end of this information sheet).

Who will conduct the research?

The research is being conducted by Catherine Vahey, a student on the University of Manchester Clinical Psychology Doctorate Programme under the supervision of Drs. Dougal Hare and Anja Wittkowski.

What the research hopes to achieve

This research will explore the attitudes and beliefs of clinical psychologists towards recent research developments that affect people with intellectual and developmental disabilities (IDDs). In particular, opinions on new developments and ideas in the field of genetics will be explored.

The research hopes to examine the influence and spread of such research and ideas and examine whether there are barriers, both personal and institutional, to their wide-spread adoption in services.

The views of both qualified and trainee clinical psychologists will be obtained. As well as looking at the overall group beliefs and attitudes, differences and similarities between the two groups will be examined to determine whether level and amount of experience has a bearing on opinions.

The research ultimately aims to inform and improve services for people with learning disabilities through greater understanding of the attitudes and beliefs of those in positions to shape and influence service provision.

What would I be asked to do if I took part?

The study researcher will aim to accumulate all possible opinion statements on the study topic i.e. opinions regarding new developments in research of relevance to

IDD. As a participant you would be asked to provide your opinions on the study topic.

There are no known expected discomforts or risks involved from participation in this study.

What demographic and personal details will I be asked to provide?

Your **full name**. Your name will not be identified on any study data or study publications. We are ethically bound to detail your name on the study consent form and this is the only study document on which it will appear.

Your email address will also be used to issue a summary of the findings at the end of the study should you wish to receive this.

What happens to the collected data?

A number of people involved in research and clinical practice in IDD, including you, will be asked for their opinions on the study topic. The accumulated opinions from the interviews and opinions drawn from relevant literature will be used to form a set of statements. Qualified and trainee clinical psychologists will then be asked to rate their level of agreement or disagreement with these statements. Analysis of these ratings will aim to show the broader attitudes and opinions towards genetic and neuroscience research into IDD.

How is confidentiality maintained?

Each participant will be identified by a study number. All data will be identified by study number only, therefore, your responses will be anonymous.

The data will be kept confidential; this means that except for the researcher and her supervisors no one will be able to view the data. The computer files that will be used for analyses will not contain any personally identifiable information; participants will be only identified by their study number. Files will be password protected and stored on an encrypted laptop and in a secure folder on the University of Manchester's network.

What happens if I do not want to take part or if I change my mind?

It is up to you to decide whether or not to take part. If you do decide to take part you will be given this information sheet to keep and be asked to sign a consent form. If you decide to take part you are still free to withdraw at any time without giving a reason and without detriment to yourself.

Will I be paid for participating in the research?

No payment will be provided for participation.

Will the outcomes of the research be published?

The data will be published in a doctoral thesis and in peer reviewed scientific journals. The outcomes may also be reported at conference presentations.

Contact for further information

Should you wish to enquire further about the research you can contact the study researcher, Catherine Vahey, by email (catherine.vahey@postgrad.manchester.ac.uk).

You may also contact the study supervisors Dr. Dougal Hare (dougal.hare@manchester.ac.uk) and Dr. Anja Wittkowski (anja.wittkowski@manchester.ac.uk).

What if something goes wrong?

The research team will endeavour to ensure that your participation in the research is fully and appropriately supported, however, should you wish to make a formal complaint about the conduct of the research you should contact the Head of the Research Office, Christie Building, University of Manchester, Oxford Road, Manchester, M13 9PL.

Appendix 3: Final Q-set (post pilot study)

1. Diagnosing different intellectual and developmental disabilities does not lead to greater understanding or change
2. The medical model of disability is more helpful than the social model of disability in intervening with people with intellectual and developmental disabilities
3. Sociocultural factors such as socioeconomic status and family background are more important than behavioural phenotyping in explaining behaviour
4. Growing up in the same family has no discernible or marked effect on the IQs of siblings
5. Intellectual ability is affected more by nature than by nurture
6. Humans are not born with innate tendencies; experience in the environment shapes all learning
7. Behavioural syndromes rarely occur
8. The cause of intellectual disability can be of equal or greater importance than the immediate and broader environment as a determinant of well-being
9. Genetically inherited traits are fixed and non-modifiable
10. Genetic screening/testing causes negative attitudes towards disability to pervade and continue
11. What's not important is genetic screening but the information it yields and how that is used
12. Awareness behavioural phenotyping and its developments is generally of a low level among staff
13. The social model of disability is more helpful than the medical model of disability in intervening with people with intellectual and developmental disabilities
14. Understanding the meaning or function of an idiosyncratic behaviour is more important than understanding the diagnostic label
15. Understanding the diagnostic label informs idiosyncratic formulations, interventions and treatment plans

16. The pathway to the same behaviour may be different for individuals with different genetic disorders
17. Impact of genetic screening on the individual and society should be considered before it is used
18. Many professionals working with individuals with intellectual and developmental disabilities are unconcerned with why someone has the impairment
19. Diagnostic labels can serve to deny people access to services
20. Service provision should be provided primarily on the basis of need not diagnosis
21. Challenging behaviour needs to be considered in the wider context of family and socioeconomic background, it is unethical to work with people with challenging behaviour and ignore this context
22. Focusing on the individual's expressed difficulties is more helpful than looking at the difficulties in the context of a diagnosis or conflicting diagnoses
23. Associating genetics with people with intellectual and developmental disabilities can undermine the progress of the social model of disability
24. The environment that a child experiences is as much a consequence of the child's genes as it is of external factors: the child seeks out or creates his or her own environment
25. It is difficult to hold both a medical and a social model of intellectual and developmental disability
26. Diagnosis specific services presume specialisation which fits with a medical model
27. Heterogeneity of presentations within diagnostic categories can render the diagnosis meaningless
28. Genetic screening can lead to negative social engineering with the creation of 'designer' societies where people with disabilities are undervalued and social/environmental influences on disability undermined
29. Inclusion in society can be enhanced by understanding individual difference that can be traced to a specific genetic disorder
30. Behavioural phenotyping can make a positive impact on quality of life for people with intellectual and developmental disabilities if the information is used appropriately

31. The medical and social models of disability can and should be integrated to enhance the lives of people with intellectual and developmental disabilities
32. Diagnosing intellectual and developmental disabilities/behavioural syndromes can hinder and create barriers to therapeutic work
33. The cause of the intellectual and developmental disability is unknown in most cases
34. Genetic disorders lead to different behavioural outcomes
35. Families usually want to know the cause of their child's intellectual and developmental disability
36. Health and social care resources should be directed towards understanding and improving social and environmental factors affecting people with intellectual and developmental disabilities
37. Health and social care resources should be directed towards disseminating and applying research, originating from all fields of relevance, affecting people with intellectual and developmental disabilities
38. The environment has less of an impact in an equal society than it does in a more unequal society
39. Genetic screening may reduce complex behaviour to genes, ignoring the impact of other factors on behaviour
40. A diagnostic label is more helpful to an individual than it is unhelpful
41. For disorder-specific services to be effective for the individual there must be no doubt in the accuracy of their diagnosis
42. Genetic screening, and its consequences, masks and denies the individuality and opportunity in people with intellectual and developmental disabilities
43. What happens in the womb influences intelligence more than anything that happens after birth
44. Genetic screening creates a battle between innate social instinct versus human rights
45. Defining services by diagnostic labels means better, more individually tailored services are delivered
46. Geneticists have a moral and ethical responsibility to the today not the tomorrow
47. No one knows what the non-genetic causes of individuality are

48. Innate abilities allow children to develop typically, absence of such abilities affects development
49. Genetic testing would not add to the quality of life for a person with an intellectual or developmental disability
50. Labelling genetic disorders/behavioural syndromes may negatively affect prognosis
51. Positive behaviour support is more influential in improving the quality of life for a person with an intellectual or developmental disability than the outcomes of behavioural phenotyping and genetic testing
52. Genetic screening and behavioural phenotyping are akin to modern day eugenics
53. The non-shared environment, such as individual school experiences, account for more differences in siblings than genes
54. Genetic screening/testing should be available to all as people have a right to information about their genetic make-up
55. The shared environment has a greater influence on sibling similarities and differences than genes
56. Genetically inherited traits/characteristics can be subject to change and adaptation by the environment - Heritability does not mean immutability
57. Degree of intellectual disability (mild, moderate, severe, profound) is a better predictor of behavioural outcomes than behavioural syndrome diagnosis
58. Genetics and behavioural phenotyping represent a shift backwards to the medical model of disability
59. Genetic aspects of a condition may be viewed as irrelevant or potentially negative by professionals/staff members
60. The science of intelligence testing is flawed
61. Genetic screening, if used in an appropriate, responsible way, has the potential to positively affect lives
62. Genes have a greater influence on sibling similarities and differences than the shared environment
63. Understanding the causal pathway to an individual difference can have a positive influence on well-being

64. Culture is the product of individual psychological make-up rather than vice versa – a person does not inherit cultural knowledge, they acquire it through experience
65. Genes have a continuing influence on individuals as they develop
66. Focusing on how genetics influences behaviour downplays the role of more important social influences
67. Diagnosing different intellectual and developmental disabilities/behavioural syndromes provide unnecessary labels and can create stigma
68. As people get older their environment has a stronger influence on their behaviour
69. The social environment is the product of individuals' innate social instincts
70. The diagnosis is more meaningful than the presenting behaviour
71. Generic services beneficial to all is the ideal but if this is unattainable disorder-specific services should be preferred
72. People with different behavioural syndromes have more similar than dissimilar behaviours
73. Behavioural phenotyping is beneficial in understanding some syndromes such as Down syndrome but this is an exception, behavioural phenotyping does not generally aid understanding as much as other factors
74. Current knowledge and understanding of different disorders is too limited to make disorder-specific services worthwhile
75. There are no direct genes for intelligence but an inherited resistance to stressors e.g. resistance to toxins which then enhances the ability to develop intelligence
76. Robustness of the conceptualisation of disorders/disabilities needs to be improved e.g. autism, otherwise the science (phenotyping) on which it is based is flawed and any predictions made on this basis are flawed and potentially destructive
77. The voice of people with intellectual and developmental disabilities in the genetic testing debate is unheard
78. In clinical practice it is difficult to keep up to date on new research developments due to time and service pressures

79. Research of relevance to people with intellectual and developmental disabilities is often difficult to access by services which affects the application of new research
80. Social instincts may mean it is natural to exclude people with intellectual and developmental disabilities (IDDs) from the group; however, our human side and social responsibility should cause us to fight against this and recognise the value to the world of people with IDDs and diversity in general
81. A partially inherited low IQ might be subject to extensive improvement through education

Appendix 4: Written Explanation of Terms in Q-set

Explanation of terms:

Behavioural Phenotyping

Concerns the distinction between one's genetic makeup (genotype) and one's ultimate physical characteristics (phenotype). It implies connections between one's genes and one's behaviour. Behavioural phenotype can be viewed as the expression of the genes on the environment and the interaction of the two.

Behavioural syndromes/genetic disorders

Syndrome: a group of signs and symptoms that, when they occur together, suggest the presence of a disorder or condition

Disorder: a medical condition involving a change to the usual functioning of the mind or body

These terms have been used together or interchangeably in these statements to reflect the often unknown cause of an intellectual or developmental disability.

Genetic screening and genetic testing

Genetic screening and testing are essentially the same thing but differ in why they are carried out.

Genetic screening is typically used with someone who is in an at risk population for developing a certain condition, for example, being screened for a condition that commonly affects people of your age or gender.

Genetic testing is typically used with someone who has a known familial risk of carrying genes linked to a specific genetic condition.

These terms are used together or interchangeably in these statements.

Intellectual and developmental disabilities

These statements use the terminology "intellectual and developmental disabilities" in reference to people who have a learning disability and/or a developmental disability such as autism.

Appendix 5: Q-sort Narrative Instruction Script

Instruction Script for Q sorting

Take the set of cards, the guide bar and score sheet. Lay the guide bar in front of you. There are 81 cards and each contains a statement with a viewpoint about research developments and ideas pertaining to people with intellectual and developmental disabilities. The statements are drawn from research literature, books and interviews with psychologists involved in research or services for people with intellectual and developmental disabilities. I would like you to arrange these statements according to your own point of view.

The question to consider when arranging the cards is “To what extent do you agree or disagree with the viewpoint expressed on each statement”. It is important that you sort according to your beliefs and personal experiences of working with an IDD population.

Please read each statement carefully and using the guide bar divide them into three piles. Place to the right those which you agree with, place to the left those which you disagree with and place in the middle statements which you are either neutral, uncertain or ambivalent.

Please count the number of cards in each pile and write that on your answer sheet. The total should be 81.

Take the cards from the agree pile and read through them again. Next select the four statements that you most agree with and place them on the right side of the score sheet below the number “4”. It does not matter which card is placed on top or bottom of the column; all statements in the same column will receive the same score. Next, select six statements that you most agree with and place them below the number “3”. Then select ten statements that you most agree with and place them below the number “2”. Please repeat this until you have placed all you cards in the agree pile. The correct number of statements in each column is indicated in the brackets at the foot of each column on the score sheet, please stick to this.

Now take the cards from the disagree pile and read through them again. Just as before, select the four statements that you most disagree with and place them on the left side of the score sheet below the “-4”. Please repeat this until you have placed all the cards in the disagree pile.

Finally read through the neutral cards and again place them in the remaining empty columns of the score sheet. Here you should attempt to place those with which you agree in the “+” area and those which you disagree in the “-“ area.

When you have finished placing your cards in the score sheet please review your distribution and once more feel free to swap around any cards.

If you are happy with your distribution I will copy the number that appears on the back of each card onto the corresponding boxes on the score sheet.

I would like to ask you some brief questions on your experience of the Q-sort process

- Why did you most agree with the four statements that you placed below the “4”?
- What did you most disagree with the four statements that you placed below the “-4”?
- Any other general feedback about the process?

Can I obtain some demographic details, namely:

The number of years you have worked in services for people with intellectual and developmental disabilities in any capacity?

The number of years you have worked in such services since qualifying as a clinical psychologist?

Thank you

Appendix 6: Guide Bar

Disagree	Neutral	Agree

Appendix 7: Participant Information Sheet and Consent Form for Study Participants

Use of Q-methodology to identify clinical psychologists' attitudes towards genetic research affecting people with intellectual and developmental disabilities

Participant Information Sheet

You are invited to take part in a study aiming to understand clinical psychologists' attitudes towards and beliefs about recent research developments and ideas that impact on people with learning and developmental disabilities.

Please take time to read the following information carefully. You can contact the study researcher if there is anything that is not clear or if you would like further information (please see contact details towards the end of this information sheet).

Who will conduct the research?

The research is being conducted by Catherine Vahey, a student on the University of Manchester Clinical Psychology Doctorate Programme under the supervision of Drs. Dougal Hare and Anja Wittkowski.

What the research hopes to achieve

This research will explore the attitudes and beliefs of clinical psychologists towards recent research developments that affect people with intellectual and developmental disabilities (IDDs). In particular, opinions on new developments and ideas in the field of genetics will be explored.

The research hopes to examine the influence and spread of such research and ideas and examine whether there are barriers, both personal and institutional, to their wide-spread adoption in services.

The views of both qualified and trainee clinical psychologists will be obtained. As well as looking at the overall group beliefs and attitudes, differences and similarities between the two groups will be examined to determine whether level and amount of experience has a bearing on opinions.

The research ultimately aims to inform and improve services for people with learning disabilities through greater understanding of the attitudes and beliefs of those in positions to shape and influence service provision.

What would I be asked to do if I took part?

The study researcher will aim to accumulate all possible opinion statements on the study topic i.e. opinions regarding new developments in research of relevance to

IDD. As a participant you would be asked to read through each of the statements, decide whether you agree or disagree with them and then rank your level of agreement/disagreement with them, options will range from 'strongly agree' to 'strongly disagree'. There are no right or wrong statements, each will simply express a different viewpoint.

This process is called a Q-sort and is part of Q-methodology. The study researcher would administer the Q-sort individually with you. After completing the Q-sort you would also be asked for brief feedback about your experience of the process.

There are no known expected discomforts or risks involved from participation in this study, however, you would have an opportunity to debrief with the study researcher, following administration of the Q-sort, to address any potential discomforts.

What is the duration of the research participation time?

The length of time taken to complete the Q-sort will have much individual variation; however, it is likely that most people will complete it within 25-60 minutes.

Subsequent feedback about the process should take no longer than a few minutes.

What demographic and personal details will I be asked to provide?

Your **full name**. Your name will not be identified on any study data or study publications. We are ethically bound to detail your name on the study consent form and this is the only study document on which it will appear.

A contact **email address**. Your email address will be used to arrange the administration of the Q-sort. Your email address will also be used to issue any additional study information; including a summary of the findings at the end of the study should you wish to receive this.

What happens to the collected data?

Once the Q-sort data has been collected from all participants it will be transferred to a computer file. The data in the computer file will be analysed statistically. Demographic data will be transferred to a separate computer file and analysed in terms of averages and frequencies.

The brief qualitative feedback about the Q-sort process will be transferred to a password-protected word document and any written records securely stored in a filing cabinet at the University of Manchester. Any hardcopy information pertaining to the Q-sort and demographic data will also be stored in this cabinet.

How is confidentiality maintained?

Each participant will be identified by a study number. All data will be identified by study number only, therefore, your responses will be anonymous, however, for the

purpose of analysis participants will be identified by their group i.e. qualified clinical psychologist or trainee clinical psychologist.

The data will be kept confidential; this means that except for the researcher and her supervisors, no one will be able to view the data. The computer files that will be used for analyses will not contain any personally identifiable information; participants will be only identified by their study number. Files will be password protected and stored on an encrypted laptop and in a secure folder on the University of Manchester's network.

What happens if I do not want to take part or if I change my mind?

It is up to you to decide whether or not to take part. If you do decide to take part you will be given this information sheet to keep and be asked to sign a consent form. If you decide to take part you are still free to withdraw at any time without giving a reason and without detriment to yourself.

Will I be paid for participating in the research?

No payment will be provided for participation.

Will the outcomes of the research be published?

The data will be published in a doctoral thesis and in peer reviewed scientific journals. The outcomes may also be reported at conference presentations.

Contact for further information

Should you wish to enquire further about the research or indicate your wish to participate you can contact the study researcher, Catherine Vahey, by email (catherine.vahey@postgrad.manchester.ac.uk). You may also contact the study supervisors Dr. Dougal Hare (dougal.hare@manchester.ac.uk) and Dr. Anja Wittkowski (anja.wittkowski@manchester.ac.uk).

What if something goes wrong?

The research team will endeavour to ensure that your participation in the research is fully and appropriately supported, however, should you wish to make a formal complaint about the conduct of the research you should contact the Head of the Research Office, Christie Building, University of Manchester, Oxford Road, Manchester, M13 9PL.

What happens next if I decide to participate?

After signing the consent form you may contact the study researcher to arrange a suitable time to complete the Q-sort. The study researcher will visit your place of work or university to administer the Q-sort. The consent form will be collected at the Q-sort administration.

Appendix 8: Factor Loadings with Categories

Table 7 shows the factor loadings again for every participant on each of the three factors. In this table the numeric loadings have also been given a categorical label. Positive loadings greater than .5 are labeled *Strongly Agree*, loadings between .1 and .5 are labeled *Agree*, loadings between -.1 and .1 are labeled *Neutral* and negative loadings between -.1 and -.5 are labeled *Disagree*. The lowest loading was -0.4502, thus, no *Strongly Disagree* category was needed. Categories have been devised by the researcher to assist in understanding the factor loadings. They are not prescribed by the methodology but are consistent with how factor loadings are signified in the methodology, for example, 14 of the 15 loadings labelled *Strongly Agree* in Factor 1 are considered to significantly load on to that factor.

Overall 28 participants agreed with Factor 1, 15 categorised as *Strongly Agree* and 13 as *Agree*. Three participants had *Neutral* loadings on this factor. Twenty-eight participants also agreed with Factor 2, 13 categorised as *Strongly Agree* and 15 as *Agree*. Three participants also had *Neutral* loadings on this factor. Twenty-three participants agreed with Factor 3, one categorised as *Strongly Agree* and 22 as *Agree*. Five participants had *Neutral* loadings on this factor and three had loadings categorised as *Disagree*.

Table 7

Loadings and Categorical Label for Each Participant and Factor

Participant	Factor 1	Factor 2	Factor 3
1	Neutral ¹ (0.0921)	SA ² (0.7522*)	Neutral (-0.0393)
2	Agree ³ (0.4548)	SA (0.6173*)	Agree (0.1643)

3	Agree (0.4599)	SA (0.5267*)	Agree (0.2254)
4	Agree (0.2184)	SA (0.7584*)	Agree (0.1694)
5	Agree (0.3909)	SA (0.6441*)	Agree (0.1796)
6	Agree (0.4372)	Agree (0.1691)	SA (0.5464*)
7	SA (0.8247*)	Agree (0.2227)	Agree (0.1233)
8	Agree (0.2031)	SA (0.6912*)	Agree (0.3047)
9	Agree (0.3083)	SA (0.7088*)	Agree (0.2336)
10	SA (0.7018*)	Neutral (-0.0040)	Agree (0.3525)
11	SA (0.7620*)	Agree (0.1635)	Agree (0.2757)
12	SA (0.7965*)	Agree (0.1451)	Agree (0.2031)
13	Neutral (0.0617)	SA (0.7744*)	Neutral (-0.0098)
14	SA (0.7758*)	Agree (0.3593)	Disagree ⁴ (-0.1065)
15	Agree (0.4748)	Agree (0.4760)	Agree (0.2685)
16	Agree (0.2159)	SA (0.6483*)	Disagree (-0.4502)
17	SA (0.6087*)	Agree (0.3311)	Agree (0.2648)
18	SA (0.7226*)	Agree (0.2308)	Agree (0.2025)
19	Agree (0.3940)	Agree (0.4125)	Agree (0.4794)
20	SA (0.6261*)	Agree (0.4816)	Neutral (0.0525)
21	Neutral (0.0279)	SA (0.7655*)	Agree (0.2498)
22	SA (0.7775*)	Agree (0.3447)	Agree (0.1892)
23	SA (0.6642*)	SA (0.5103)	Agree (0.1806)
24	Agree (0.3466)	SA (0.7745*)	Agree (0.1254)
25	SA (0.8154*)	Neutral (0.0980)	Agree (0.2578)
26	SA (0.6623*)	Agree (0.3281)	Agree (0.2532)
27	SA (0.5774)	Agree (0.3686)	Agree (0.4543)
28	SA (0.7808*)	Neutral (0.0317)	Disagree (-0.1611)
29	Agree (0.2193)	SA (0.8081*)	Neutral (0.0507)
30	Agree (0.4420)	Agree (0.3139)	Agree (0.4996)
31	SA (0.8263*)	Agree (0.2716)	Neutral (0.0635)

Note. ¹ Neutral = loadings between -.1 and .1

² Strongly Agree (SA) = loadings greater than .5

³ Agree = loadings between .1 and .5

⁴ Disagree = loadings between -.1 and -.5

* = Scores significantly loading onto a factor

Appendix 9: Reflective Comments for Each Factor Array

Factor 1 “Most Agreed” with Statements

Statement 21: *Challenging behaviour needs to be considered in the wider context of family and socioeconomic background, it is unethical to work with people with challenging behaviour and ignore this context*

“I’ve had a lot of clients who show behaviour that challenges due to abuse” (Q)

“I agree it is unethical to ignore this, ignore context assumes behaviour is just about person and not a reasonable response to environment. Not going to get far with intervention” (Q)

“From my experience and research the environment is key factor. I do believe environment shapes behaviour but within confines of what is inherent to the individual. Own experience, through changing environmental factors proves environment has impact” (Q)

“Believe it’s very important from clinical experience and academic teaching. Can’t formulate on diagnosis alone” (T)

“Challenging behaviour more influenced by what goes on around a person. Ignoring that...can’t change or reduce behaviour.” “Challenging behaviour is not a diagnosis, it’s a social construction” (T)

“Similar as before (meaning response in relation to statement 31- in that integration of social and medical models is important). Wider context etc. we don’t exist in a vacuum, consider wrong things if you ignore holistic” (T)

“Really agree, always have to know about family and wider system to effect any change regardless of whether it is a phenotypic behaviour etc.” (T)

“From experience of working with challenging behaviour it is unethical not to consider family context and background. This may be maintaining the challenging behaviour or protecting it from being much worse. In terms of intervention you need to consider family to see, regarding the capacity of intervention to work” (T)

“Made me think regarding behaviour and how about it’s about antecedents and triggers, more about attachment and relationships – internal world, obviously important to understand behaviour” (T)

“You need to take all factors into account, genetic, environment, systemic factors etc.” (Q)

Statement 31: *The medical and social models of disability can and should be integrated to enhance the lives of people with intellectual and developmental disabilities*

“Maximum benefit can be derived if both models are taken into account” (T)

“Both provide something valuable, I recognise they may disagree but they can provide something complementary to each other. Integration – capture complexity ‘ideal world’” (T)

“For me this promotes a holistic approach to people with IDD and how you might support them. I think that having a diagnosis increases understanding and gives you a reference frame to work with, for example, social aspects. Integration – gives a rounded view of someone” (T)

“The strengths of the medical and social models should be integrated to benefit the lives of individuals rather than a continued battle between the models” (T)

“Role of psychologist part of this is to use different models and use them together. If you’re not doing this you’re not doing your job. Good way to work in general” (T)

Statement 30: *Behavioural phenotyping can make a positive impact on quality of life for people with intellectual and developmental disabilities if the information is used appropriately*

“If you know about the phenotype and understand how a person acts and responds then that helps design an intervention” (T)

“Again, like point 11, phenotyping has the potential to enable us to understand the individual needs of clients leading to better informed formulations and intervention plans” (T)

Statement 15: *Understanding the diagnostic label informs idiosyncratic formulations, interventions and treatment plans*

“I have done a lot of autism work. I see diagnosis as part of formulation especially in autism, some construct validity for concept of autism which is supported by data and can help to make predictions about how a person may respond. Key question is inform not dictate” (Q)

“Goes back to nature-nurture debate. I think if you’ve a diagnostic label, especially in relation to particular behavioural syndromes you need to know about that, if you don’t you might use an intervention that is doomed to failure. More about managing than eliminating behaviour” (Q)

“By understanding diagnostic label gives you an idea why somebody behaves or responds in a certain way. Understanding helps in intervening” (T)

“I do believe this one - gives you such a head start if you know what you’re looking for. Adds a bit of weight when working with staff team, it’s a helpful heuristic or schema” (T)

Factor 1 “Most Disagreed” with Statements

<p>Statement 1: <i>Diagnosing different intellectual and developmental disabilities does not lead to greater understanding or change</i></p>
<p>“I think there’s evidence to say that’s not true, in forensic and mental health stream, in medical model, in family units. Having an appropriate diagnosis allows system to appropriately diagnose somebody” (Q)*</p> <p>“If you’re able to identify syndrome then gives you a realistic picture of what you can change” (Q)</p>
<p>Statement 52: <i>Genetic screening and behavioural phenotyping are akin to modern day eugenics</i></p>
<p>“So different”. “Genetic screening is about information for preparation to ensure person is helped in right way and not trying to reduce social value like eugenics did” (Q)</p> <p>“I think intention of genetic screening is very different to the intention 100 years ago. Intention now is to find a way forward to be realistic regarding changes. Culture has moved to the individual, intentions are sound” “99.9% of work done is in order to help provide understanding and help for individuals and families” (Q)</p> <p>“Interestingly I commented on this in explanation for part 11. It has potential to be abused however not researching or screening could lead to a disservice and not meeting needs” (T)</p> <p>“Misconception about genetic screening. It’s about how the information can be helpful e.g. Prader-Willi syndrome, Lesch-Nyhan syndrome” (Q)</p>
<p>Statement 25: <i>It is difficult to hold both a medical and a social model of intellectual and developmental disability</i></p>
<p>“Only if you suffer from rigid set of values or you are drawing from very narrow section of evidence base” (Q)</p> <p>“That’s formulation and that’s what we’re supposed to do. No reason why two can’t be integrated” (Q)</p> <p>“I am able to do this every day.” “Patients benefit more” (T)</p> <p>“Should integrate, how you do this is difficult. Clinically neglectful not to hold both in mind” (T)</p> <p>“Bit like what I said before regarding models” (See response to Statement 31 above) (T)</p> <p>“As a psychologist it should be possible to understand role of medical model and see</p>

<p>how it fits with social models. Should be done very well!” (Q)</p>
<p>Statement 70: <i>The diagnosis is more meaningful than the presenting behaviour</i></p>
<p>“I think the two are equally important. If you ignore presenting behaviour you are not going to get very far” (Q)</p> <p>“Being able to identify syndrome does not remove impact of environment. Diagnosis useful not for” (Q)</p> <p>“A diagnosis without a presenting problem would not be seen (in services)” (T)</p> <p>“Neglectful of individual differences – contexts and lives in amongst a label. Lose that quality clinical care” (T)</p> <p>“I do think when it comes to it genetic diagnosis is helpful, ‘more meaningful’ part of sentence I disagree with. Diagnosis doesn’t necessarily tell you much. Doesn’t tell you much regarding presenting behaviour, it’s different in cases where there’s a known behavioural phenotype. This statement is too black and white” (T)</p> <p>“I think having a diagnosis can help improve understanding but it is unethical and wrong to put all behaviour down to label. It’s more important to look at presenting behaviour than label” (T)</p> <p>“Important to understand meaning of behaviour” (T)</p> <p>“Similar. Assume behaviour is needs led, that’s where you would focus attention rather than assuming from diagnosis” (T)</p>

Note. *In all tables in Appendix 9, statements followed by the letter T in brackets indicate those made by trainee clinical psychologists and statements followed by the letter Q in brackets indicate those made by qualified clinical psychologists

Factor 2 “Most Agreed” with Statements

Statement 14: *Understanding the meaning or function of an idiosyncratic behaviour is more important than understanding the diagnostic label*

“You must understand the personal meaning of the behaviour” (Q)

“We spend a lot of time talking to families regarding behavioural condition or why it occurs and all complex factors are often unseen as more weight is put on diagnosis.” Takes a lot of effort to talk with families regarding how much that (diagnosis) offers but this is one piece of a complex formulation.” “Most important bits of work not diagnosis related” (Q)

“Whilst I do acknowledge that individual behaviour can have a genetic influence it doesn’t tell you very much regarding what to do, I don’t believe behaviours influenced by genetics are unchangeable” (Q)

“Fits with my view, function way more important. Doesn’t matter what diagnostic label is understanding the function is predominantly more important to me. In some ways diagnosis can be helpful but understanding the function is more meaningful. That’s what you would be looking for without a diagnosis” (Q)

“Because diagnosis is only useful understanding behaviour as a heuristical device. It doesn’t explain cause or function of behaviour. Some information is more dangerous than no information if you go of the diagnostic label” (Q)

“This can inform the idiosyncratic formulations, treatment plans that is tailored to specific individual need” (Q)

“Working with clients with challenging behaviour in my view is looking at why they are displaying it, its function, not trying to link behaviour with diagnosis, not trying to diagnose behaviour but trying to understand it. Focusing on diagnosis distracts from supporting people. Understanding diagnosis does not give answers. It feeds into believe ‘they have autism so there’s nothing we could do about it.’ It’s deterministic” (T)

“Understanding meaning behind behaviour is dynamic – tells something regarding the needs. Label tells you nothing about a person” (T)

Statement 20: *Service provision should be provided primarily on the basis of need not diagnosis*

“Just agree. Most people would say need.” “There’s a variation of need within diagnosis”. “Gatekeeping” (Q)

“People on fringes miss out.” “Should be based on need.” Participant doesn’t always agree with this statement as “you have to know diagnosis to access certain services” (Q)

“I struggle to see anyone disagreeing with this”. “Need goes way beyond just diagnosis. You cannot determine all of someone’s needs based on diagnosis.” “You generate delusions of need based on diagnosis” (Q)

“This is a far cry from reality – in an ideal world with ample resources” (Q)

“I think it’s an absolute that people should receive what they need. Need to look at individuals when assessing them as there is differentiation within a diagnosis” (T)

“Need is more reliably identified and about providing services that merit here and now pattern than diagnosis which may not be relevant. Diagnosis not homogenous – ASD exception, there are usually some connecting factors” (Q)

Statement 13: *The social model of disability is more helpful than the medical model of disability in intervening with people with intellectual and developmental disabilities*

“Not sure what medical model intervention is for IDD other than as a pharmacological straitjacket” (Q)

“Draws on wider systemic factors that impact on well-being and inform treatment” (Q)

“I think social model is more appreciative of holistic individual”. “Homogenous labels of medical model are unhelpful” (T)

“This links to previous one in terms of understanding the right intervention for people with IDs. You need to understand history and experience. You then have a better formulation, intervention and outcome when treating the diagnosis. Social model helps to understand, it is proactive. It looks at traumas, losses, relationships that might lead to current difficulties. Medical model explains through condition” (T)

Statement 21: *Challenging behaviour needs to be considered in the wider context of family and socioeconomic background, it is unethical to work with people with challenging behaviour and ignore this context*

“It is unethical to work with people and not consider wider context” (Q)

“I work in a service with focus on challenging behaviour and positive behavioural support; this is probably why I’m very passionate about this. Massive reason for referral to psychology. If we ignore context we are not going to be able to understand function of behaviour or the formulation” (Q)

“Much more compelling evidence about the environment” (Q)

Factor 2 “Most Disagreed” with Statements

<p>Statement 43: <i>What happens in the womb influences intelligence more than anything that happens after birth</i></p>
<p>“Intelligence is influenced by a range of factors, this is life-long” (Q)</p> <p>“It’s a rod to beat women with” (T)</p>
<p>Statement 9: <i>Genetically inherited traits are fixed and non-modifiable</i></p>
<p>“Disagree with anyone arguing either way (re genes versus environment), naïve to look at things in this way. It’s an interaction of genes and environment” (Q)</p> <p>“If we thought that then we wouldn’t do our job” (Q)</p> <p>“Behavioural traits are predicted by genetic make-up but are highly susceptible to environmental factors”. Participant used example of Asperger syndrome to describe this – “spontaneous occurrence in people with Asperger syndrome of becoming adept at social situations overtime, with no interventions but through social modelling” though genetic make-up would not seem to predispose this (Q)</p> <p>“Traits are an expression of something, not fixed, changeable” (Q)</p> <p>Participant said she had recently read about epigenetics and said “traits are highly modifiable with lifestyle” (T)</p> <p>“I don’t believe that things are fixed and non-modifiable. People can change given opportunities and support”(T)</p>
<p>Statement 2: <i>The medical model of disability is more helpful than the social model of disability in intervening with people with intellectual and developmental disabilities</i></p>
<p>“Medical model is more dominant but not more helpful” (Q)</p> <p>“Integrate them, naïve to think one is better” (Q)</p> <p>“Medical model little more than descriptive, it doesn’t tell you what to do.” “In a rare exception it does but then it doesn’t tell much about what to do” (Q)</p> <p>“Social model is better for intervention including formulation, consultation with staff, community teams etc. (I’m) much more interested in the social model. I may ask about physical health but not about diagnoses. Environment and wider context are more important” (Q)</p> <p>“Counter opposite.” “Tells you nothing regarding diagnosis, put them in a bin” (Q)</p> <p>“Clinical experience is indicative that it is pertinent to consider systemic factors”</p>

Participant said “don’t think medical model is more helpful” (Q)

“Both important, social model perhaps more so, links to number 13 (in most agree), applies to both” (T)

“Medical model assumes too much contribution from genes and downplays importance of early social experiences” (T)

“My belief is that social psychological model of understanding any kind of difficulty is always more helpful in trying to understand them and their behaviour” (T)

“More evidence for social model of disability being relevant and meaningful” (Q)

Statement 70: *The diagnosis is more meaningful than the presenting behaviour*

“Diagnosis does not have that much scientific validity. They are clues regarding things that go together” (Q)

“Same reason as before.” “Complexity is ignored in diagnosis but the presenting behaviour is often more complex” (Q)

“Labelling not always useful.” “It gets in way of work you do.” “Better to work with presenting feeling and/or behaviour” (Q)

“It’s the presenting behaviour that needs to be examined.” “Genetic conditions can predict certain behaviour e.g. chin tapping behaviour in Lesch-Nyhan syndrome but this is quite rare. Even in those circumstances diagnosis had no meaning without the presenting behaviour. Can’t change genetic diagnosis but you can modify the behaviour.” “Diagnosis does not tell about the communicative function of the presenting behaviour” (Q)

“Presenting behaviour is more important than diagnosis because diagnosis doesn’t mean they will present with certain behaviours. Presenting behaviour has no impact on what diagnosis is” (Q)

“A load of rubbish, ridiculous statement” (Q)

“We can understand an individual more effectively by analysing the function of their behaviour” (Q)

“There are so many possible reasons for learning disabilities and interaction with other factors.” “Behaviour is often not related to diagnosis.” “Diagnosis is informative but not helpful” (T)

“The function of behaviour needs to be looked at. If a person is angry then what’s happening now?” (T)

“A diagnosis or label doesn’t tell you much about an individual’s behaviour. Looking at meaning behind the behaviour is more important to inform how we work

with them.” “Heterogeneity and complexity, hard to capture that accurately in one label” (T)

“Flip-side, most meaningful is what people say and do” (T)

“Presenting behaviour is clearly more important than diagnosis – two people with same diagnosis presenting with different behaviour” (Q)

Factor 3 “Most Agreed” with Statements

<p>Statement 11: <i>What’s not important is genetic screening but the information it yields and how that is used</i></p>
<p>“Most important thing to grapple with is ethics of genetic screening. Used to have a baby or not then it is not useful, is immoral. Used to increase understanding then the better I feel about it” (Q)</p>
<p>Statement 17: <i>Impact of genetic screening on the individual and society should be considered before it is used</i></p>
<p>“Most important thing should be considered before it is used. Genetic screening used to some degree and it’s not considered, what it means to people, especially in some countries. Every human being has rights. Genetic screening is fine as long as no decisions are made regarding child birth” (Q)</p>
<p>Statement 23: <i>Associating genetics with people with intellectual and developmental disabilities can undermine the progress of the social model of disability</i></p>
<p>“I think people with learning disabilities have a huge amount to offer society. Society isn’t created by just intelligence. Other things are important – characteristics like being loving, caring, funny, strong, love for gardens etc. carry as much value for society. Social rather than medical/intellectual models” (Q)</p>
<p>Statement 28: <i>Genetic screening can lead to negative social engineering with the creation of ‘designer’ societies where people with disabilities are undervalued and social/environmental influences on disability undermined</i></p>
<p>“I think this is so true. ‘Designer’ societies occur if genetic screening goes wrong (used to reduce learning disability rather than understand it). End up with a flawed society” (Q)</p>

Factor 3 “Most Disagreed” with Statements

<p>Statement 2: <i>The medical model of disability is more helpful than the social model of disability in intervening with people with intellectual and developmental disabilities</i></p>
<p>“Not true – social model is hugely important. What happened in society is that the medical model has existed for 100 years and social model only since 1970s, we’re just beginning to have social model but pendulum might swing back to medical model. Some benefit to defining cause – families feel less pressure, less responsibility as they see it as part of phenotype” (Q)</p>
<p>Statement 9: <i>Genetically inherited traits are fixed and non-modifiable</i></p>
<p>“Inherit but environment impacts on life. Hardest part is about how services are set up” (Q)</p>
<p>Statement 43: <i>What happens in the womb influences intelligence more than anything that happens after birth</i></p>
<p>Participant said it was what happens in womb has some importance – gave personal example (Q)</p>
<p>Statement 70: <i>The diagnosis is more meaningful than the presenting behaviour</i></p>
<p>“Both important, both are meaningful” (Q)</p>

Appendix 10: Distinguishing Statements for the Three Factors

Table 8

Distinguishing Statements for all Three Factors

Statement	F1	F2	F3	Q-sort Rank
13 The social model of disability is more helpful than the medical model of disability in intervening with people with intellectual and developmental disabilities	1	4 ¹	1	
17 Impact of genetic screening on the individual and society should be considered before it is used	3	1	4	+ ² on all factors
51 Positive behaviour support is more influential in improving the quality of life for a person with an intellectual or developmental disability than the outcomes of behavioural phenotyping and genetic testing	1	3	1	
73 Behavioural phenotyping is beneficial in understanding some syndromes such as Down syndrome but this is an exception, behavioural phenotyping does not generally aid understanding as much as other factors	-2	-1	-3	- on all factors
20 Service provision should be provided primarily on the basis of need not diagnosis	3	4	-3	
14 Understanding the meaning or function of an idiosyncratic behaviour is more important than understanding the diagnostic label	2	4	-1	
22 Focusing on the individual's expressed difficulties is more helpful than looking at the difficulties in the context of a diagnosis or conflicting diagnoses	1	3	-1	- on F3 + on F1 & F2
18 Many professionals working with individuals with intellectual and developmental disabilities are unconcerned with why someone has the impairment	1	1	-2	
77 The voice of people with intellectual and developmental disabilities in the genetic testing debate is unheard	3	3	-2	

Statement	F1	F2	F3	Q-sort Rank
63 Understanding the causal pathway to an individual difference can have a positive influence on well-being	1	-1	2	
34 Genetic disorders lead to different behavioural outcomes	2	-1	2	- on F2 + on F1 & F3
48 Innate abilities allow children to develop typically, absence of such abilities affects development	1	-2	2	
23 Associating genetics with people with intellectual and developmental disabilities can undermine the progress of the social model of disability	-1	1	4	- on F1 + on F2 & F3
26 Diagnosis specific services presume specialisation which fits with a medical model	-1	1	1	
29 Inclusion in society can be enhanced by understanding individual difference that can be traced to a specific genetic disorder	1	-3	-1	- on F2 & F3 + on F1
47 No one knows what the non-genetic causes of individuality are	-2	-3	1	
1 Diagnosing different intellectual and developmental disabilities does not lead to greater understanding or change	-4	-1	1	- on F1 & F2 + on F3
25 It is difficult to hold both a medical and a social model of intellectual and developmental disability	-4	-2	3	
27 Heterogeneity of presentations within diagnostic categories can render the diagnosis meaningless	-1	2	-3	- on F1 & F3 + on F2
28 Genetic screening can lead to negative social engineering with the creation of 'designer' societies where people with disabilities are undervalued and social/environmental influences on disability undermined	0	1	4	+ on F2 & F3 N on F1
39 Genetic screening may reduce complex behaviour to genes, ignoring the impact of other factors on behaviour	0	3	2	
11 What's not important is genetic screening but the information it yields and how that is used	3	0	4	
30 Behavioural phenotyping can make a positive impact on quality of life for people with intellectual and developmental disabilities if the information is used appropriately	4	0	3	N on F2 + on F1 & F3
61 Genetic screening, if used in an appropriate, responsible way, has the potential to positively affect lives	3	0	3	

Statement	F1	F2	F3	Q-sort Rank
67 Diagnosing different intellectual and developmental disabilities/behavioural syndromes provide unnecessary labels and can create stigma	-1	<i>0</i>	-2	- on F1 & F3 N on F2
32 Diagnosing intellectual and developmental disabilities/behavioural syndromes can hinder and create barriers to therapeutic work	-3	<i>0</i>	-2	N on F2
36 Health and social care resources should be directed towards understanding and improving social and environmental factors affecting people with intellectual and developmental disabilities	2	3	<i>0</i>	N on F3 + on F1 & F2
49 Genetic testing would not add to the quality of life for a person with an intellectual or developmental disability	-3	<i>0</i>	-3	- on F1 & F3 N on F2
24 The environment that a child experiences is as much a consequence of the child's genes as it is of external factors: the child seeks out or creates his or her own environment	-1	-2	0	- on F1 & F2 N on F3
40 A diagnostic label is more helpful to an individual than it is unhelpful	<i>0</i>	-2	3	
3 Sociocultural factors such as socioeconomic status and family background are more important than behavioural phenotyping in explaining behaviour	0	3	-1	
72 People with different behavioural syndromes have more similar than dissimilar behaviours	0	-2	1	Different ratings of N, + & - on the factors
66 Focusing on how genetics influences behaviour downplays the role of more important social influences	-1	2	0	
55 The shared environment has a greater influence on sibling similarities and differences than genes	0	2	-1	
10 Genetic screening/testing causes negative attitudes towards disability to pervade and continue	-2	1	0	
58 Genetics and behavioural phenotyping represent a shift backwards to the medical model of disability	-3	1	0	

Note. ¹ A factor rating in italics highlights that the statement was distinguishing for the indicated factor (statements can be distinguishing for more than one factor)

² N denotes *neutral*, + denotes *agree* and - denotes *disagree*

Appendix 11: Consensus Statements for the Three Factors

Table 9

Consensus Statements for all Three Factors

Statement	Q-sort Rank
19 Diagnostic labels can serve to deny people access to services	
21 Challenging behaviour needs to be considered in the wider context of family and socioeconomic background, it is unethical to work with people with challenging behaviour and ignore this context	
35 Families usually want to know the cause of their child’s intellectual and developmental disability	+* on all
56 Genetically inherited traits/characteristics can be subject to change and adaptation by the environment - Heritability does not mean immutability	factors
60 The science of intelligence testing is flawed	
78 In clinical practice it is difficult to keep up to date on new research developments due to time and service pressures	
80 Social instincts may mean it is natural to exclude people with intellectual and developmental disabilities from the group; however, our human side and social responsibility should cause us to fight against this and recognise the value to the world of people with IDD’s and diversity in general	N on F3 + on F1 & F2
81 A partially inherited low IQ might be subject to extensive improvement through education	N on F1 & F3 + on F2

44	Genetic screening creates a battle between innate social instinct versus human rights	
54	Genetic screening/testing should be available to all as people have a right to information about their genetic make-up	N on all factors
59	Genetic aspects of a condition may be viewed as irrelevant or potentially negative by professionals/staff members	
79	Research of relevance to people with intellectual and developmental disabilities is often difficult to access by services which affects the application of new research	

53	Labelling genetic disorders/behavioural syndromes may negatively affect prognosis	- on F3, N on F1& F2
50	Current knowledge and understanding of different disorders is too limited to make disorder-specific services worthwhile	- on F1 & F3, N on F2

74	Intellectual ability is affected more by nature than by nurture	- on F1 & F2, + on F3
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5	Growing up in the same family has no discernible or marked effect on the IQs of siblings	- on F1 & F2, N on F3
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4	Growing up in the same family has no discernible or marked effect on the IQs of siblings	
7	Behavioural syndromes rarely occur	
9	Genetically inherited traits are fixed and non-modifiable	
43	What happens in the womb influences intelligence more than anything that happens after birth	
46	Geneticists have a moral and ethical responsibility to the today not the tomorrow	- on all factors
68	As people get older their environment has a stronger influence on their behaviour	
70	The diagnosis is more meaningful than the presenting behaviour	

Note. * N denotes *neutral*, + denotes *agree* and - denotes *disagree*