The development of an online tool to facilitate collection of video data in British Sign Language (BSL): principles, process and implications for Deaf people's engagement with research and service development

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Contents **Page number**

Tables and figures **Page number**

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Abstract

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The development of an online tool to facilitate collection of video data in British Sign Language (BSL): principles, process and implications for Deaf people's engagement with research and service development

Introduction: Deaf people who use British Sign Language (BSL) face multiple barriers to accessing services, including the interpreted communication of complex information. Signed languages have different grammar and syntax from English and take full advantage of their visual modality through the inter-relationship of signs. Previous research identified the need for Deaf people's contribution to service development and research. However, there are challenges to facilitating this, including that Deaf BSL users in the UK are geographically widely dispersed and researchers ideally need to be Deaf or at least fluent BSL users themselves to allow the detail of culture and communication to be conveyed: interpretation of data from one language into another for the researcher's benefit risks losing crucial richness and nuance from a participant's contribution.

Purpose: To develop a means of Deaf people's autonomous participation in data generation using the vehicle of a putative data collection tool concerning access to and use of genetic counselling (a 'use case' approach). Through that process, to engender a generative approach to the identification and collection of terminology in BSL in this field of interest.

Methods: A literature review across medical, educational, Deaf studies, linguistic and information technology fields informed the subsequent consultation with information technology (IT) specialists across the University. Several different technologies theoretically capable of capturing video data, necessary to record signed language input, were explored. The information content for the use case was developed from previous information resources and from an acted scenario filmed specifically for the study. The tool was then developed using two proprietary pieces of software (Selectsurvey questionnaire development software and the University's version of a user-generated content site: the video library server (VLS)) and a video capture application in Flash available as freeware which was modified to meet the requirements of a visual-spatial language. The three software components were linked together using bespoke Javascript and Java coding to create an integrated outward-facing website for the user, capable of capturing video data as streamed files direct to a secure server for later download by a researcher.

Results: This study determined the essential features of an online data capture tool; incorporated the features into a theoretical use case aimed at collecting context-rich and in-depth data from which a technical specification could be drawn; identified the available technologies with the capacity to realise such a tool; and built, tested and piloted the tool to evidence its feasibility.

Conclusions: The resulting tool has the capacity to present information in BSL and English and enables participants to respond asynchronously, remotely and securely in BSL via webcam, or using spoken or written English. The IT technologies used are changing fast, but there are principles underpinning such a tool that can be carried forward to future software dedicated to presenting and capturing video data, generally and also specifically in signed languages.

16th November 2017.

Declaration

No portion of the work referred to in the thesis has been submitted in support of an application for another degree or qualification of this or any other university or other institute of learning.

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Dedication

For Dot

Professor Dorothy Trump
mentor, colleague, friend, supervisor and self-confessed fellow geek

24th January 1964 – 26th March 2013

‘Further up and further in.’ (Lewis, 1956 p153)

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This work was primarily undertaken within the Social Research with Deaf People Research Group (SORD) in the Division of Nursing, Midwifery and Social Work, School of Health Sciences and Professor Young has been the primary supervisor from 2011 onwards. From 2007-2011 Professor Young and Professor Trump co-supervised me. I am registered in the Division of Evolution and Genomic Sciences, School of Biological Sciences, but have been linked with both divisions within the Faculty for the duration of the project.

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The Author

Rachel Belk’s professional background is as a genetic counsellor with a BSc (Hons) Genetics and an MSc Genetic Counselling. She has worked clinically for over twenty years in general and cancer genetic counselling and as a specialist deafness genetic counsellor. This work has given her practical experience and knowledge of communicating complex and fast-developing information cross-culturally and cross-linguistically. Her previous research and development work related to her clinical job has been mainly in genetics, information and deafness, with a particular focus on improving access and communication within health services. This led to a shift in role in 2007 to take up a part-time Doctoral Research Fellowship from the NIHR from which this MPhil dissertation is the primary output. During this period, she has developed her interest in and understanding of how communication between individuals is dependent on many more factors than simply the concepts under discussion: for example, the semantic and pragmatics of the language(s) being used, whether interpretation or translation between languages is taking place, the language history of the individuals, their background knowledge of the subject, and the positionality and relationship between the individuals. She is particularly interested in the potential of IT to overcome communicative challenges and in their application to real life problems.

Introduction

This thesis developed from an identified area of concern within health service provision – communication between Deaf British Sign Language (BSL) users and hearing people in a complex fast-changing area of health information: genetics and genetic counselling. The need for research that focuses on communication, and terminology in particular, was backed up by previous work evidencing the barriers faced by d/Deaf (see page 16) people in accessing health services, both to reach an appropriate service and then to take full advantage of it. Poor communication is known to be a significant contributor to these barriers. Further, although there is a growing body of work that has sought to document and explore the experiences of d/Deaf people, there is a paucity of research and developments that have enabled flexibility in how d/Deaf people engage with research projects as participants, and with services as users and consumers. Specifically, there are a number of conditions that, together, make a unique situation:

- d/Deaf people have great diversity in language preference and fluency
- The population of BSL users is of low incidence and widely geographically dispersed
- BSL is a visual spatial language without an orthographical form
- Information technology and the internet are now used widely and to great effect in information provision, healthcare and research. However, the majority of these applications are still text-based, making them inaccessible or less accessible for people whose first or preferred language is not an oral/written one.

The thesis presents one solution to facilitate engagement: the development of a remote, asynchronous, secure, online questionnaire tool that allows BSL users to participate, autonomously, in a research study in their preferred language(s). The detail of the technologies used and developed was necessarily of its time (2008-2009), given the rate of change of hardware and software. However, at the time of the research, there was no off-the-shelf solution available, nor had the requirements for such a tool to be effective for BSL users been researched in depth. The identification of principles and implications that stood apart from the technology of the time remains valid and there is still no easily available solution to this most basic of challenges: the engagement of visual language users in research. Furthermore, the principles identified through this study can be applied to other situations, both for visual language users to engage with other research or services, and for other groups that prefer or require non-written forms of communication. These wider implications are argued in Chapter 6 with reference to the current literatures available.

The following brief introduction gives an overview of some key facts related to the community(ies) of d/Deaf people in the UK and to British Sign Language. This information begins to evidence the statements made above about the particular cultural, linguistic and geographical situation which this thesis addresses. In addition, it sets out why and how genetic counselling may be relevant to a d/Deaf person and how such services are organised in the UK. This gives context to the complexity of the communicative challenge within genetic counselling for any individual, but particularly if the communicative exchange is between users of different languages. This complexity supports the case for developing a means by which Deaf people's linguistic and cultural preferences can be researched without being filtered through a majority language (English) which may not be their preferred means of communication. Finally, this introductory section defines certain concepts and terms as they will be used throughout the thesis.

Chapter 1 takes, as its starting point, an exploration of the diversity of ways in which deafness and signed languages are experienced and understood. Describing this diversity and its lack of recognition by the majority (hearing) world underpins why the current hegemony does not facilitate engagement by a minority population who experience the world visually rather than audiologically and whose language does not fit into the existing predominant channels of communication.

Chapter 2 explores genetic counselling as an exemplar of a health service where communication is crucial and central. d/Deaf people are potential users of genetic counselling services, both related to the causes of deafness and to other genetic conditions. However, the number of culturally Deaf people accessing genetic counselling is low. Lack of availability of information in BSL about genetics and genetic counselling services may be a factor. In addition, genetics and genetic developments are widely discussed in the general population, but Deaf people have less access to equivalent discussions in BSL. Within the genetic counselling consultation itself, good communication is fundamental because the information is complex and decisions can be emotionally difficult or ethically challenging. There are therefore communication challenges with all genetic counselling clients, but chapter 2 argues that these are potentially greater, not only if the professional and client are using different languages, but when other factors (cultural, environmental, lack of awareness) affect the level of mutual understanding gained and the relationship formed between health professional and client. Building upon chapter 1, this examination supports the case of need for a means by which Deaf BSL users can contribute to service improvement and research -

avoiding, where possible, communication that is always mediated through a third party interpreter or where Deaf people do not have the option to use BSL.

Chapter 3 considers research into language as the bridge between meaning to an individual (both semantic and pragmatic) and transmission of that understanding through information provision and dialogue. It considers work done both on understanding within a single language and on translation between two languages. This closer examination of the relationship between meaning and language, and the recognition of how the resources of different languages influence communication, also underpin the need to create opportunities whereby individuals can contribute to research, debate and development in their language of preference.

In chapter 4, the literature discussed in chapters 1 to 3 builds the rationale for this thesis. The chapter argues for an ontological and epistemological orientation that can address the stated aims and objectives. The background literature reviewed leads to an understanding of the problem in general terms: that health services research and development may seek the views and knowledge of d/Deaf people, but conventional and commonly used methods of data collection, such as online or written surveys, will not work for d/Deaf people whose English is not necessarily fluent. In addition, meeting the rights of Deaf BSL users to information and access in their own language needs an adaptation of the methods of data collection and feedback to fit the visual requirements of a language with non-written form. The chapter then moves from that broad statement of the problem to describing the methods used to develop a means of remote data capture.

Chapter 5 reports the detailed development and properties of the remote data capture tool, whilst Chapter 6 summarises the outcomes and contextualises the implications of the work described.

Number of d/Deaf people in the UK, identification terms and language use

The terms ‘deaf’ and ‘Deaf’ are used throughout this thesis. Conventionally (Woodward, 1982), the noun ‘deafness’ and adjective ‘deaf’ refer to the audiological measure of deafness. ‘Deaf’ with a capital D refers to an identity in the same way as one might capitalise a nationality. British Sign Language (BSL) is the first or preferred language for most Deaf people in the UK. Deaf people both self-identify as belonging to a linguistic and cultural minority and are recognised as such by others. ‘deaf’ can also be used to identify someone with deafness who is not Deaf, usually someone who is not a sign language user. d/Deaf is used when referring to both of these groups, or when not seeking to distinguish between them or in acknowledgment of the fluid and situational nature of identities. There are other terms used to refer to deafness in the general population and a person with deafness is likely to have personal preference or objection to certain terms. For example, ‘hard of hearing’ is often used to mean a mild or moderate audiological deafness and/or age-related deafness; ‘deafened’ is used to refer to acquired deafness, often more than a mild or moderate hearing loss; many Deaf people object to being described as having a hearing loss when they have not lost their hearing, but have been born deaf; the term ‘hearing impairment’ is still used widely by health professionals, but is objected to by many d/Deaf people themselves.

10 million people in the UK (1 in 6 of the population) have some level of deafness (Action on Hearing Loss (previously Royal National Institute for the Deaf), 2014), of which two thirds are over retirement age. Just over 800,000 of this total are, in audiological terms, severely or profoundly deaf. Severe deafness has been defined as a level of deafness where the quietest sounds that can be heard unaided are between 70 and 94 decibels and profound deafness as a level of deafness at which the quietest sounds that can be heard unaided are 95 decibels or more (Mazzoli et al., 2003). 135,500 people of working age have severe or profound deafness and the majority will have had this since childhood (Action on Hearing Loss (previously Royal National Institute for the Deaf), 2014). The aim of digital hearing aids and cochlear implantation is to improve hearing thresholds, ideally to those within the speech range, so that individuals have access not just to sound but to sounds that enable them to decode speech. However, no intervention can fully restore hearing (Humphries et al., 2012). Someone with severe or profound deafness is likely to find speech difficult to follow with hearing aids and may use lipreading and/or BSL may be their first or preferred language. However, BSL is also used or preferred by many people with lesser audiological levels of deafness.

An individual’s communication choices will be influenced by many factors including whether they have been deaf since birth or become deaf later in life, whether they have other d/Deaf family members, their childhood communication environment and their educational experiences such as whether they attended a deaf or a hearing school (Mitchell and Karchmer, 2005; Mitchell and Karchmer, 2004b). Other factors include developments in hearing technologies and earlier identification of deafness in infants. Over the past 20 years, both the minimum age for, and hearing threshold for candidature for cochlear implantation has reduced significantly (National Institute for Health and Care Excellence, 2009; British Cochlear Implant Group, 2016) and the quality of digital hearing aids has improved markedly (University of Manchester, 2017). Universal newborn hearing screening, which enables identification of deafness as early as eight weeks of age, is now standard in most economically developed countries of the world (Public Health England, 2016; World Health Organisation, 2010). An estimated 100,000 people in the UK are BSL users (British Society of Mental Health and Deafness, 2010). BSL is a complete language which is grammatically and structurally distinct from written and spoken English and which is more similar to other signed languages than to English (Sutton-Spence and Woll, 1999b). Some people in the UK who are born deaf are monolingual in BSL and others are bilingual in English or other spoken/written languages. Of the latter group, some, not all, Deaf people are balanced bilinguals (similar levels of fluency in both languages) and others are multilingual e.g. in several signed languages. Although a proportion of Deaf people have excellent fluency in written English, on average the Deaf population has depressed levels of literacy (Mayer, 2007). Those people who predominantly use BSL are recognised as a minority cultural community (the Deaf community) and BSL now has formal governmental recognition as an indigenous language of the UK (Department for Work and Pensions, 2004; Smith, 2003). In Scotland, BSL has a protected status in law with obligations on public services to promote equality of access to goods and services through it as well as outlawing discrimination on grounds of BSL use (The Scottish Parliament, 2015).

Aetiology of deafness

Deafness at birth or in early childhood

In approximately half of children with severe or profound deafness present by the age of 5 years, there is a genetic cause (MacArdle and Bitner-Glindzicz, 2010). Non-genetic causes include infections during pregnancy (e.g. rubella and cytomegalovirus (CMV)), infections after birth (e.g. mumps or meningitis) or complications of prematurity or birth trauma (Fortnum et al., 1996; Bitner-Glindzicz, 2002). Of those people whose deafness has a genetic cause, two-thirds have deafness alone with no other physical signs or associated health problems. One third of people have a syndromic cause where deafness is part of a recognisable pattern of signs or symptoms.

Health professionals providing services related to deafness may refer families to genetic counselling services to discuss a possible genetic aetiology. The majority of those referred are hearing parents with deaf children. Because of the large proportion of childhood deafness that has a genetic cause, a genetics consultation, including diagnostic genetic testing of the Connexin 26 gene for all children with severe or profound deafness and sometimes testing of other genes, is part of the recommended investigations (British Association of Audiological Physicians and British Association of Paediatricians in Audiology, 2009; British Association of Audiovestibular Physicians and British Association of Paediatricians in Audiology, 2008; MacArdle and Bitner-Glindzicz, 2010).

At least sixty different genes that can cause deafness have been identified and more than a hundred additional genes have been implicated as being involved in the development and function of the ear (Van Camp and Smith, 2014). This complexity makes it difficult to pinpoint the cause of deafness in many children. This is particularly true if both parents are also deaf or hard of hearing as the causes of their deafness are more likely to be different. With current tests, it is only possible to determine the probable cause of deafness for a maximum of 75% of children (MacArdle and Bitner-Glindzicz, 2010).

Of the genes causing non-syndromic sensorineural deafness (SNHL), approximately 75-80% are autosomal recessive, another 10-15% are autosomal dominant and the remaining 5-15% are X-linked or mitochondrial genes or have a chromosomal cause (Bitner-Glindzicz, 2002). The figure of 90% of children with any level of deafness being born to hearing parents (Schein, 1989) is widely used and the high proportion of SNHL that is recessive in origin (whereby both parents would usually not have deafness) largely explains these proportions. Conversely, the number of different genes involved in the ear and hearing means that no more than 10% of children of two parents with deafness have some deafness themselves (ibid.). This ‘90-percent rule’ (Schein, 1989 p106) is easily remembered, hence its common use, but has been challenged as a likely overestimate (Mitchell and Karchmer, 2004a).

There are usually no additional distinguishing features that can help to make a clinical diagnosis pointing to a specific gene, although in some cases a characteristic audiogram may be suggestive of a particular gene. Unless a distinguishing feature does suggest that testing of a particular gene or genes can be offered, it has not been possible previously to offer comprehensive genetic testing of a large number of genes involved in the ear. Testing of one particular gene, Connexin 26, is routinely offered as it accounts for up to 50% of autosomal recessive SNHL (Petersen and Willems, 2006). Capacity to test more genes is changing as massively parallel genetic sequencing techniques increase the throughput of genetic testing in the laboratory and/or gene chips are developed which allow a number of genes and changes within them to be tested at the same time (Hilgert et al., 2009; Shearer et al., 2010; Wang et al., 2011).

There are at least 400 syndromes described of which deafness is one feature (MacArdle and Bitner-Glindzicz, 2010). Most are very rare, but a small number are seen more frequently within the genetic counselling clinic. These include Usher syndrome (deafness and retinitis pigmentosa), Waardenburg syndrome (heterochromia, depigmented patch of hair or skin, early greying of hair, and dystopia canthorum in one type), Alport syndrome (thin basement membrane disease progressing to loss of renal function), Branchio-oto-renal (BOR) syndrome (dysplastic kidney or renal agenesis, cysts or pits near the branchial arches), Pendred syndrome (enlarged vestibular aqueduct (EVA) and/or Mondini defect (lack of usual number of cochlear turns), goitre, hypothyroidism or hyperthyroidism) and Jervell and Lange-Nielsen (long QT interval picked up on ECG). Some have fewer implications for future health, but the accompanying signs just help with diagnosis e.g. Waardenburg syndrome types 1 and 2, whereas others have significant associated health risks e.g. Jervell and Lange-Nielsen syndrome which predisposes to sudden cardiac death. Additional syndromic causes are being identified with new laboratory techniques such as microarrays which can identify a small section of missing or duplicated chromosome which may contain several genes including one associated with deafness.

Another cause of deafness is Auditory Neuropathy Spectrum Disorder (ANSD), in which deafness is caused by a difficulty with nerve signal transmission rather than a problem within the cochlea. Its contribution to childhood deafness is still not fully understood, though is a subject of much current research (Manchaiah et al., 2011). A diagnosis of ANSD is made when an otoacoustic emissions (OAE) test gives a normal result (as this test measures the cochlear echoes sent back from the functioning hair cells), but an auditory brainstem response (ABR) test gives an abnormal result, showing that the auditory signal is not being processed correctly by the brain. The diagnosis may initially be missed for otherwise well children who receive newborn hearing screening through the national programme. This is because newborn hearing screening in England and Wales uses only the OAE test (compared to the protocol for babies in neonatal special care, who receive both an OAE and an ABR test). Auditory neuropathy can be caused by single gene changes or can be a feature of neuropathic genetic conditions such as Charcot-Marie-Tooth disease. It is also associated with environmental causes such as complications of prematurity.

Age-related deafness

The great majority of people with deafness have mild or moderate deafness that has developed in older age. Environmental factors and ageing itself are significant contributing causes, though there are genetic factors as well (Van Eyken et al., 2007).

More is starting to be understood about these genetic influences in acquired deafness, such as genes involved in early onset otosclerosis (Van Camp and Smith, 2014). Some genes have a variable effect on hearing or can be non-penetrative, meaning that a person carrying a change in one of these genes will not necessarily develop deafness. There are, however, single gene conditions causing profound deafness in adulthood which can be clearly diagnosed. One such condition is neurofibromatosis type 2 (NF2): the majority of people with this condition develop benign tumours on the hearing nerves in their twenties and thirties.

Relevance of genetic counselling to d/Deaf individuals

As a separate issue to their deafness, d/Deaf people of course have the same chance as any other member of the population of being at risk of inherited diseases. They may benefit from, for example, being referred because of a history of breast cancer in their mother and maternal aunt and being offered the options of breast screening or other testing. Regardless of Deaf people's views towards genetic counselling for deafness (see Chapter 2), lack of equality in access to genetic counselling could, therefore, have implications for their health and well-being with respect to unrelated genetic conditions.

Genetic counselling around possible genetic causes of deafness may also be relevant to d/Deaf individuals. It is recognised that increased knowledge in itself is an outcome of genetic counselling that is valued by individuals and can increase their sense of control or support their self-identity (Berkenstadt et al., 1999; Payne et al., 2007). Richards points out that people are very interested in where certain characteristics have come from:

> *The public's knowledge and beliefs about inheritance have not arisen de novo with the coming of the new genetics, or even with Mendelian genetics at the turn of the century: they have long been part of family culture. Much family talk is about particular characteristics of family members, who these may have been acquired from, and who they may be passed to.* (Richards, 1996 p249)

This may be even more the case when the characteristic is something which distinguishes the individual as a member of a minority cultural community, as is the case for an adult who considers him or herself Deaf, even if that person does not see their deafness as a disability or something to be avoided. Arnos and colleagues, from the genetic counselling service based at Gallaudet University, USA (the largest Deaf-only university in the world) state that:

deaf individuals have a deep curiosity about the cause of their own deafness and the implications for future generations. When genetic information is presented in a manner that is sensitive to their cultural and linguistic differences, deaf people are very enthusiastic about participation in the genetic counseling process. (Arnos et al., 1991)

Even individuals with an environmental cause to their deafness may gain useful information through genetic counselling because it may identify the cause and so give information about a low chance of their future children inheriting deafness from them. In addition, 70% of people with significant deafness have a partner who is also deaf (Mitchell and Karchmer, 2004a) and assessing an accurate chance of deafness/typical hearing in future children would need to consider both partners.

The majority of parents of deaf children are hearing and, as such, are far more likely to consider their child's deafness as a deficit or disability, at least initially. However, most parents do not ask to use the knowledge gained through genetic counselling actively to avoid having another child with deafness by, for example, use of prenatal testing. These figures are not published, but are known to genetic counsellors through their clinical practice. Brunger and colleagues asked parents of deaf children why they were interested in having a genetic test and the most common reason given (by 93%) was simply to identify a cause – the knowledge being a positive outcome in itself (Brunger et al., 2000). In addition, this knowledge can help with practical planning around education and language. This empowerment through understanding can extend to the deaf child within a hearing family who may increasingly question their self-identity as they get older (Ahmad et al., 2002). If the cause of deafness is known, this can sometimes give additional information about whether the deafness is likely to progress or remain at the same level, whether there are likely to be additional problems such as balance and whether cochlear implants are more or less likely to be of benefit (MacArdle and Bitner-Glindzicz, 2010).

Aside from information about the cause of deafness or chance of recurrence, there is the chance that someone with deafness has a syndromal genetic cause with accompanying signs or risks to health. Many d/Deaf people are aware of syndromic causes. For example, there is a specific BSL sign for Usher syndrome as a well-recognised syndrome. From a genetic counselling professionals' perspective, an additional benefit of aetiological investigations, as well as helping to identify a cause of the deafness, is therefore to identify or exclude potential health problems that could be monitored in the future with the aim of reducing mortality or morbidity (MacArdle and Bitner-Glindzicz, 2010).

There are a small number of individuals, usually hearing, who may wish to take active steps to avoid having a child with deafness or other health problems that are part of a syndrome that includes deafness. In this case, if the cause has been identified, prenatal testing or pre-implantation genetic diagnosis (PGD) may be options. The acceptability of these options to avoid non-syndromic deafness has been discussed since before they became technically possible (Chadwick and Levitt, 1998; Middleton et al., 1998) and they remain highly controversial, particularly amongst the Deaf community (Emery et al., 2010). The individual gynaecologist who would carry out a termination of pregnancy if a foetus was identified as carrying a gene for deafness through prenatal testing would have to agree that this was a serious enough condition within the stipulations of the Abortion Act (UK Government, 1967). This agreement is by no means a certainty. Non-syndromic autosomal recessive sensorineural deafness has been approved as a condition for which PGD can be offered as well as several syndromal causes of deafness (Human Fertilisation and Embryology Authority, 2014).

The structure of genetic counselling services in the UK

In the UK, genetic counselling services are provided by 21 regional centres within the National Health Service as part of tertiary care i.e. providing a highly specialist area of healthcare across a wide geographical area. Genetic counselling professionals see families with the whole range of conditions that have a definite or possible genetic cause. A significant proportion of referrals are now because of a family history of particular cancers: most commonly breast, ovarian and bowel. The broad range of conditions seen by genetic counselling services – and I avoid the word illness specifically in the context of this thesis – means the family experiences vary greatly and, coupled with individual coping mechanisms, family composition and other differences, necessitate a service which is capable of responding in a highly individualised way.

Genetic counselling services are provided by two groups of clinicians: Clinical Geneticists and Genetic Counsellors. Consultant Clinical Geneticists are doctors with specialist registrar training in clinical genetics following initial post-registration experience in a variety of disciplines, but usually paediatrics, adult medicine or oncology. Genetic Counsellors in the UK have varied backgrounds, but the majority have undertaken specific Masters level training to enter the profession and are recognised through professional registration (Genetic Counsellor Registration Board, 2012b).

There is an overlap in the roles of the doctors and genetic counsellors (Kerr et al., 2006). Doctors provide a greater focus on diagnosis and genetic counsellors on ongoing psychosocial support, but both are involved in risk assessment, genetic testing and communication of genetic information. For a more detailed overview of roles within genetic counselling services and their evolution, Walker’s summary is helpful (2001), as are many of the core texts used by genetic counsellors and doctors during their training (Harper, 1993; Uhlmann et al., 2009).

Given the complexity of genetic information, as well as its potential relevance to other family members, clients receive a detailed summary of the information discussed in clinic (Hallowell and Murton, 1998). This has been found to serve as a good reference in the future, sometimes years hence (Stayner and Kerzin-Storrar, 2004). However, use of written information clearly depends on the reading ability of the user. English is a second language for many Deaf people and the average reading age of a BSL user is between seven and a half and ten years (Solomon, 1994; Conrad, 1979; Powers et al., 1999). Whilst guidelines suggest that written leaflets should be pitched at a maximum reading age of 12 (Nicholls, 2003; Kent, 1996) and plain English used as much as possible in letters and leaflets, it is usually impossible to simplify complicated genetic information to this lower level. Deaf people may have extra difficulty in accessing accurate facts about genetic conditions and genetic counselling from other sources and therefore the information supplied through their contact with genetic counselling services is potentially even more important than for most people. In addition, the implementation of the Equality Act 2010 in the UK (UK Government, 2010; UK Government, 2011), following on from the Disability Discrimination Act 1995 (UK Government, 1999), means that there is now a legal obligation for public sector organisations to ‘take steps to meet the needs of persons who share a relevant protected characteristic that are different from the needs of persons who do not share it’ (UK Government, 2010 p96). For these reasons, there is a strong argument for genetic information resources to be produced in BSL as well as English and for pre-existing information in English to be translated into BSL.

Representation of BSL lexical items within the English text of the thesis

The English gloss of BSL lexical items is given using the conventions set out by Sutton-Spence and Woll (1999a). Where it is important for the reader to understand the form of the lexical item, it is described using the handshapes also summarised by these authors (ibid.).

‘Linguistic’ is used in the context of this research project in the sense of its first dictionary definition ‘of or relating to language’ (Collins, 2003). This definition is used in contrast to its second definition ’of or relating to linguistics’ (ibid.) where linguistics is defined as ‘the scientific study of language structure’ (ibid.). This distinction is important in that, while semantics is an important consideration during the literature review, and some of the major morphological features of BSL are described on occasion to explain how a concept is expressed, the study is considering language and communication in a broader sense than a linguistic analysis of BSL.

Conclusion

This introduction has set out factual information about d/Deaf people living in the United Kingdom, the provision of genetic counselling services through the NHS, the causes of deafness, the ways in which genetic counselling may provide information of value to d/Deaf people and the complexity of information about genetics and genetic counselling. These areas have been introduced to underpin the three subsequent chapters which explore, in turn, the different ways in which the experience of being d/Deaf have been constructed, genetic counselling services and their relationship with Deaf people as an exemplar of communication within health services, and the relationship between language, knowledge, understanding and communication. These chapters describe the landscape in which the argument is made for developing a method that allows Deaf BSL users flexibility of engagement in their preferred language(s), both to overcome potential barriers to full expression if there is only the option to use the majority spoken/written language (English), and to meet the rights of BSL users to have information and access in their own language. This is the essential backdrop to the thesis which seeks to develop a means of autonomous engagement of BSL users in research in order for them to shape the research that will ultimately fulfil the goal of improved access and services for Deaf people.

Chapter 1: The constructions of d/Deafness

> *'What though you cannot express your minds in those verball contrivances of man's invention; yet you want not speech, who have your whole body for a tongue, having a language more naturall and significant, which is common to you with us, to wit, gesture, the general and universall language of human nature.'* (Bulwer, 1644).

1.1 Introduction

Lane, in his seminal work, *The Mask of Benevolence*, posits two alternative ways of 'knowing' a culture: inside out as a Deaf person or outside in as a researcher (Lane, 1999 pXIX). The distinction is of central importance to this research, given that at the outset it is important to recognise that I am a hearing researcher. This positioning affects how I come to and present the literature review in this and in subsequent chapters. Where I stand and what I know (whether from inside or out) will change depending on the subject of the literature reviewed and its various aspects; I am not Deaf but I am a genetic counsellor, for example. Therefore, this chapter begins by exploring different perspectives towards deafness. It considers the reasons why deafness has historically been viewed almost exclusively from a medical and scientific viewpoint and examines why the histories of Deaf people have been less visible. The chapter thus aims to contextualise the published work within what is a contested field. It does not draw upon an exhaustive literature search, but presents the core texts and core arguments.

In deciding on those core texts and arguments, key works were identified through wide reading throughout the Deaf studies and disability studies literature, beginning with a focus on the prominent authors e.g. Bauman, Ladd, Lane and Oliver, and using their work to access the underlying philosophical arguments and texts. In parallel, discussion with colleagues with academic and personal understanding of the arguments around deafness and ways of being informed my understanding and identified further routes for reading. This chapter is an overview of these key arguments as a framework within which the subsequent literature can be understood.

1.2 Historical and philosophical constructions of hearing and speech

We create our individual realities through the perceptions gained through our senses, modified by our cognitive interpretations and influenced by the social world surrounding us. This statement's apparently simplicity is unpacked in Chapter 4, but is stated here to suggest why the majority hearing population re-enforces a shared interpretation of the world in which sound is crucially central to communication. The use of the voice by the majority of humans to communicate led Rousseau (1762) to justify the centrality of hearing amongst the senses because, he said, hearing alone is linked with an 'active organ' i.e. the voice. Rée (1999) explores some of the earliest philosophical explanations of the nature of sound and voice. He describes how Amman (1692) and Helmont (1685) both supported the view of the voice as a link to the soul, partly due to the idea of breath being an expression of the spirit of God and therefore the most direct means of expressing one's innermost being. Herder (1800), writing a century later, made the distinction between the expression of sound as 'narcissistic' (releasing the inner being) and the voice as 'echoic' (linking speaker to speaker). In this understanding, the voice became the 'artificial repetition or language' (ibid.). Amman (1700) had gone on to develop his earlier ideas, stating that 'human speech was no longer capable of 'vividly uttering the very essence of things''. He meant that the voice and words can only ever give an approximation of the meaning of something as it is filtered through the individual's own interpretation. This recognition by Amman that voice is not a direct link to reality is a fascinating forerunner of research into the link between meaning and language that is explored in Chapter 3.

1.3 Historical and philosophical constructions of signed languages

The common factor throughout the deliberations above was the centrality of hearing. Rousseau's claim that hearing is the only sense with which an 'active organ' is directly linked can be challenged: this may be true if voice was the only means of communication. However, signed language as a means of communication has been recognised since Graeco-Roman times. Socrates and Aristotle referred to deaf people signing (Ladd, 2003 p91) and the Turkish Ottoman Empire held signed languages in particularly high regard, having up to 200 Deaf servants at court who were seen as particularly discreet by virtue of not communicating by voice with the wider population. For both Deaf and hearing court members, it was 'of much use to those who attend the Presence of the Grand Signior, before whom it is not reverent or seemly so much as to whisper' (Miles, 2000).

Already this latter example raises the proposal that there were times and situations when signed language had distinct advantages over spoken language and there was recognition of this. However, these occasions were sporadic and infrequent over the last two millennia and, for most of this period and particularly through the majority of the twentieth century, the phonocentric (Derrida, 1976 p12) worldview has been responsible for the ways in which the general perception of d/Deaf people and signed languages have been constructed. Later sections discuss in more detail alternative perspectives of deafness and the individuals and movements who have argued that signed language is the equal of spoken language or even exceeds its capacity.

What Rée (1999) highlights in his framing of the historical background is a distinction between speaking a language and understanding it i.e. between language and communication. The earliest attempted ‘cures’ of deafness within Christian cultures and the desire to teach d/Deaf people a language were motivated by a wish to bring them closer to God as, without language, they could not access nor understand Christian teachings. Even some individuals now seen as central historical figures in the championing of signed languages, such as Sicard, had these same motivations: ‘the abbé Sicard…was both grammarian and priest; he labored to educate the impoverished deaf in order to save their immortal souls.’ (Lane, 1984 p13). Sicard later shifted his stance to value signed language as of equal value to spoken language in the communication of both concrete concepts and abstract ideas, but still used it in the context of a tool to teach speech, thereby ‘restoring’ d/Deaf people to the hearing world. Ladd also references signed languages as most commonly used in this way: as an ‘educational tool’ to ‘achieve normalcy’ (Ladd, 2003 p25).

However, arguments began to be advanced that signed languages were a means of communication equal to any other language rather than solely a tool. For example, Stewart (1983) criticised ‘the educators of the deaf for confounding ‘the gift of speech with the gift of reason’’. By this, he meant that d/Deaf people who learnt speech did not necessarily understand the concepts they were expressing through spoken language.

Both supporting and contradicting these perspectives were arguments proposed by philosophers from diverse areas of study as to whether d/Deaf Sign Language users could think in the same way and with the same complexity as hearing people. Much of the debate centred around whether sign or speech had been first to develop as a means of communication for humans. Tyler, an anthropologist who had made a study of the structure of signed languages, used it to illustrate his work on the origins of humans and argued that signs were ‘a more primitive language form’ than speech (Kyle and Woll, 1985b p47). In contrast, Condillac, a 17th century philosopher, argued that thoughts were based on images and ‘were not always representable in speech and were more directly related to gestures’ (Kyle and Woll, 1985b p47). He took the view that, in human development, speech had developed after gesture and that several words were often needed to represent one gesture. His argument has interesting parallels with current research investigating the modality-dependent resources of spoken and signed languages as discussed in Chapter 3 and investigated through this research.

Bringing the debate forward to the present day, although BSL has been recognised as a minority national and indigenous language (Department for Work and Pensions, 2004; Smith, 2003), social attitudes towards signed languages still tend to regard them as aids for a disabled group rather than equivalent to a spoken language. This was illustrated by two interviews, a fortnight apart, which were conducted by John Humphrys and James Naughtie respectively on the Radio 4 Today programme. The first was with Tomato Lichy, a Deaf artist and designer (BBC Radio 4, 2008b), and the second was with President Nicholas Sarkozy of France (BBC Radio 4, 2008a). In both interviews, interpreters were used, for BSL and French respectively. Although Humphrys began by explaining plainly that the voice heard on the radio would be that of the BSL interpreter voicing the BSL interpretation, it was clear later in the interview that he saw the interpreter as being for the benefit of the Deaf person rather than to enable communication between different languages:

> *Tomato Lichy: You say it's a serious disability. I disagree with that. We have an interpreter here for you to be able to understand me. If I go to a Deaf Club or a d/Deaf academic conference with thousands of Deaf people, you would be lost. You're the one with the disability, because you can't use sign language.*
>
> *John Humphrys: Isn't that a slightly perverse point? I, after all, don't need somebody to sign for me. I can hear the music of Beethoven or listen to a play by Shakespeare or pop music or whatever it happens to be - you can't. So therefore you have a disability. Surely that's simply a fact?* (BBC Radio 4, 2008b 3min 19sec)

The respect given by Naughtie to the French-English interpretation in the later interview was evident through the fact that the need for interpretation was not even commented upon: the President began to speak in French and it then faded after a couple of seconds into the English interpretation. This gave the impression of a meeting of two equal languages.

1.4 Hegemony and colonialism: the relevance of these theories in the relationship between the hearing and Deaf communities

The phonocentric worldview is understandable, to some extent, as it is much more difficult to imagine experiences outside one's own physicality. Many hearing people, probably the majority, do not question the status quo and this easy acceptance of what is 'natural' contributes to the dominant hegemony.

A starting position for understanding the relative positions of the alternative constructions of deafness is Gramsci's description of hegemony: 'the 'spontaneous' consent given by the great masses of the population to the general direction imposed on social life by the dominant fundamental group; this consent is 'historically' caused by the prestige (and consequent confidence) which the dominant group enjoys because of its position and function in the world of production' (Gramsci, 1971 p12). Gramsci balances hegemony against domination (by which he means the use of force) as a necessary pairing to maintain the current order, but judges that, in a democratic society, hegemony will almost always be adequate.

Marx (1887) argued that capitalism (and the importance of individualism within it) meant that economics was the single most important factor in determining the dominant hegemony. Gramsci (1971) highlights other public and private factors, particularly culture and the media, in reinforcing hegemony and Hamilton (1987 p8) summarises the insidiousness of these factors thus: 'It is the sheer taken-for-grantedness of hegemony that yields its full effects – the 'naturalness' of a way of thinking about social, economic, political and ethical issues.'. Gramsci's understanding of hegemony recognises the complexity and nuance behind evolving ideologies, not just seeing economics as solely dominant.

Jackson-Lears (1985 p571) explains how Gramsci posited the development of competing ideologies as new 'historical blocs' with both cultural and economic components. These could challenge, at times, the dominant hegemony and sometimes gain the ascendancy. Once established, less powerful minorities may comply with their subordination through unconsciously taking on aspects of the dominant culture. This does not need to happen through a concerted effort at 'brainwashing' on the part of government or other dominant group, but happens through the tendency of public discourse to highlight some forms of experience while ignoring or suppressing others. Thus the dominant hegemony is supported by the role of churches, schools and medical services, amongst others (ibid. p572).

When the concept of hegemony is considered in relation to the alternative constructions of deafness, it is clear that the hearing culture and the medical model of deafness within it are dominant in the majority of populations today – and have been for most of recorded history. Scientific and medical knowledge increased quickly from the Renaissance onwards (Gribbin, 2002) and were ahead of the social sciences in providing explanations of human diversity and cultures. Coupled with the obvious practical achievements of science in treating illness and prolonging life, these two factors provide particularly strong reasons why the medical model became pre-eminent and remained part of the dominant ideology. This medical model will be explored further in the next section.

In *The Mask of Benevolence* (1999), Lane develops the idea of colonialism to explain how the view of Deaf individuals and their community is constructed by the hegemonic hearing community. He draws parallels between the colonialism of African populations and the Deaf population by the hearing 'colonisers'' imposition of their own values and argues that the colonisers judge the members of the minority group by their own standards rather than understanding the minority culture as different, but not inferior. He illustrates this by the example of a comparison between the words used to describe African people and Deaf people in scientific studies of personality and psychology (Lane, 1999 p34-6). He finds great similarity in the choice of terms, which are predominantly negative. He attacks this hearing construction of d/Deaf people's personality traits by unpacking the reasons why testing would give different results and finds that, for example, tests were often administered in English rather than their national Sign Language.

He goes on to define 'audism' as a term to describe this objectification of Deaf people by the dominant hearing culture. Specifically, that audism is 'the corporate institution for dealing with deaf people, dealing with them by making statements about them, authorizing views of them, describing them, teaching about them, governing where they go to school and, in some cases, where they live; in short, audism is the hearing way of dominating, restructuring and exercising authority over the deaf community.' (Lane, 1999 p43). The crucial issue, and one that is returned to in the following section, is that audism does not require conscious acts by individuals in order to be present: it is the insidious influence of the dominant hegemony in all aspects of society's operation that results in audism permeating the interactions within and between Deaf and hearing people. If one accepts that Deaf people have a separate culture and community, then one also has to accept that, as a group, their alternative views and involvement in matters concerning their own deafness has been almost entirely absent from debate.

1.5 The medical model of deafness

There are two distinct ways of viewing deafness and, until one challenges oneself, the second – that deafness is a difference rather than a deficiency - may not even occur to a hearing member of a phonocentric community, partly because there is no reason to challenge the dominant hegemony as explored above. The majority population measures individuals against what is accepted as normal: if considering a single trait, this is likely to be both one that is common rather than unfamiliar and understood, or at least accepted, as normal by wide consent. Through these criteria, deafness is not seen as normal. If judged by a hearing person using the standard of what the loss of a sense means in relation to their own experience, it is likely to appear self-evident that a deaf individual would be disabled by that loss. From this constructed connection develops the medical model of disability as being solely focussed on the individual and the physical impairment or difference that they have.

Corbett (1996 p4) postulates that the reasons for this categorising of normal and abnormal include the human desire for individuals to separate themselves from the frightening ‘other’ that they do not understand. She argues that society exists by individuals finding their place within the hierarchy and that being able to label others as deficient in some way supports an individual’s self-esteem as ‘normal’. These often unconscious motivations may well be a true part of the picture, but it can be argued that another part is the relative difficulty all humans face in placing themselves in another’s shoes and experiencing the world as others do. As Rée (1999 p17) explains at the start of his exploration of the nature of sound, he would imagine himself as a child losing his hearing and how he would experience that. Through this imagining, it would be almost impossible to gain an accurate impression of what it would be like to be born deaf as he was measuring deafness against his knowledge of hearing and sound rather than defining it in its own right. Inverting this idea to consider the experience of deafness from the perspective of a person born deaf, it explains why Deaf people reject the description of themselves as having hearing loss: it is illogical to use the term ‘loss’ when hearing has never been experienced.

Lane (1999 p24) explores how it was that medical professionals were given power to define deafness through this model and suggests that it is related to the normal and abnormal as discussed above: hearing parents whose child is born with or develops deafness find the loss of communication very difficult to manage and seek help from medical professionals to try and remove this difficulty. He argues that it is medical professionals’ role to diagnose and label infirmity so that cures can follow and, from this, deafness could easily be fitted into a category of loss. From the perspective of those working in a medical field, there would be the argument that they have a strong altruistic motivation for trying to help those who are deaf gain hearing. Their role is to aid the individual to regain their health and faculties as completely as possible and it is, therefore, their duty to do all they can to achieve this. It cannot be denied that deaf people do not have a sense that the majority of people find extremely useful and a great source of pleasure. However, the difficulty with this argument remains that the measure of lack of hearing as a deficit is made in the context of a phonocentric hegemony.

Lane (1999 p24) gives an example of a cochlear implant specialist who, when asked by a Deaf member of an audience to whom he was presenting whether he also explained to parents the alternative options of signing and participation in the Deaf community, responded that ‘We tend to present things from our point of view’. There may be multiple reasons for this response: the professional’s own lack of knowledge about the alternatives, possibly his reluctance to challenge the parents’ own medical perspective on deafness at a time when they are dealing with their child’s recent diagnosis and probably because he wishes to help the family - and something concrete like technological aids to hearing is judged, under the medical model, as something far more than just exploring other ways of looking at the deafness.

The impact on Deaf people of using purely the medical model to define deafness and the lack of recognition of language and cultural differences is argued to be great. Lane (1999 p23) comments that d/Deaf people are not participants in research about deafness, but are ‘merely…passive objects of that research’. To give two specific examples, he firstly quotes research that showed that the variation in the way intelligence tests were administered to deaf children could affect the ‘measured IQ’ by up to thirty points (Lane, 1999 p51). He goes on to illustrate a medical dismissal of signed languages thus: ‘‘As hearing children grow up,’ a 1978 textbook on the ‘psychology of the deaf’ explains, ‘they replace physical violence with verbal aggression. But the deaf cannot do this as they have no language.’’ (Colin, 1978 p82; quoted in Lane, 1999 p54).

One area not yet touched upon and yet which will be crucial to this study is the semantics and pragmatics of language. Jackson Lears (1985) references the importance of the choice of words in enforcing the dominant hegemony. He discusses the work of Bakhtin, a Russian literary critic who emphasised culture ‘as a many-voiced conversation’ where multiple meanings of the same terms showed a ‘plurality of value-laden perspectives’ (ibid. p591). In the context of a hegemonic culture, this choice of language can be ‘internally persuasive rather than merely authoritative’ (ibid.). The application of these ideas to deafness is powerful: ‘hearing loss’ and ‘hearing-impaired’ are two examples of terms used commonly in medical settings, but which members of the Deaf community, as previously discussed, would reject or define in alternative ways from their own perspectives. At the risk of deviating briefly from the aim of this chapter, this also highlights the importance of this study and the careful choice of methodology: the same cultural meaning of terms, once translated into BSL, cannot be assumed – words are not neutral, even if attempting to define an objective scientific concept.

The dominance of the medical model is accepted. However, there are some issues that should be discussed in relation to some of the medical and educational evidence presented by Lane as discredited. For example, he reports a study that found the proportion of children with severe emotional disturbance in a school for d/Deaf children to be five times higher than in comparable schools for hearing children ((Schlesinger, 1985 p106) quoted in (Lane, 1999 p56)). He presents this as audist misjudgement of d/Deaf children’s behaviour due to a lack of awareness of cultural differences and this may, indeed, be a significant factor. However, it could be argued that to give this single explanation for all this difference would result in missing actual problems and therefore would do a disservice to the children. Hindley (1997) argues that there is a real increase in the incidence of psychological problems in d/Deaf and hearing-impaired children (he defines these two groups on the basis of their educational setting). He explores several possible causes of this increase and concludes that the most important are communication problems and the influence of these on relationships and individuals’ self-image. The cause of these communication problems could also be explained by the surrounding audist society, but the problems described by Hindley could have a real impact on the d/Deaf individual’s mental health as well as the perceived difference from their hearing peers to which Lane refers. This distinction is important. Both aspects are part of the relationship between individual and environmental factors which defines the extent to which a person is disabled by a physical difference: this relationship is explored in the next section.

Ladd (2003 p35) asserts that the medical model is ‘deliberately constructed’. However, it can be argued that a distinction should be made between a fully conscious ‘deliberate construction’ on one hand and, on the other, compliance with the status quo by definite actions, but actions that are carried out in an unconscious way because one is too busy or has not had the impetus to challenge oneself to recognise the prevailing hegemony and one’s acceptance of it. This latter position may, for example, apply to many health professionals working within an NHS system which, almost without exception, considers deafness in the medical sense. This compliance with the status quo could be said to be deliberate construction in the sense that nothing concrete has been done to stop it, but there is no evil mastermind constructing this model. This argument against Ladd's completely clear demarcation is supported by Jackson Lears (1985 p587), who comments that 'to avoid getting shot down, proponents of hegemony should beware of attributing a single mentality to large institutions' and adds 'abandon any assumption that there is a straight line linking intentions, actions and effects' (ibid. p588).

1.6 Models of disability

This section begins with an exposition of Oliver's landmark book, *The Politics of Disablement* (Oliver, 1990), which constructs a persuasive argument to explain the political frameworks that have shaped developing models of disability. A central part of the argument is summarised by Stone (1985) thus: 'The definition of disability against 'the norm' is a shifting ground which, it is argued, is necessary to fit the economic and political need of the society so that the balances of production and need are kept under control' (quoted in Oliver, 1990 p40).

Comte's 'evolutionary model' (1855) suggested two main factors influencing the view of disability: the 'mode of thought' and the 'mode of production'. The mode of thought or human intellectual process breaks down into three consecutive stages of development:

…each of our leading conceptions…passes successively through…the Theological, or fictitious; the Metaphysical, or abstract; and the Scientific, or positive. In other words, the human mind, by its nature, employs in its progress three methods of philosophizing, the character of which is essentially different… (ibid. p27)

In other words, as paraphrased by Oliver (1990 p29), these stages mark a shift from 'a religious interpretation of reality to a more naturalistic one and finally to a scientific way of understanding both the natural and social worlds'. These are mirrored by a tendency for society's view of disability or, in a broader sense 'deviance', to shift from moral problem to legal and then to medical (ibid.). As a result, the 'management' of disabled people in society has moved from it being a religious concern to a legal one (particularly manifested in the institutionalisation of disabled people), to a medical framework (which has latterly been linked to the move towards community management of people with disabilities).

The mode of production is separated into the consecutive steps of 'spontaneous participation' (feudal society), 'separation…from the rest of society' (capitalist society) and ‘integration’ (socialist society) (Oliver, 1990 p26). Oliver explains the shift in the view of disability to individual pathology (which is still the prevalent view) as a development caused by capitalism, which encourages a focus on individual able-bodiedness, the development of wage labour and the ability of the individual to make an economic contribution. In this model, the focus shifts from ‘the individual’s contribution…to the family, the community…in terms of labour’ and disabled individuals become excluded (ibid. p47).

Several theories have been advanced to explain how disabled people are treated in society and Oliver postulates that the reality is likely to be a combination of these theories working together. Abrams’ ‘enlightenment theory of social welfare’ argues that what happens to disabled people is due to ‘the progressive evolution of reason and humanity’ (Abrams, 1982; quoted in Oliver, 1990 p37). An alternative model is Marxist in that the mode of production is the crucial factor and is labelled by Abrams as the ‘necessity theory of social welfare’ (ibid. p38). The ‘action theory of welfare’ proposed by Parker ((1988) quoted in (Oliver, 1990 p38)) suggests that additional factors such as families’ views and the intentions of people working in institutions complicate the issue. It is also significant to consider the ‘power theory of welfare’ (ibid.): why some groups, particularly the medical profession, have much greater influence on the management of disabled people.

This last point is of particular relevance to the issues raised in the previous section. Oliver brings together four main theories of medicalisation and argues that a combination of the four influences is likely (Oliver, 1990 p49). The first of these theories is ‘enlightenment’, which sees medicine as progressive and predominantly a force for good, influenced by the ‘rise of science and the progress of humanitarian ideas’; the second is ‘necessity’, which sees medicine as assuming an important role in social control; the third is ‘action theory’, which highlights the ‘struggle between various groups to impose their own specific set of meanings upon particular social phenomena’ (ibid. p50) and the fourth is the ‘power theory’ which has three strands: the perceived ‘superiority of medical knowledge’, the good organisation of individuals within the medical profession and the strong connections with the ‘ruling class’ (ibid. p51).

Stone (1985) adds a further layer to the explanation of the changing perception of disability: that it is not capitalism in itself that is the influence, but the tendency of increasing ‘rationalisation’ (ibid.) to accompany capitalism. By this, she means the increasing standardisation of disability and the measurement of work and need. Oliver (1990 p40) later brings in Stone’s views again to support his argument for what he sees as the central role of the medical profession in defining disability: Stone (1985 p51) argues that doctors were reluctant participants in the increasing standardisation and measurement of disability mentioned above, but that they took on the task believing that a separate group of government doctors might be sanctioned to carry it out if they did not.

So the current prevalent view in the western world is disability as personal tragedy with the focus squarely on the individual's physical impairment (Oliver, 1990 p1). Oliver strongly supports the argument that disability should be deconstructed into different layers of experience. He uses the views of Brisenden (1986) to illustrate this: 'In order to understand disability as an experience, as a lived thing, we need much more than the medical 'facts', however necessary these are in determining medication. The problem comes when they determine not only the form of treatment (if treatment is appropriate), but also the form of life for the person who happens to be disabled.' (Brisenden, 1986 p173).

Oliver (1990 p4) critiques the *International Classification of Impairments, Disabilities and Handicaps (ICIDH)* adopted by the World Health Organisation (WHO) (Wood 1981) that was current at the time he was writing and argues that it inadequately incorporated the social aspects of disability. Although this scheme accommodated a distinction between individual impairments and external disabling factors, Oliver argues that the focus and choice of language was still medical and grouped disabled people as separate from the normal majority. The WHO scheme used a three level measurement: 'impairment as abnormality in function, disability as not being able to perform an activity considered normal for a human being and handicap as the inability to perform a normal social role' (Oliver, 1990 p4). Oliver prefers to advance the definition given by the Union of the Physically Impaired against Segregation as being an acceptable summary of the social model of disability:
Impairment: lacking part of or all of a limb, or having a defective limb, organism or mechanism of the body;
Disability: the disadvantage or restriction of activity caused by a contemporary social organisation which takes no or little account of people who have physical impairments and thus excludes them from the mainstream of social activities (UPIAS, 1976 p3-4).
What this definition does not include is a distinction between limitations which could be ameliorated by societal changes and other personal limitations in function which could not be adequately overcome by environmental adjustment.

In 2002, WHO produced an updated version of *ICIDH*, the new title of which emphasised the change in focus: the *International Classification of Functioning, Disability and Health* (ICF) (World Health Organisation, 2002a). This framework is intended to aid the description of both disability and health states for use in research and policy planning. The introductory document stressed the aim of moving away from the individual as the sole focus for measuring disability: ‘Previously, disability began where health ended; once you were disabled, you were in a separate category. We want to get away from this kind of thinking. We want to make ICF a tool for measuring functioning in society, no matter what the reason for one’s impairments.’ (World Health Organisation, 2002b p3). WHO described the scheme as recognising the validity of both the medical model and the social model by integrating the two into the ‘biopsychosocial model’ (ibid. p9) with biological, individual and social aspects of health. The dimensions of this new model are body functions, body structures, impairments of function or structure, activity (the limitations of which are equivalent to disabilities in the previous model), participation (the restrictions of which are equivalent to handicaps) and, additionally, measures of the environmental factors (physical, social and attitudinal) which surround the individual (ibid. p10).

These distinctions allow a more accurate measure of the practical impact of deafness on everyday life for a Deaf individual who predominantly uses BSL. In particular, when looking at activity and participation, two separate areas are considered: ‘performance’ (problems encountered in the person’s current environment) and ‘capacity’ (limitations experienced by the person if they are without necessary assistance or adaptation) (World Health Organisation, 2002b p2). A Deaf person living with relatives who use a signed language and working in an environment where communication issues had been overcome would not measure their performance as affected in any way by their impairment. Living within a hearing majority population, their capacity would certainly be affected, but it is clear that this is a function of the relationship between the person and their environment rather than an individual problem. This allows a clear shift in perspective from the medical model to a social model, particularly when the individual themselves does not consider themselves disabled.

Oliver’s (1990) core argument is that it is the movement towards individualisation that has altered the focus of the ‘problem’ of disability onto the individual rather than society. It should be recognised that society is responsible for the prevailing view as well as being the solution to disability – that society can change and that this should be viewed as a more morally correct approach than focussing on altering the individual person and hence losing some of the diversity which enriches the population.

Deafness is not mentioned widely in Oliver's book, but reference to the shifting definitions of particular conditions is made often: 'A disorder in one place and at one time is not seen as such in another: these social perceptions and definitions influence both the provision of care, the demands of those being cared for, and the size of any count of health needs' (Susser and Watson, 1971 p35) quoted in (Oliver, 1990 p13). This recognition of the potential for future change is highly relevant to this thesis as deafness is certainly a 'disorder' (ibid.) that is seen in different ways by different groups. The societal conditions which have led to the dominant medical perspective may shift in the future such that a social definition of deafness gains more acceptance.

Oliver finally links his argument back to hegemonic principles, describing ideologies such as 'disability as personal tragedy' as becoming 'common sense' because they are so 'deeply embedded in social consciousness generally that they become facts' (Oliver, 1990 p80). He ends on the point that disability movements have not yet achieved equality by any stretch, but have been successful in getting opposing views to those of the existing hegemony on the agenda. This leads us onto the next stage: the alternative constructions of deafness.

1.7 Deafness defined as a cultural and linguistic difference

This section presents the argument that contrasts the medical definition of deafness as the loss of a sense and defines deafness instead as an integral condition of the body. As Ladd points out 'the issue of loss has no meaningful reality' [for congenitally deaf people] (Ladd, 2003 p14). The Deaf community experience the world visually with a depth which may well surpass that which can be achieved by a hearing person, yet as a minority culture, the issue of missed visual experiences by the hearing majority is never discussed.

Ladd argues that Deaf people are 'organic and linguistically whole beings' (Ladd, 2003 p72) and Kyle and Woll second this in saying that 'deaf culture, like all cultures, is carried through the language' (Kyle and Woll, 1985b p9). Bauman's (2005) paper defining communication between Deaf and hearing people as the barrier that defines the 'contact zone' is extremely helpful in highlighting the equality of the Deaf and hearing worlds. The paper begins by reviewing the 2002 case of the Deaf lesbian couple from the US who aimed to increase their chances of having a deaf baby by choosing a Deaf sperm donor. He argues that both sides are so influenced by the medical model of deafness that it 'obscures' part of the argument and proposes that we need to reframe deafness within the social model in order to lead us to 'the ironic claim that seeking a deaf child could actually mitigate the conditions of disability within the family' (ibid. p311).

The definition of the 'contact zone' is the point where the hearing and Deaf worlds meet and this highlights that, if all the family are deaf, then the hearing world is not within the family home even if it comes right to the front door. The scene is then set for 'a visually centered episteme to emerge that results in lived experiences not predicated on the lack of a sense, but on the plenitude of a visual culture with its own norms of language acquisition and identity development.' (Bauman, 2005 p313).

The paper also makes the point that being hearing is indeed more convenient than being deaf, but this, in itself, does not 'lead to an increased value of one's life'. In response to a newspaper article written by Jeanette Winterson (2002), Bauman adds that it is not a 'simple fact' (Winterson, 2002) that being hearing is better than being deaf. Indeed, the author comments that 'one would also wonder how Winterson would respond to the claim that it is a simple fact that it is better to be heterosexual than homosexual' (Bauman, 2005). Clearly Deaf people have a disability while living within a phonocentric society, but, returning to the previous section, they would argue that, despite having an impairment within that definition, they are not disabled in any way when their linguistic preferences are allowed.

Ladd argues that deafness should be defined not by the medical model, nor by the social model, as both see deafness as problematic, albeit the latter as a result of the surroundings rather than the individual. Rather, deafness should be defined by a third positive model: the culture-linguistic model (Ladd, 2003). He highlights the problems in successfully putting this case: 'therein lies the crucial distinction between majority and minority cultures – the former are under no obligation either to make explicit the beliefs which drive their actions, let alone to have to justify their actual existence.' (ibid. p21). There is a 'double yoke' of not having the resources within the community to enable them to argue their case effectively, while the surrounding majority culture means that it is almost always members of that culture that carry out the investigation of the minority group.

In support of this argument, Kyle and Woll (1985b) add that 'it is evident that [the Deaf community] is not to be assessed in terms of a population driven together by lack of some skill (such as speech or hearing), but rather, one brought together by a conscious desire to interact meaningfully and to share information and (perhaps as a direct result) to share experience and belief' (ibid. p21). Language is the binding force leading to the development of a unique culture and has three particular roles: as ‘a symbol of social identity, a medium of social interaction, and a store of cultural knowledge’ (Lane et al., 1996 p67). The first of these – social identity – is addressed in section 1.8 as part of the discussion about individual identity. The opportunities for social interaction in a signed language are limited for most Deaf people by the reality of being isolated within the hearing population for the majority of home and work life. For this reason, the opportunities themselves have historically been a central part of Deaf culture: residential Deaf schools and local Deaf clubs. The Deaf community in the UK is strongly opposed to mainstreaming of d/Deaf children into hearing schools (Ladd, 2003 p43), seeing this as depriving these children of the opportunity to learn the language and culture which comes most naturally to them. Even when the residential schools are predominantly oral/aural in teaching approach, communication amongst children outside the classroom is in Sign Language (Lane et al., 1996 p425). Deaf clubs provide a similar opportunity for Deaf adults to socialise in a relaxing situation where they can use Sign Language and, in addition, keep in contact with friends, many of whom will have attended one of the same small number of residential schools (Kyle and Woll, 1985b p11). Many Deaf clubs have a long history, often having grown from school alumni societies or religious meetings (ibid.). There is a long tradition of Deaf clubs from different regions meeting up for sport or social events (Lane et al., 1996 p131) and links between clubs led to the development of national organisations. For example, Manchester was the setting for a meeting of twenty Deaf representatives in 1877 who came together to set up the National Deaf and Dumb Society – the forerunner of the British Deaf Association (Kyle and Woll, 1985b p11). The fight for self-determination against the dominance of hearing professionals has been a constant feature throughout the history of Deaf organisations and perhaps had its finest hour with the successful outcome of the campaign for a deaf candidate to be appointed as the first d/Deaf president of Gallaudet University in 1988 (Lane et al., 1996 p128).

Returning to the third feature linking language and culture – that of language as a store of cultural knowledge – many Deaf cultural mores are directly related to the visual nature of the language. For example, taking turns to speak is signaled in a very different manner: it is impolite to start speaking while another is still doing so and, in any case, if the current speaker is looking elsewhere, one would not be seen. Rather, when the speaker has finished what they are saying and is ready to give permission to hand over the conversation, they will look at the other who is clearly ready to respond. Similarly, secrecy is not seen as a virtue within Deaf culture, possibly as a result of the difficulty of having a private conversation in a signed language, but also because openness and directness are valued and therefore secrecy can be seen as rude (Lane et al., 1996 p74). A final example of the blending of Deaf culture and language is the fact that Deaf individuals have their own name sign: not the alphabetical finger-spelling of their English name, but a single sign that distinguishes them, perhaps by physical appearance or character or a combination of features.

Lane et al. (1996 p159-161) define four characteristics of minority groups that can explain why individuals within it bond strongly such that a separate culture develops. These are: a shared physical characteristic such as deafness or language, individuals choosing to identify as members of the group and others recognising them as such, marriage/partnership within the group, and external oppression directed towards group members. The Deaf community fulfils all these criteria: the first and last have been discussed already and would both be strong explanations for why finding a partner within the group is common – 90% of Deaf people have a partner who is also d/Deaf (ibid.). The second characteristic – that of individual belonging in the minority group – is revisited in section 1.8.

There is persuasive evidence that deafness should, indeed, be defined through a cultural and linguistic model if it is also accepted that Deaf people themselves, who do not experience their deafness as a disability, are self-evidently the best-qualified group to make this decision.

It may be a bold suggestion, but it could be argued that scientists with a working knowledge of and reference to the theory of evolution should find it easier than others, despite a positivist background, to see the current status quo of typical hearing and deafness (and, indeed, humans themselves) in the world as transitory and based on an infinitely complex set of conditions that make it even more important not to make judgements on what is 'normal' and what is 'other'. It is impossible to know whether tomorrow, the 'other' could not be better adapted. That is only one possible way to understand conflicting perspectives, but calling on evolution highlights that our judgements on what is 'normal' are hugely influenced by society in the widest sense: biology, genetics, geography, psychology and philosophical theories.

Taking a slightly different perspective, Menand (2004) optimistically wrote in his essay on '*The Science of Human Nature and the Human Nature of Science*' that 'There is intelligence in the universe. It is ours. It was our good luck that, somewhere along the way, we acquired minds. They released us from the prison of biology.' (ibid. p20). This use of our intelligence to release us from biological restrictions could be interpreted in two ways: that intelligence allows us to develop technological ways of ameliorating barriers caused by physical limitations and that we are consequently able to avoid potential restrictions imposed by them. This interpretation uses a medical model of biological restriction. The alternative interpretation is that our intelligence allows us to see other ways of measuring an individual beyond medicine and evolution: socially and individually-constructed realities are just as valid, but must be understood in their historical context in order to avoid looking at them only from the perspective of the dominant culture.

1.8 Development of Deaf-World and Deafhood

Kyle and Woll (1985b) state that ‘Just as in any group separated in some way, deaf people through their language negotiate and agree on a construction of reality’ (ibid. p50). That identity of the Deaf community has evolved greatly since the revival of signed languages beginning in the 1970s.

Hearing society in western cultures is much more focussed on the individual. Deafness is a culture that forms around groups, memories and, like Queer culture, is not intergenerational for the majority of Deaf people. In the past it has not been written down, though the explosion of video blogs and alternative ways of recording experience could change this. Part of the definition of being Deaf is being part of the community. Mindess (2006) suggests that the Deaf community is not in the minority here in that 70% of the world’s population live in cultures of ‘collectivism’ rather than ‘individualism’. In addition, there is a ‘strong belief in cultural collectivism’ (Ladd, 2003 p58) within the Deaf community, as exampled by the fact that the Deaf academics and professionals are not always the natural leaders because of the Deaf belief in working together from the grassroots: ‘attitude is…all’ (ibid.).

Ladd (2003 p40-1) uses two definitions of community that he feels are particularly pertinent to the Deaf community: ‘the quality of holding something in common, as in community of interests’ and ‘a particular quality of relationship (as in communitas)’. He explains that ‘Deaf world’ was the collective term of choice until about twenty years ago, but this term slowly gave way to ‘Deaf community’ as a recognition that Deafness is one of many communities rather than a ‘binary opposition of ‘hearing world’ and ‘deaf world’’ (ibid. p41). The identity of the community is continuing to evolve and be debated. Over recent years, the term ‘sign language community’ has sometimes been used instead of ‘Deaf community’ in recognition of language’s crucial role in community. For example, the British Deaf Association (2014a) rebranded themselves several years ago as ‘Sign Community’ with ‘British Deaf Association’ in smaller letters under the main logo and ‘unity’ within ‘Community’ highlighted. However, they have recently reverted to British Deaf Association to make clear their focus on campaigning and empowerment led by and for Deaf people.

Ladd (2003 p3) made a significant contribution to the debate in coining the term ‘Deafhood’. This term tried to grasp a ‘deaf epistemology’ (Young and Ackerman, 2001; Young and Temple, 2014a p36): a way of collecting the grassroots views of Deaf people so as to understand the Deaf community from the inside looking out rather than, as has always been the case previously, views of the Deaf community being made by (predominantly) hearing researchers looking in.

Ladd points out that the point has been reached where Deafness has attained some cultural power through visibility and influence. For example, pop groups use signed songs on stage, Marlee Matlin acted in a mainstream US television series as a signing Deaf political professional and there is greater awareness of signed programmes. However, he argues how much further there is to go by highlighting that other minority or potentially disadvantages groups such as Black people, LGBTQ people, and women now have broad agreement with their perspectives from society which is not based in any way on ‘charitability or kindness’ (Ladd, 2003 p27). In Deaf and disabled issues, by contrast, ‘the mask of benevolence’ (Lane, 1999) is still visible. This is particularly evident in the way services are provided. For example, Deaf people are forced to acquiesce to the recognition of their deafness as a disability to gain access to benefits such as Access to Work that will pay for interpreters in order to remove the communication barriers that handicap (World Health Organisation, 2002a) them. A Deaf colleague (Rogers, 2008), during a discussion about genetic counselling services for deafness, pointed out that it was necessary for a Deaf person to collude in the recognition of deafness within the medical model in order to attend the clinic.

In summary, there is not yet economic or political power, but a new ‘historical bloc’ (Jackson Lears, 1985 p572) is emerging which may yet challenge for a more prominent position in the current hegemony.

1.9 Deaf Gain

Since 2009, there has been increasing discussion of Bauman and Murray’s (2009) posited concept of Deaf Gain – a ‘reframing of “deaf” as a form of sensory and cognitive diversity that has the potential to contribute to the greater good of humanity’ (ibid.). They argue that Deaf people contribute to three types of diversity: cognitive, cultural and creative through their language and their visual way of experiencing the world.

1.10 Individual constructions of Deaf identity

Deaf identity is something that one is given through acceptance by the community as much as something one chooses to take on. One widely used model is that described by Baker-Shenk and Cokely (1980 p54). Their model for membership of the Deaf community is based on ‘strength of positive attitude’ in four areas: audiological, linguistic, social and political. If an individual is particularly strong in some areas, then level of deafness and competency in BSL may not actually be the most crucial factors. However, the fact that there is ongoing debate about whether hearing children of Deaf adults (known as HMFD in the UK – hearing, mother father Deaf to reflect the BSL word order and CODA in the US – child of Deaf adults) can be fully part of the Deaf community, shows that some audiologically-measurable deafness is an important prerequisite for membership. Likewise, as BSL is the language of the Deaf community in the UK, at least using it sometimes would be a prerequisite. A signed language is such a strong part of the political argument for the existence of the community, as discussed in section 1.6, that without any Sign Language, an individual would have great difficulty being seen as attitudinally Deaf.

Other factors likely to influence an individual’s position with regard to their own deafness and their place in the hearing and/or Deaf worlds are their family and friendship networks, their choices of modes of communication and discourse and their place of education. The last of these is likely to have had a powerful influence on the first two for the reasons discussed in previous sections. Corker (1996 p33) highlights the influence of poor self-concept on the positioning of the individual: she stresses the impact that ‘vertical relationships’ with hearing family and professionals and ‘horizontal relationships’ with peers like themselves, such as would happen in a residential school, have on the development of an individual.

Jackson Lears (1985 p578) comments further on the feelings that an individual has internalised about their deafness and discusses the ‘hidden injuries’ that a member of a minority takes on board and that shape them as they grow. He also highlights the influence that one’s private situation can have on a lack of political awareness (and further relates this to the wider public consequences resulting from a lack of grassroots involvement). This is particularly pertinent for d/Deaf people as the majority grow up in hearing families with the resulting lack of access to background spoken conversation and information. Deaf people commonly grow up with mediated communication being standard, either through an interpreter or by hearing people modifying their communication. This gives a constant experience of not being able to determine one’s own safety, not being able to control who one can communicate with and being reliant on other people. These factors could all play a part in forming the individual (Napier et al., 2017).

Corker (1996) writes about the internalised struggle that individuals go through as they learn to balance their autonomy and self-esteem against the pull of intimacy and belonging to a group within which they feel accepted. She points out that different groups ‘have a series of *a priori* narratives which impose relatively closed systems of understanding on people’ (ibid. p19). In addition, if the individual has a minority difference such as deafness, they will have been under pressure from childhood to meet the expectations of the hegemonic culture before they reach the point where they can ‘‘come out’ as a member of that oppressed group’ (ibid.). This means that an individual may go through a series of transitory self-identities during childhood and on into adulthood. Carty (1994 p40) suggests the following developmental stages during an individual’s adjustment to their d/Deaf identity:

- Confusion – arising from the realisation that one is not the same as everyone else
- Frustration/anger/blame – as responses to a lack of understanding or acceptance of difference on the part of others
- Exploration – deciding to explore self-identity options with deaf and/or hearing communities
- Identifications and rejections – during the ongoing process of exploration
- Ambivalence – following the recognition of negative characteristics of the group with which one is identifying
- Acceptance – of one’s personal and social identity once one has sufficient information and experience (ibid. p42).

Corker (1996 p59) refers to Carty’s (1994) developmental stages, but highlights that the boundaries between groups within the d/Deaf community are less clear than the boundaries between racial or cultural groups or groups of people with different disabilities. This is because of the fluid and personal adjustments to communication that d/Deaf people have to constantly make based on their surroundings. Not only does this mean that it is more difficult for the d/Deaf community to present a united front politically, but it is more difficult for individuals to find and negotiate their position within the community.

Writing as an experienced counsellor working with d/Deaf people, Corker (1996) stresses the importance of an individual reaching a balance between the inner identity ‘that values honesty over conformity’ (ibid. p61) and the outer constraints of identity that influence how one is seen by others and that are imposed by membership of a socially constructed group.

1.11 The definition of Deaf in the context of this thesis

‘Deaf’ will be used throughout this thesis to be inclusive of any person with an audiologically-measurable deafness who signs, at some times and in some situations, with any level of fluency and, in addition, has an understanding of Deaf culture and considers themselves part of the Deaf community to any extent. The author is a British Sign Language user, has good awareness of Deaf culture and contact with many members of the Deaf community, but is not d/Deaf and therefore is defined as a researcher outside the group being studied.

Chapter 2: Genetic counselling services, their development and relationship with society and Deaf people

2.1 Introduction

The context for this chapter is set out in the introduction to this thesis. The introduction includes a description of modern genetic counselling from the perspective of the professionals that deliver it together with the structure of genetic counselling services in the UK. It includes a brief summary of the causes of deafness to explain why genetic counselling has relevance for Deaf people in relation to their deafness. It also highlights that Deaf people have the same chance as the rest of the general population of having an unrelated genetic condition or illness themselves or in their family history.

This second of three literature review chapters takes the same approach as Chapter 1 by exploring how the subject under scrutiny has been socially constructed. It considers the place of genetic counselling services within society and looks at where and why there are difficulties in the relationship between such services and some groups in society, including Deaf people. It illustrates that being aware of different views towards the genetic counselling topics addressed during a consultation can contribute to clearer understanding on both sides. In other words, communication is not only about the practicalities of individuals speaking the same language (literally or metaphorically), but to what extent participants in a communicative exchange understand each other’s position with respect to a particular social construction – that might be genetic counselling or it might be the experience of being Deaf.

The literature that informs this chapter encompasses both non-empirical and empirical research and argument. The specific reach of the chapter means that, as a professional working within the clinical field, I began with a strong working knowledge of the existing literature. Continuing literature searches built on this initial knowledge through the research period. A systematic review was not carried out. Literature searching was performed as an iterative process including review of the contents pages of the following key journals in genetic counselling and research involving d/Deaf people between 2007 and the present:

Journal of Genetic Counseling; Social Science and Medicine; American Journal of Medical Genetics; American Journal of Human Genetics; Journal of Medical Genetics; Clinical Genetics; European Journal of Human Genetics; Journal of Deaf Studies and Deaf Education; American Annals of the Deaf; Sign Language Studies.

As well as the above, additional publications by key authors were reviewed, citation tracking (Greenhalgh, 2006 p24) was used to look forward at other papers that had cited those already identified and reference lists were studied to identify earlier papers. This approach became easier as the digital resources increased in power and could also suggest related papers. At the start of the period of study, Ovid was used to search Medline, PsychInfo, and CINAHL simultaneously. From 2009, Web of Knowledge was used in preference to Ovid for its ease of use and reach. In January 2011 the University adopted Ex Libris/Primo Central and in mid-2013, the Library Search function, built by the University to work within this system, became the single search point for electronic content within all the e-journals and e-books to which the University subscribes as well as all articles within Primo Central. The latter encompasses all the above subscribed collections and, in addition, a large amount of grey literature (University of Manchester, 2015; Ex Libris Group, 2015). I have used Library Search since shortly after its adoption.

Although the body of relevant literature that these approaches identified was considerable, the aims and objectives of the thesis overall, and of this chapter specifically, guided what was discarded as well as the approach to appraisal. The non-empirical literature was chosen for its relevance to and strength in supporting the key arguments, particularly in the first half of the chapter. The principal writers were identified within the disciplines of genetic counselling, Deaf studies and disability studies. The empirical literature was not appraised by direct use of standard frameworks or checklists such as Popay et al. (1998) or CASP (Critical Appraisal Skills Programme, 2013). Rather, it was appraised with knowledge of the common content of these tools and with reference to widely agreed standards of excellence e.g. clearly stated aims and objectives, a clearly defined sample, comprehensive methodology/methods, evidence of ethical approval. The papers’ inclusion in high quality peer-reviewed journals was also taken as a likely measure of quality, but this was not assumed.

The chapter begins by summarising the development of clinical genetic counselling services as a distinct speciality within health services, the influence of society on the path this development has taken and, indeed, why genetic counselling is positioned within health services. The rich canon of research that has developed symbiotically alongside the genetic counselling clinical service is used to illustrate genetic counsellors’ growing understanding of the process and outcomes of the service they offer. The chapter argues that the key concept underpinning all other processes and outcomes within genetic counselling is effective communication. ‘Effective communication’, in itself, is a phrase which must be unpicked and defined, but this discussion is part of chapter 3. If this assertion is accepted, then a priority within genetic counselling research is the investigation into communication. However, the role of communication within a process or outcome is not always made explicit. The argument is illustrated by considering one of the most central and referenced components of genetic counselling: *non-directiveness* in the provision of information and facilitating of decision-making and the ongoing debate as to whether it can and should be achieved. It is argued that the fundamental tenets of genetic counselling, such as non-directiveness and maximising the autonomy of the client, can be seen as part of a distinct professional genetic counselling culture. Such a culture allows individuals within it to understand each other in cultural shorthand. However, there is therefore a need for these professionals to remain aware that their clients and others outside may not share the same view, either though lack of knowledge and/or because of their own cultural values and constructions of genetic counselling services.

There are known challenges and barriers in the interactions between Deaf people and health services as a whole. The chapter moves on to explore what is known about the nature of these barriers within genetic counselling, but also in broader health settings where there are understandings that are transferable to the genetic counselling setting. For the purpose of this chapter, barriers are defined as anything that impedes understanding and communication between two or more parties. That may mean, for example: practicalities in the availability of communication support because systems are not responsive to the needs of Deaf people, unilateral or bilateral lack of understanding of the cultural values and perspectives of the other party, or challenges whilst actually communicating because of the complexity of information or unfamiliar terminology. The definition of barriers used here therefore includes not only barriers that prevent people coming to services in the first place, but barriers that hinder clear communication once Deaf people are within the health service. The chapter examines the work that has been done in this area using a process and outcomes framework similar to that used in genetic counselling research. Use of such a framework helps to illustrate what is understood about the implications of the barriers (outcomes) and which barriers have been identified. However, not enough is yet understood about how different barriers operate, interact and contribute to each other, nor the mechanisms which create them: the process. If effective communication is at the root of effective genetic counselling, then it could be argued that, theoretically, all barriers can be understood, if not ameliorated, from the perspective of a failure of some aspect of communication.

In summary, Chapter 1's exploration into the different constructions of deafness aims to inform this chapter's summary of the interaction of Deaf people and health services (with a particular focus on genetic counselling services). Further, the chapter argues that communication is central to the relationship between Deaf people and health services. Chapter 3 then investigates terminology, language and understanding in greater depth.

2.2 Genetic counselling as an evolving speciality within health services

2.2.1 History of genetic counselling services within a changing society

The aims, content and process of the giving of health-related genetic information have changed dramatically and continuously since the start of the twentieth century (Harper and Pierce, 2010). That the scientific content has changed by degrees of magnitude is self-evident (ibid.) and increased genetics knowledge has inevitably influenced both the aims and process. However, there is also evidence that change in both aims and process has been significantly affected by the surrounding society and by the conscious choices of individuals working in the area. These conscious choices – specifically those of some key individuals – are based on personal beliefs (which, in themselves, would be affected by the context in which the individuals lived and worked) and conscious reflection on the lessons learned from earlier decades. As Galton said in 1909, with some prescience, given his legacy of an idea about which opinion has shifted so significantly (see below), 'social opinion operates powerfully without us being conscious of its weight' (Thom and Jennings, 1996 p214).

This statement means that any documents about genetic counselling have to be viewed as an historical reflection of their time: that there exists a cohort effect that means the social context must be also understood as part of the influence on what genetic counselling services have become today.

The first geneticists to identify some of the building blocks that explained inheritance, such as the identification of chromosomes and the use of Mendel's laws to explain recessive inheritance of genetic conditions, restricted themselves to objective observation of these phenomena. However, Francis Galton's choice to study physical traits including intelligence, criminality and poverty led him to conclude that genetics could be used to improve the health of the nation and he was the first to coin the term 'eugenics' to explain this idea (Walker, 2001). It is interesting to reflect that the use of a word which translates directly from the Greek as 'good genes' could change over time to encapsulate a concept so prejudiced and discredited: if ever there is an example of a word that does not only have an objective meaning, but has layers of additionally socially-constructed meaning, this is it.

Thom and Jennings, in their excellent history of genetic counselling, acknowledge that the term eugenics has different meanings (1996 p224). One distinction is between positive and negative eugenics, defined as the encouragement of those with 'desirable' traits to reproduce (positive eugenics) and the discouragement or prevention of those with 'undesirable' traits from reproducing (negative eugenics). Although eugenics is now generally taken to refer to genetic traits, in the early 20th Century it was also used by some people to mean any theoretical perspective or practical approach that supported survival of desirable traits – including environment improvements such as housing or education. Nevertheless, this perspective still encapsulates the idea that people purporting to be eugenicists are making judgements on which 'desirable traits' should be encouraged.

From Galton's initial ideas, the eugenic movement spread to over thirty other countries where it was highly influential in shaping how genetic information was given: generally as advice against reproduction for anyone judged 'less fit' (Thom and Jennings, 1996). The direct influence of the eugenic movement caused its most horrific abuses during the Nazi era, with widespread forced sterilisation on the basis of race and disability as well as the later mass slaughter in extermination camps (Muller-Hill, 1994). But the Nazis were not the only group to abuse genetic principles, with several other nations, most notably the US and Scandinavia, enacting legislation to enforce state-supported sterilisation for groups of disabled people (ibid.).

Thom and Jennings (1996) argue that the history of eugenics can be understood more clearly by considering the influencing factors as falling into three areas: the perspectives towards people with disabilities i.e. the current prevailing view towards particular conditions, the science about the causes of conditions and the ethics of the proposed solutions. The first of these has a significant overlap with Chapter 1 of this thesis and particularly with the historical evolution of views towards disability (Oliver, 1990). As far as the scientific basis was concerned, the early eugenicists were basing their theories on little empirical knowledge, but Gillott (2001) argues that one of the major reasons why this did not significantly hinder the movement was that it was predominantly driven by political fears of social decline rather than directly by scientific advances. It has been argued that the prevailing social movement of modernism in the late 19th and early 20th centuries was instrumental in the development of ideas, such as eugenics, that supported the good of society as a whole in the national interest rather than the rights of the individual (Turda, 2010). Modernism also valued scientific method highly and, in this context, quasi-scientific approaches such as those used by eugenicists found fertile ground in society. In this context, options such as sterilisation, despite taking away individual choice, were considered ethically acceptable (Muller-Hill, 1994).

After the Second World War and condemnation of Nazi abuses in the name of eugenics, public opinion began to move against the infringement of individual rights in pursuit of population-based goals. Nevertheless, sterilisation of people with learning disabilities continued as late as the 1970s in the US and Scandinavia (Walker, 2001). Alongside these explicit negative eugenic actions, the philosophy of eugenics continued to be a driving force for some of the individuals pivotal in developing early medical genetics clinics. A medical model evolved in the 1950s which aimed to provide information about empiric risks, although little diagnostic testing was available. However, the assumption was still that the main aim of this intervention was to prevent affected individuals being born. One of the most prominent proponents of this perspective was Professor Cedric Carter at the Institute of Child Health, London – his personal positive eugenic philosophy was that intelligent people had an obligation to have children (Clarke, 1984). Alongside this philosophy, he strongly espoused the view that the main aim of his service was the reduction in the number of children born with disabilities and he continued to hold this view throughout his working life (Carter, 1974; Thom and Jennings, 1996).

According to Thom and Jennings, the single most important individual working for a change of focus from eugenic ideas to human genetics was Professor Lionel Penrose at the Galton Laboratory. His work was mostly on the causes of intellectual disability and he moved the arguments subtly and incrementally to a more neutral position over the course of his career. He made it clear that he thought medical geneticists could not know everything and therefore, intellectually, should not make value judgements. For example, he stated ‘Subcultural mentality [sic] must inevitably result from normal genetical variation and the genes carried by the fertile scholastically retarded may be just as valuable to the human race, in the long run, as those carried by people of high intellectual capacity.’ (Thom and Jennings, 1996 p228). He was also clear that there should be a separation between the science and how it is used: ‘human genetics is a science and eugenics is an ideology’ (ibid. p230).

2.2.2 Genetic counselling as a client-centred service

Nevertheless, there was still a significant gap to be bridged between this theoretical perspective and the shift in service emphasis that then occurred through the 1970s and 1980s. This shift was towards a position of genetic counselling provision that was non-judgemental and worked towards maximising the autonomy of the client by giving information in a non-directive manner. This emphasis on individual autonomy was the direction in which medicine had been moving since the Second World War as a reaction to the historic paternalism of doctors and was one of the four core principles of biomedical ethics set out in Beauchamp and Childress' seminal 1970s textbook (2008). Clinical genetics was following this trend within medicine, but had also been debating its ethical position for longer than most specialities because of the explicit reaction against the legacy of the eugenic movement. Client autonomy and non-directive counselling to facilitate autonomy continue to be the two central tenets of genetic counselling, particularly in relation to reproductive choices, and maximising autonomy is one of the professional standards to which genetic counsellors in the UK commit themselves (Association of Genetic Nurses and Counsellors, 2012; Genetic Counsellor Registration Board, 2012a). However, the extent to which these tenets can be achieved has continued to be an area of great debate and research within the profession and this argument is summarised and developed in section 2.4.

The definition of genetic counselling that is most commonly referred to was written in 1975 by a committee within the American Society of Human Genetics (Ad Hoc Committee on Genetic Counseling, 1975). Although, as discussed in the next section, the depth of understanding of the process and outcomes of genetic counselling has moved forward significantly, the 1975 definition is still valuable and largely salient to current practice (and very relevant to this thesis), in emphasising genetic counselling as 'a communication process'. Crucially, the focus of the definition is on the empowerment of the individual rather than any external or population based outcome measures.

2.3 The development of process and outcome research in genetic counselling

After the acceptance of these central principles of genetic counselling in the UK, Western Europe, North America and Australasia, later definitions began to add more detail about the process. These definitions had different emphases and balance between the diagnostic, risk assessment, educational and counselling aspects of the genetic counselling exchange (Kessler, 1997; Harper, 1993), but all had in common the components of technical knowledge, relaying the information in an understandable way to the client and conveying it in an empathetic and supportive manner: components that distinguish the role of a modern genetic counselling professional from either a genetic scientist or a generic counsellor. Kessler (ibid.), for example, explored these co-existing aspects by separating genetic counselling into two models: the teaching and the counselling model, but recognised that, in practice, aspects of both approaches are brought together in the clinic setting and the emphasis changed according to the specific situation.

These papers illustrate the close relationship that was already emerging between the clinical service and research that had developed from practice. The drivers for the research were partly to continue reflective practice in understanding the psychosocial content and implications of genetic counselling, but also because the 1990s was a time when there was pressure for all branches of medicine to prove their value and effectiveness through measurement of desired outcomes and so justify their existence. Chadwick (1993) and Clarke (1990; 1993; 1997a) both felt that pressure from Government to use health economics arguments and, through them, health service management would lead to the use of blunt measurement tools such as the effect on birth incidence of genetic disease. They agreed that this is an inappropriate measure, indicative of the earlier focus of genetic counselling. Chadwick suggested that patient satisfaction questionnaires are suitable tools to measure, amongst other factors, the extent to which patient autonomy in decision-making is achieved. Clarke agreed with the use of questionnaires to client and referrer and also put forward workload audit as a descriptor of what is done by clinical genetics professionals.

In a later book chapter, Clarke argued that ‘genetic counselling is a process centred on the clients and their need to understand the condition in their family…the ethos of genetic counselling, then, is for the clients to set the agenda…’ (Clarke, 1997a p169-170). He was not changing the pre-existing definition of genetic counselling, but was rather emphasizing further that the success of genetic counselling should not be measured on crude outcomes of birth incidence, but on measures that related directly to the autonomous and information-giving nature of the process, without making assumptions about the client’s own agenda or that they necessarily want to prevent a genetic condition occurring. The next question would be exactly how client satisfaction with the experience of genetic counselling could be measured – and with suitable depth and complexity to reflect the complexity of the genetic counselling intervention itself.

In response to this challenge, Berkenstadt and colleagues (1999) brought together outcome measures in a new concept known as perceived personal control (PPC) and developed from it a questionnaire evaluation tool that had three domains: cognitive, decisional and behavioural. They found that individuals who were given more definite information such as diagnosis or specific chance of recurrence had higher PPC scores, supporting the idea that knowledge, in itself, increases the sense of control over a situation.

Shortly after this paper, Biesecker and Peters (2001) reviewed the current evidence and gave clarity to the difference between outcome studies and process studies. They argued that understanding what is going on within the health interaction is equally important to measuring outcomes, and that, in fact, the two are symbiotic: what professionals and clients see as the desirable outcomes or what they expect the outcomes to be (which they may or may not agree with) will influence both how they approach the consultation and how successful they judge it to be. Biesecker and Peters posited a new definition which was not significantly different in its scope from earlier definitions, but the emphasis was on the dynamic and complex nature of the interaction rather than breaking it down into constituent parts. It did not specifically mention the diagnostic element of genetic counselling and as such, could be regarded as more of a working aim for a genetic counsellor focussing on the psychosocial elements of the exchange rather than a clinical geneticist:

Genetic counselling is a dynamic psychoeducational process centered on genetic information. Within a therapeutic relationship established between providers and clients, clients are helped to personalize technical and probabilistic genetic information, to promote self-determination and to enhance their ability to adapt over time. The goal is to facilitate clients' ability to use genetic information in a personally meaningful way that minimizes psychological distress and increases personal control. (Biesecker and Peters, 2001)

Subsequent papers began to review the current frameworks for measuring outcomes and explored their use following the sequencing of the human genome which, it was argued (Wang et al., 2004), would widen the focus of genetic counselling from reproductive issues and more towards health issues that affect individuals in their own adult life – multifactorial conditions such as cancer and Alzheimer disease. Kasparian et al. (2007) and Payne et al*.* (2007; 2008) continued this synthesis of the available outcome measures, the latter group with the explicit aim of investigating what clients value within the process (McAllister et al., 2008) and how this knowledge could be used to develop new or improved psychosocial outcome measures (McAllister et al., 2011a; McAllister et al., 2012; McAllister et al., 2011b). Recognising the commonalities between Berkenstadt et al.'s PPC measure and the concept of Empowerment that they had developed, McAllister et al. collaborated to test the validity and reliability of their measures (2012). The Genetic Counseling Outcome Scale (McAllister et al., 2011b) that they have developed is a 24 item questionnaire with five dimensions: decisional control, cognitive control, behavioural control, emotional regulation and hope. It has potential to be used to measure change as a result of a genetic counselling intervention or a research intervention similar to a genetic counselling consultation. The measures are not measuring satisfaction with the process directly, but rather measuring the impact of the genetic counselling on the client's well-being.

Although this research is of great value to the profession in terms of supplying robust outcome measures that are based on what clients say they value about the process, it does not answer the questions about what actually happens within a genetic counselling session and, further, how this affects the outcomes. Many studies have now attempted to look at the internal process of genetic counselling through both the investigation of separate components as well as observation of sessions in their entirety. Such a large body of research cannot be adequately covered here, but, for example, components such as understanding of genetic information (Gale et al., 2010) and understanding of risk figures and choices made by clients (Smith et al., 2002; Vos et al., 2011; Michie et al., 2005) have been studied not only in terms of simple client recall after counselling, but also in terms of how these components were presented and discussed within the session.

Hallowell and Richards (1997) carried out a helpful review of studies that had looked at recall and understanding of risk figures. They found a wide range in the proportion of individuals who recalled risks correctly and proposed factors that influenced recall including mode of transmission, reproductive intentions, time since the counselling session, previous experience of the familial condition, subjective perception of risk and the way in which risk figures were presented. However, they concluded that measuring only risk recall is of limited value as it does not recognise the dynamic and continually-adjusting nature of the process 'within a particular emotional and sociocultural context' (ibid., p31). Further, there is an important distinction between correct recall and actual understanding of the risk figure so that it can be used by the individual and they stress that there is difficulty in defining understanding. This distinction (between knowledge and understanding) is one that is explored in Chapter 3.

Several reasons could be proposed as to why research has focused on some aspects of the genetic counselling process to a greater extent: one is that risk assessment or information-giving is a more active aspect and therefore more measurable compared with the subtleties of staying with a client in a supportive relationship whilst they worked on a decision or adjustment. The latter aspect is far harder to make visible and might be judged as apparent inactivity if only a blunt measure was used, but is anything but in terms of the engagement and work needed by both the client and the professional. A second reason has parallels with quantitative versus qualitative research generally – that quantitative research which measures, categorises and sorts out the complexity appears cleaner than 'thick' qualitative data and is attractive for that reason. These areas of research are valuable in having explored specific pieces of the genetic counselling jigsaw, but there is also a need for further qualitative observational research into the complex interaction of all components within a genetic counselling session.

To summarise so far, the previous section has highlighted how the changing social context in which genetics knowledge was developing influenced a move from the eugenic focus on population outcomes to the modern genetic service focus on autonomy and individual outcomes. Further, changes in the health services of the UK and other countries have contributed to the development of robust outcome measures and, in order to develop these measures, more process research has been developed. Given that this thesis proposes that communication is core to achieving a successful outcome, the next section discusses one of the central tenets of genetic counselling – non-directiveness – as an illustration of the complexity of a deceptively simple idea and the centrality of communication in trying to achieve it.

2.4 Non-directiveness

Section 2.1 explains how the historical influences on the development of genetic counselling was at least partly responsible for non-directiveness being considered an essential part of modern genetic counselling. However, Clarke (1991) was largely instrumental in initiating the debate that continued through the 1990s as to whether totally non-directive genetic counselling could ever be achieved. In his later expanded and thoughtful chapter on whether the idea of non-directiveness was now too restrictive (Clarke, 1997b), he pointed out that analysing only what happens in an actual consultation is too narrow to answer the question as to whether non-directiveness can be achieved. This is not to say that the principle of maximising client autonomy through giving information as neutrally as possible and without coercion is any less important. However, in addition to this challenge within a genetic counsellor's individual practice, there is a wider argument that the very availability of genetic tests for an inherited condition and making a client aware of them may give an implicit recommendation of their use, regardless of the professional’s own views and the way in which the information is given. Clarke suggests that there are personal benefits for genetic counsellors in adhering to a non-directive stance: that it allows them to feel they are unbiased and not supporting an option with which they may not personally agree. In other words, if one considers the wider social context, some options (reproductive, testing etc) may be challenging to the professional’s own values as well as to the client’s values. It can be argued that non-directiveness is part of a distinct genetic counselling culture and is an ethical stance that works for its members (genetic counselling professionals) in helping them to feel comfortable within that culture as much as it does for the populations they serve. Other professional groups, such as psychotherapists and counsellors, also provide non-directive counselling and support with the aim of empowering individual choice. However, genetic counselling combines this philosophy of individual choice with the option of medical interventions such as carrier or prenatal genetic testing which may be ethically contentious. It is the non-directive approach in this context that distinguishes genetic counselling from both other types of counselling and other branches of medicine. In the latter, professional recommendation of a particular course of action has historically tended to take precedence over complete client autonomy.

Having said this, increasing genetics knowledge has changed some genetic counselling situations by the addition of an intervention with a clear medical health benefit, such as cancer screening by mammography or colonoscopy. In these situations, there is a growing consensus that qualified professional recommendation of this intervention is appropriate. As Clarke pointed out (1997b), the ethical imperative of beneficence could take precedence over autonomy in this situation. Although these situations are a challenge to the core position of non-directiveness, they have not fundamentally removed it as the starting position from which the service is offered. At the moment, they are seen as exceptions. However, this position could change if more treatments for genetic conditions are developed in the future and would raise further questions about the ability of genetic counsellors to maintain a neutral position.

As highlighted earlier, the professionals within modern genetic services support the view of the individual attending the service rather than making the judgement themselves. However, there are still legal and professional limits as to what choices are available and it can therefore be argued that the available options do not allow complete autonomy on the part of the client. For example, pre-implantation genetic diagnosis to determine gender of a foetus is not allowed in the UK (except in the case of a risk of a sex-linked condition such as Duchenne Muscular Dystrophy) (UK

Government, 2008). Another example are the professional guidelines that exist to guide and potentially limit testing of children for conditions which will not affect them until adulthood or in their own family planning, or which will not change any treatment or management decisions until later life (British Society for Human Genetics, 2010). Thus decisions are not made by the individual health professionals, but limitations are set by law or professional guidelines. Some researchers have argued that, not only is this not achieving non-directiveness, but is actually potentially eugenic. Gillott (2001), for example, makes the point that, whatever the intention of testing, whether as population-based screening or as individual parental choice, the outcome may be to avoid having a child with a specific condition. He points out that ‘whether or not these procedures are thought to be “eugenic” will depend on what that emotive term is taken to mean’ (ibid.). The definition of eugenic, he says, does not depend on whether there is a conscious intention. Again, this returns to the point at the start of the chapter that ‘eugenic’ is defined differently by different individuals and groups. He points out that some people opposed to prenatal testing and screening might agree that the aim and focus of genetic counselling have changed, but still object on the basis that the very availability of these options makes a value judgement on people who have the same conditions. He argues that individuals’ decisions about prenatal testing are not based only on these value judgements, but also on their own capacity for what they think they could manage within their family and how they envisage a child with that condition would cope. This may be true, but the riposte by those opposing that argument is that social factors will affect individuals’ knowledge and views about that condition and, if so, that it could still be seen as society influencing individual decisions, regardless of whether there is state-led involvement in policy.

Even a straightforward description of genetic service development can be analysed as to how society might influence its direction. For example, Donnai (2002) clarified genetic services not only as genetic counselling, but also as incorporating population screening programs. At the time, these were predominantly based on biochemical screening, but, as she predicted, there was potential for population genetic testing for predisposition genes and this technology is now starting to be realised through next generation sequencing techniques. The point to be made here is that the decisions on what testing is included in public health screening are made at the policy level and therefore by the majority view. It is difficult to incorporate the views of minority populations who may feel differently about the inclusion of screening for specific conditions within a testing programme – such as the Deaf population. Burke et al. (2001), in a discussion of the ethical and social implications of genetic tests, highlighted clinical validity (high penetrance of gene effect and high risk) and effective treatment as being two major axes on which to judge whether a gene test should be offered. However, they do not mention the severity attached by the clients themselves to the condition in their family as being a factor affecting which tests should be offered: it could be suggested that the absence of this factor illustrates that the majority view within the population is taken-for-granted in many instances. These arguments are by no means restricted to genetic counselling and testing: there are commonalities with screening and medical programmes in other areas, including those around deafness. For example, Young et al*.* (2006) argue that parental informed choice following the diagnosis of deafness in a child may not be fully informed for many reasons, but one reason is that the very decision to set up screening for deafness and the choices that follow signals an implicit recommendation by the professionals involved. The guidelines now used by the UK National Screening Committee (2012) to evaluate whether a screening programme should be set up cover four areas: the condition, the test, the treatment and the screening programme. However, within these areas, there are significant points which could be interpreted very differently by different groups. For example 'there should be an effective treatment or intervention for patients identified through early detection, with evidence of early treatment leading to better outcomes than late treatment' (ibid.) are open to very different interpretations of effective treatments or interventions in the case of childhood deafness.

The range in severity of the genetic conditions that can be tested for has increased in recent years, such that we have now moved beyond the situation even ten years ago when the focus was still primarily on the identification of loci for unequivocal 'diseases'. For example, despite an individual's own views about whether or not to have testing for Huntington disease, there is wide agreement in the general population that the availability of genetic testing both as a predictive test for the individual and as a prenatal test is not only acceptable, but a good thing. However, next generation sequencing techniques such as massively parallel sequencing (Bell et al., 2011), microarray testing and exome sequencing (Biesecker, 2010) raise questions about what should be included in such a test, given the impossibility of a client having knowledge or experience of all the conditions which could be included – deafness, in Bell's paper, being one of them. There is, of course, a distinction between carrier genetic testing and prenatal genetic testing, but the question of which conditions to offer testing for is still a valid one. These ethical questions are now a major focus of attention (Goldstein, 2011) as next generation sequencing techniques are implemented in NHS service laboratories and research laboratories.

Deafness is by no means the first potentially inherited condition where the ethics of prenatal testing availability, allowing the choice of ending an affected pregnancy, have been debated (Shakespeare, 1998; Parens and Asch, 2003; Shakespeare, 1996). Shakespeare clarifies his argument using the framework of the social model of disability which distinguishes between impairment (biological) and disability (social), though his more recent work (Shakespeare, 2006) argues that this model should be re-examined from a critical realist stance that acknowledges that the distinction between impairment and disability is less clear-cut. He points out that the impairment ‘equated with disease, illness and poor quality of life’ caused by different genetic conditions is hugely variable and that, for some conditions, such as achondroplasia (a common type of short-limbed dwarfism), the majority of the problems faced by an affected individual are disabling social factors rather than the impairment of the condition itself (Shakespeare, 1998). These issues are largely the subject of Chapter 1, but are reiterated here in the context of the differing perspectives that people with a particular condition may have from the general population.

In the recent past, the difference between the perspective of many in the Deaf community and the hearing majority was seen most explicitly in the 2007/8 debate over the Human Fertilization and Embryology Act (UK Government, 2008) which contains a clause prohibiting the use of pre-implantation genetic diagnosis or donor gametes for the purpose of actively increasing the chances having a child with a particular condition. Regardless of the fact that few people ask to use PGD to avoid deafness in a child and requests to use PGD to select for deafness are almost unknown, the introduction of the bill to the Lords specifically referred to deafness in the context of the clause, meaning that a value judgement was potentially being set in law between a deaf person and a hearing person. This judgement was vigorously opposed by the Stop Eugenics campaign and others (Emery et al., 2008; Blankmeyer-Burke et al., 2008; Stop Eugenics, 2008). The Government would have argued that their position was not eugenic in its intention i.e. the intention in not allowing PGD to select for deafness was not to decrease the incidence of deafness. Rather, it could be suggested that the decision was taken from a position of the hearing majority seeing it as ethically unacceptable to use this technique to select for what is seen by the majority as a disability. Again, this comes back to the definition of eugenics: whether there has to be an active intention or whether it is still eugenics if it is passive or unconscious. Eugenics continues to be a highly emotive word and genetic counsellors are likely to be horrified by the idea that they could be involved in a eugenic activity, but it is important that professionals continue to question the ethical basis on which they practice.

What these examples seek to illustrate is that genetic counsellors see the service as one that prioritises neutrality and non-directiveness, but it may still be viewed differently by clients who suspect another agenda and expect negative attitudes towards the condition in their family. Thinking specifically about deafness, Deaf potential clients may experience negative attitudes, even if unintentional – genetic counsellors, despite their best efforts to understand the client's perspective, may not have an awareness of Deaf cultural norms and may find it difficult to appreciate the Deaf experience of deafness as simply a different way of being rather than a disabling condition. If the counsellor believes they are understanding and tailoring their communication to the position of their client and yet are unwittingly ignorant of cultural differences and values, this lack of knowledge may contribute to not achieving a non-directive consultation.

Research on the attitudes of Deaf people towards genetics is valuable in providing genetic counsellors with advance knowledge of the perspective a client may bring, though sometimes additional understanding of Deaf culture is needed to contextualise the results. For example, Middleton et al. (1998; 2001) found that a proportion of Deaf people would prefer to have a deaf child and a very small number would consider pre-natal testing with the intention of proceeding only with a confirmed deaf pregnancy. That information can be better understood by hearing people if they also understand the wider context: that a deaf adult who had a deaf child would potentially have fewer communication barriers and a shared perspective within the family (Bauman, 2005). Seen within this context, it can be understood that Deaf people are often pragmatic about the chances of having deaf or hearing children and that a common perspective would be 'as long as the child is healthy' which for a Deaf person would mean either a deaf or a hearing child. In addition, there are studies in other areas of genetics, particularly cystic fibrosis carrier and prenatal testing (Decruyenaere et al., 1998; Denayer et al., 1992), that suggest that fewer people actually have a test than say they would theoretically. It is important to consider that these papers were also asking about a theoretical test so the position could be similar with prenatal testing for deafness.

To summarise this section, we have explored non-directiveness in genetic counselling as an example of a deceptively simple concept which is multifaceted and probably unattainable. The previous section had explored societal influences on its positioning as a central tenet of genetic counselling. This section has presented the several major challenges to non-directiveness made over the last two decades. These include the societal context in which genetic counselling is provided, which influences the development and availability of particular genetic tests and procedures. Potential and actual clients attending genetic counselling may agree with the acceptability of options available, but alternatively they may not, and may have a different conception of genetic counselling as a result. This suggests that striving to understand the client's personal perspective is equally important as it will affect how they receive information and whether they receive it neutrally, even if the counsellor gets close to delivering information and support in a neutral and non-directive manner. In addition, there is increasing potential for intervention such as cancer screening which can challenge the suitability of a non-directive position.

Given these challenges, it is suggested that genetic counsellors are aiming for something subtly different from non-directiveness. Rather, their approach is non-partisan. As in the original understanding of non-directiveness proposed by genetics professionals such as Clarke (1991), the central aims remain: to inform clients of all the options and support the decision that the client judges most appropriate for themselves. In addition, the approach is explicit about the medical evidence which supports recommendation of a particular option and recognises the variation in the individual and cultural perspectives that a client may bring. Balancing all these facets within a consultation is a dynamic process requiring the genetic counsellor to be highly sensitive, self-aware (including awareness of their own views towards the condition under discussion (Enns et al., 2010)), and responsive to what the client is saying (and not saying). Communication is central and it is language as both a tool and a barrier to conveying meaning that is explored in Chapter 3.

Now that these perspectives towards genetic counselling by providers and (potential) clients have been explored, the final sections of this chapter move to consider what is known about other barriers that exist for Deaf people in accessing health services in general and genetic counselling services in particular.

2.5 d/Deaf people's interaction with health services and genetic counselling

2.5.1 General access to health services

The excellent review by Harmer (1999) gives a thorough introduction to the intersection between deaf people and health care services. She makes the valuable point that it is not only deafness in itself that affects access to services, but that deafness affects many other things such as access to background knowledge, communication and support within the family, confidence and self-identity, educational opportunity, ability to access healthy living information and employment. For example, factors within the home, such as people from minority ethnic groups having several languages in the home, may well affect the communication background prior to their first interaction with health services. All of these other factors could affect an individual's health even before the direct issue of access to health services is considered. Communication is also key to all of the issues above.

Harmer adds that D/deaf clients may have experienced health care as a child (and perhaps also as an adult) only as a passive recipient because of lack of explanation on the part of the professionals. As adults, therefore, they may be unprepared for involvement as an active participant supported to make autonomous decisions. In the context of this thesis, it is important to consider, therefore, that Deaf people's antipathy towards genetics may not be isolated, but part of a general suspicion towards medical and health services because of their experience of deafness always being treated as a pathology, with the predominant focus being on their ears and deafness, rather than the whole person.

An increasing number of outcome and observational studies have looked at D/deaf people's satisfaction and reported problems with access to general health services (Pereira and Fortes, 2010; Cardoso et al., 2006; Iezzoni et al., 2004; Steinberg et al., 2006; Steinberg et al., 1998; Steinberg et al., 2002; Barnett, 1999; Signhealth, 2008; Signhealth, 2009a; Signhealth, 2009b; Royal National Institute for the Deaf, 2004; Signhealth, 2014) and set out basic guidelines on communication in health settings (McAleer, 2006). There are common themes across these studies such as lack of awareness by health professionals about practicalities in communicating effectively. These practicalities include enquiring about preferred language and booking a BSL interpreter or other communication support where required, wrongly assuming that written English will always be an appropriate means of communicating with a D/deaf person, not taking the necessary additional time in a consultation that may help to overcome communication barriers, communicating with hearing family members about the D/deaf person rather than directly with the person themselves and not making adjustments that could help build up a rapport with a D/deaf person such as moving away from a desk/computer input and making greater use of body language and eye contact.

There is evidence that D/deaf people have a greater number of visits to their GP compared to hearing peers, that a significant number have taken the wrong amount of medication due to lack of communication with a health professional and that a significant number have missed medical appointments due to oversights such as patient names being called rather than a visual alert (Royal National Institute for the

Deaf, 2004; Signhealth, 2008). Despite these well-documented barriers and the reasonable assumption that they will have a negative impact on d/Deaf people's health as a result, there has been only limited research documenting D/deaf people's health (Ridgeway, 1998; Connolly et al., 2006; Fellinger et al., 2005). A much anticipated report in the UK by the Signhealth charity (2014) has recently been published to add to the evidence. This report has shown concerning results. For example, the 300 Deaf participants who underwent a full health check were twice as likely as the general population to have raised blood pressure and four times as likely to have pre-diabetic levels of blood sugar. The results from the 533 Deaf people who responded to the online survey and the 47 in-depth BSL interviews backed up previous evidence that many had great difficulty in making appointments with GP surgeries and hospitals and then in communicating effectively with their healthcare provider.

Although there are a number of other recent studies documenting these practical barriers and investigating outcome health measures for D/deaf people (Barnett et al., 2011; Fellinger et al., 2012), there is, as yet, little process research about the content of health consultations with D/deaf people. The importance of communication is implicit in many studies, but there are parallels with the genetic counselling research discussed earlier, in that much research into communication has so far looked at simpler measures such as information recall (Baldwin et al., 2012) or background health knowledge (Zazove et al., 2009; Pollard and Barnett, 2009). There is, as yet, little qualitative research about the complexity of a health care consultation with Deaf people and the next section discusses this dearth of research in the specific context of access to genetic counselling services.

2.5.2 Deaf people's access to genetic counselling

There are two facts in relation to Deaf people's access to genetic counselling that are most frequently cited in the literature and, as such, it is possible that they have influenced the type of research carried out to date. The first is that the number of Deaf people seen in the genetic counselling clinic is a very small proportion of all those for whom it may be applicable. Figures from the Royal National Institution for the Deaf (rebranded in 2012 as Action on Hearing Loss) suggest that, from the North-West population of 4.6 million people, there are likely to be as many as 5000 BSL users (Action on Hearing Loss (previously Royal National Institute for the Deaf), 2014). In the Genetic Medicine Department in Manchester, approximately 150 families a year are seen for whom the primary reason for referral was deafness. However, only a small proportion of these are adults likely to identify as part of the Deaf community, as a BSL interpreter is booked for a genetic counselling consultation in Manchester on average

10 times a year. This is a small proportion of all those for whom genetic counselling may give useful information, if a discussion of deafness, its causes and related health issues is included in the indications for referral to the service. Reflecting similar findings, Arnos and colleagues reported in the US that, of 175 families contacted with likely hereditary deafness, 58% had a clear family history of deafness or a clear syndromic cause, but only 16% had been referred for genetic counselling (Arnos et al., 1991). These low referral rates could be due to individuals not wishing to be referred, health professionals not offering a referral, or Deaf people not being aware of the service and its potential benefits and therefore not seeking a referral. The group in Arnos et al.'s study were parents of deaf children, so they comment that the proportions are likely to be smaller for deaf adults where there may be additional cultural and linguistic differences.

The second factor is the views of Deaf people towards genetic counselling services. It has been documented for many years that some Deaf people have views ranging from caution towards genetic technology to direct equation of genetic counselling services with a negative eugenic approach and an agenda on the part of genetic counselling professionals to eradicate deafness (Middleton et al., 1998; Burton et al., 2006; Stern et al., 2002; Taneja et al., 2004; Withrow et al., 2009; Middleton et al., 2001; Brunger et al., 2000; Dennis, 2004). Whilst it is neither assumed that suspicion towards genetic counselling services is universal amongst Deaf people, nor that it is the most significant factor in the low numbers of Deaf people who attend for genetic counselling, cultural values and beliefs are nevertheless likely to be important in the decisions Deaf people make about whether they wish to access genetic counselling services (Boudreault et al., 2010; Middleton et al., 2010a). Given the debate around the clause in the Human Fertilisation and Embryology Act 2008 that was discussed in section 2.4, and considering the significant increase in the number of deaf children who now receive cochlear implants at a young age over the last twenty years, it is not surprising that a significant number of Deaf people conclude that genetic services and medical services as a whole have at least an unconscious, if not conscious, agenda to eradicate deafness.

It could be considered, therefore, whether improving accessible information about genetic counselling e.g. in BSL, strongly visual and/or plain English formats, and therefore highlighting the genetics professionals' perspective towards their service, might increase the number of Deaf people who choose to access the service. Whilst this may be the case, it is equally important to improve the communication within a consultation itself, both practically around information transfer and to enable mutual understanding by the client and the counsellor of the other's agenda and perspective. This brings us full circle to the other side of the debate in section 2.4: that of non-directiveness and the genetic counsellor's need both to understand the wider implications of what they are offering and how this may be perceived by the client, based on the attitudes and beliefs they bring to the consultation.

Section 2.4 explored the research that has been done from the perspective of genetic counselling professionals and related researchers. However, there is now also a sizeable body of research from the disability studies perspective that is debating the relationship between disability and genetics. Whilst some of this is by Deaf researchers (Emery et al., 2010; Blankmeyer-Burke et al., 2008; Scully, 2008; Lane, 2002), there are many other researchers, disabled and non-disabled, commenting on the ethical relationship between deafness, disability and genetics (Chadwick and Levitt, 1998; Johnston, 2005; King, 1999; Madeo et al., 2011; Hayry, 2004) and the impact of this ethical relationship on the client/counsellor relationship that is formed (Evans et al., 2004).

It has already been argued that a mutual understanding of the positions client and counsellor bring to consultations cannot be achieved without communication that is in the correct register, that makes adequate provision for accurate interpretation, if necessary, and that therefore achieves effective transmission of information. The onus is on the professional to achieve this level of communication. There has been some work both to improve the information available about genetic counselling for deafness (National Deaf Children's Society, 2006; Belk, 2008) and to publish suggestions for practical adjustments to be made in the provision of genetic counselling for d/Deaf people (Arnos et al., 1991; Middleton, 2006; Belk, 2006; Middleton et al., 2010b).

There is also continuing engagement between members of the Deaf community and genetics professionals through joint research and advisory group membership (Leigh and Marschark, 2005; Blankmeyer-Burke et al., 2008). One significant co-existence of genetics and deafness alongside each other has been the Genetic Services Centre based in Gallaudet University in Washington D.C. Back in 1991, I. King Jordan, the first Deaf president of Gallaudet University, made a significant statement of purpose in saying 'I believe that the genetic counseling community has taken the necessary first step by changing the locus of responsibility from societal to individual, in effect defusing the old eugenic argument that the individual should sacrifice his or her reproductive rights to the "greater good of strengthening the race."' (Jordan, 1991).

However, research into aspects of communication between hearing genetic counselling or other health professionals and D/deaf people is very limited. One of the few studies looked at provision of genetic counselling related to Connexin 26 genetic testing and showed a statistically significant increase in Deaf participants' knowledge of related genetic concepts before and after counselling (Baldwin et al., 2012). Within other studies about Deaf people and health services, there is sometimes (as in the broader genetic counselling literature) reference to specific aspects of language, when the main focus of the paper is not on language. One such example is in Andrade Pereira and de Carvalho Fortes' paper (2010) from Brazil about Deaf people's access to health consultations when they give the example of the word 'allergy': the Portuguese word is similar to the word for happy and this led to misunderstanding for a Deaf person when pen and paper was used to communicate and he confused the two words.

Research on communication and language within genetic counselling, related areas of science and BSL has predominantly been around specific terminology and translation (Scottish Sensory Centre, 2014; Craigie High School Dundee, 2008; Wolverhampton University, 2005c; Genetics and genetic counselling translation team, 2005; Belk, 2006). There are parallels here with work that has been done on translating genetic terminology accurately between spoken languages such as English to Urdu (Shaw and Ahmed, 2004). There are also parallels with other subject areas with unusual terminology or a need for historical accuracy where systematic study has produced BSL glossaries and translation (Deaf Professionals in Mental Health, 1997; British Sign Language Bible Translation Project Team, 2014). This area of research is mentioned here simply to highlight its existence, but is a starting point for the detailed exploration of language in Chapter 3.

Unlike the evolution of research about outcomes and, to some extent, about process discussed earlier, the evolution of research about communication within genetic counselling does not have the same arc of development that can be traced through the last half century. Rather, there are specific pieces of research that have taken different approaches to investigating language: some have focussed on the actual words and specific terminology, whereas others have looked at how the language used by genetic counsellors within sessions affects client satisfaction, and yet others have looked at the sociolinguistic dimension to a genetic counselling consultation. In these latter complex studies of interaction, there is overlap with studies that have looked at the process of genetic counselling by observing complete sessions. When the cohort is viewed as a whole, it is helpful to consider a distinction (though generally not one that is made by individual authors) between communication studies, which highlight the language first and investigate its impact on an aspect or aspects of the outcome, and process studies, where the primary focus is on the intention or content of the session and the language is a means to that end. The two areas are closely interlinked and the main difference is a matter of emphasis in the stated aim and main outcome measures. Some of these existing studies are referenced in Chapter 3 as part of the exploration of language, but the distinction is proposed here as a framework for identifying the gaps in knowledge and establishing the niche for and necessity of future research.

2.6 Conclusion

This chapter has explored how external social factors have interacted with growing genetics knowledge to shape the current service situated within health services. The service has developed client-centred outcomes and an increasing understanding of process, signalling professionals' commitment to maximising client autonomy. Published reflections on the concept of non-directiveness have been used to show that it is not only how the individual genetic counsellor delivers their contribution to a consultation that is important: external factors including the social context in which the service is delivered and the perspectives that clients bring to the consultation affect how they interact with the information and options with which they are presented. The chapter has asserted that exploring components of the communication between client and counsellor will contribute to understanding the quality of the relationship and the mutual understanding between client and counsellor.

The final sections summarised the research into Deaf people's interactions with health services and specifically genetic counselling services and argued again that communication challenges and differences in cultural perspective are at the root of lack of understanding between health professionals, the majority of whom are hearing, and Deaf clients.

Modern genetic counsellors are in an apparently contradictory position. They are involved in a field of medicine that has the technology to detect the causes of deafness, potentially prevent some people being born who would be deaf and, in the future, potentially be channels for genetic research findings that could 'cure' deafness e.g. stem cell treatments or gene therapy. At the same time, they are committed to providing a service which is centred on their clients' wishes and choices. Is it contradictory or is it not surprising that, given the long history of ethical challenges faced within medical genetics, the genetic counselling service has evolved to fully appreciate those ethical challenges and face them head on? However, the internal process of the genetic counselling consultation is still not fully understood and therefore neither are the influences brought to the consultation by both client and counsellor. Language and communication influence and are influenced by individual and societal use and therefore the language used in the topic of genetics and inheritance needs to be further understood.

Chapter 3: How concepts are communicated in science, genetics and health: what's in a word – and what's not in it?

> *'When* I *use a word,' Humpty Dumpty said in rather a scornful tone, 'it means just what I choose it to mean – neither more nor less.'*
> *'The question is,' said Alice, 'whether you* can *make words mean so many different things.'*
> *'The question is,' said Humpty Dumpty, 'which is to be master – that's all.'*
> *Alice was too much puzzled to say anything, so after a minute Humpty Dumpty began again. 'They've a temper, some of them – particularly verbs, they're the proudest – adjectives you can do anything with, but not verbs – however,* I *can manage the whole lot of them! Impenetrability! That's what I say!'*
> *'Would you tell me, please,' said Alice 'what that means?'*
> *'Now you talk like a reasonable child,' said Humpty Dumpty, looking very much pleased. 'I meant by "impenetrability" that we've had enough of that subject, and it would be just as well if you'd mention what you mean to do next, as I suppose you don't mean to stop here all the rest of your life.'*
> *'That's a great deal to make one word mean,' Alice said in a thoughtful tone.*
> (Carroll, 1871 Chapter 6)

3.1 Introduction

Chapters 1 and 2 made clear that language plays a major part in shaping societies and cultures. They also showed that language is the tool, but communication within and across cultures is the application of these tools and is key to understanding other cultures and facilitating debate. Communication is fundamental to improving access to knowledge and, more importantly, engagement with this knowledge so that it can be personalised and used. To understand communication better, it is necessary to explore how meaning is conveyed through language.

This chapter extends the argument further by addressing the relationship between terminology and access to information and knowledge through consideration of context, comprehension and translation. Is it enough merely to translate terminology into the linguistic preference of clients in order to maximise engagement and promote understanding? Or is there something more fundamental about creating conditions for the promotion of knowledge and understanding that extend beyond the translation and/or explanation of terminology? Knowledge and understanding of concepts as well as the communication of them through language are some of the most complex and yet taken-for-granted phenomena that an individual experiences: taken-for-granted precisely because this experience is ubiquitous and familiar.

The chapter considers previous work on terminology and the promotion of knowledge, engagement and understanding within science, genetics and health studies – engaging with some studies concerning spoken/written language(s), before focussing specifically on signed languages, including BSL. This work is considered with reference to some of the main epistemological positions articulated within current language studies. The term ‘language studies’ is used here to refer collectively to the differing, but overlapping perspectives of disciplines including linguistics, psycholinguistics and sociolinguistics, and touching on philosophical perspectives towards language. The term ‘language’ is used here to encompass all components of human communication, from the linguistic levels such as morphology, syntax, lexicology and semantics (Crystal, 2010b) through to extralinguistic pragmatic factors: metaphor, emotional and cultural meaning (ibid.). Through this exploration, the chapter locates the specific concern of this study within the wider debate about the inter-relationships of language, communication and knowledge transfer. In particular, it will draw attention to the similarities and differences between challenges faced in the promotion of knowledge and clinical engagement in spoken/written language contexts and those encountered in visual/signed language contexts. In this respect, the significance of cultures as well as modalities will be explored.

The literature search strategy followed a similar process to that described in Chapter 2. In addition, combinations of search terms were used for searching content of both titles and abstracts. Some of these search terms were MeSH terms and others not, given that the literature was multi-disciplinary. They included language, terminology, translation, communication, deaf, British Sign Language, Sign Language(s), signed language(s). Most of these terms have multiple meanings within different subject areas and gave very large numbers of hits. It was therefore more successful to identify and focus on the key writers and combine author names with the other search terms. The iterative searching described in Chapter 2 was then used again to both review reference lists retrospectively and, prospectively, find the newer literature which was citing the papers already reviewed. The journals listed in Chapter 2 again formed the core publications to meet the scope of this chapter.

The appraisal of the literature followed the same approach to Chapter 2. For the non-empirical literature around language meaning and translation, sociolinguistics, philosophy of language and translation theory, key writers and arguments were selected. These included work by Baker, Crystal, Emmorey, Meier, Pinker, Slobin, Temple and Young. For the empirical research, the criteria for relevance were different for the literature about English and that about Sign Languages, or translation to/from Sign Languages. The research that had looked at language meaning in applied settings in English or other spoken/written languages was restricted to genetics, science and medicine. The research that looked at signed languages more broadly included the communication of concepts in varied subject areas, but in a real-life setting. The empirical literature focused on the most contemporary work and also included contemporary examples as they arose, even if there was little in the peer-reviewed literature. There is one area of related relevance which is recognised as having a substantial body of published literature, but which was judged to be outside the scope of this review: this is detailed translation theory and interpretation studies.

The crux of this chapter is to demonstrate that identifying the ‘right word’ for something in a particular language – be that the language of origin or in translation to another language – is complex. Terminology or jargon words may be artificially created, loaned or loan-translated (Turner et al., 1998 p162) from another language; may have evolved from a word used originally with an entirely different or closely related meaning; may be translated literally (as semantically close a word as possible to the original) or dynamically (a word which pragmatically reflects the original meaning with reference to cultural and sociolinguistic factors) (Baker, 2011). Fundamentally, terminology must also be made meaningful in the context of its use: on what level the user has engaged with the meaning of the words and what scaffolding is needed around it in order for the user and recipient to develop a shared understanding of a referent (Elbourne, 2011 p15). This scaffolding will vary between languages and cultures – and this includes differences in grammar, modality and in pragmatic factors such as cultural and sociolinguistic meaning(s) in context.

Thus, an examination of terminology not only concerns lexical equivalence between languages, but also engages with the varied resources that different languages might possess to express concepts. The grammatical and structural properties of languages, as well as their lexicons, are not identical; allowing some forms of expression in one that might not be easily produced in another. For example, German has a strong tradition of producing compound words for new ideas made out of previous common words in the German language (e.g. das Lebensmittelgeschäft identifies a grocery store, but translates literally as ‘the stuff of life shop’), whereas French, over the last 40 years in particular, has adopted many loan words from other languages, particularly

English (e.g. le weekend), rather than coining compound or new words in French. Slobin (1996) illustrates beautifully how aspects of a picture story are attended to to a greater or lesser extent when it is retold in different languages. English, for example, has greater capacity to convey trajectories of motion whereas Spanish allows the relative positions of the characters in the story to be more easily conveyed. For signed languages, in comparison with spoken languages, the key underlying difference in the resources available for development of terminology or producing appropriate translation is modality: signed languages are visual and spatial. In translation between any two languages, full use of the available resources is part of the complexity of producing a product that not only reflects the semantic content, but is culturally recognisable and acceptable.

When a novel, fast-changing and/or technical subject is under discussion, there is a tendency to focus on the terminology: the new concepts and the new words that mark them. For example, consider the rush of new terms, not in existence 20 years ago, that have entered languages around the world in response to the ubiquity of computers and the internet. These include verbs such as to Google or to Facebook someone. Some of these terms have transcended their original locus to become more widespread in common parlance, such as ‘running out of RAM’ or ‘to program’. Indeed, this thesis proposal developed from an earlier translation project precisely because the translation process highlighted the difficulty of deciding upon appropriate, culturally recognisable and acceptable BSL terminology in a fast-changing topic area like genetics, where new English terminology was constantly being coined. In seeking to understand how conceptual knowledge is communicated and engaged with in order to promote understanding (in this case between professional and client), the terminology cannot be considered in isolation. The context of the communication situation and the language biographies of those interacting are also of relevance. This chapter will argue that much research considering information in a specific subject or clinical setting has not explicitly acknowledged within their methods or results the potential impact of the context around the terminology presented to participants. This may be because much of the published work in this area is practical, service-based and outcome-focussed rather than aiming to dissect the language itself.

However, despite the entreaty to consider terminology within a context, there is also good reason to investigate terminology itself. Lexical items develop precisely in order to label one particular idea and the existence of these units of meaning therefore allows *productivity*. Productivity is one of the distinguishing and defining features of human language: the ability to create new meanings through novel combinations of words

(Hockett, 1960). These units of meaning are therefore also an obvious starting point when researching the relationship between the terminology used by individuals and their understanding of the concepts conveyed. Laugksch's review and discussion paper on scientific literacy (2000) provides one framework against which existing research into the understanding of terminology can be compared. He argues that the term 'scientific literacy' itself appears simple, yet can be defined and interpreted in varied ways (ibid.). One of the models he introduces is Shamos' concept of three incremental levels of literacy (Shamos, 1995 p87). This model starts with *cultural scientific literacy*, which describes the idea of basic familiarity with scientific terms and is likely to be the level of scientific literacy of the majority of educated adults. The next level is *functional scientific literacy*, which describes the ability not only to understand a scientific vocabulary, but to be able to use it, albeit in a non-technical way. The first level describes a passive knowledge of terminology, such as might be needed to read an article in the media, whereas the second is active and engaged and would allow an individual to summarise and relay the content to someone else. The third level is *true scientific literacy* which requires background knowledge of the subject in order to appreciate fully the implications of the information.

Laugksch (2000 p81) summarises the definitions of scientific literacy proposed by the varied models into three easily referenced levels, broadly comparable to Shamos' model (1995): 'function-in-society', 'competent' and 'learned'. Previous research into use and understanding of genetic and health-related terminology and information is not always explicit about what level of literacy is being investigated or identified, but the three levels highlight significant differences in how an individual can utilise the knowledge they have. Kiefer (1988) proposes three levels of knowledge which would appear to map usefully onto the aforementioned levels of scientific literacy, though not with exactly equivalent meaning: '*linguistic knowledge*, which, roughly speaking, concerns the core meaning of lexical items, *conceptual knowledge*, which has to do with the predictable modifications of the core meaning in various contexts [the semantic meaning], and *encyclopedic knowledge*, which comprises the rest, i.e. knowledge associated with a word but which is not immediately relevant to linguistic structure [pragmatic modification of the meaning].' (ibid.). Laugksch's focus is on how competently language is used, whereas Kiefer is looking at what the user is conceptualising. These two frameworks can be referenced later when considering how terminology is 'known' by participants in existing published studies.

In what follows, the first section looks at research investigating participants' recall and understanding of terminology. Initially, this focuses on studies that considered one language only, but then moves to studies that have investigated translations of terminology between languages and/or creating or collecting terminology in BSL. Collecting existing terminology raises the concept of language-in-use: defined as a functional lexicon that has been engaged with by members of that linguistic community and that is understood at least to the level of how to use it in context ('competent').

The second section considers studies that have grappled with language in a context-rich setting. These include pragmatic aspects of understanding such as the emotional engagement with particular knowledge; the emotional impact of specific terminology; the impact of language on specific outcomes in a clinical setting; the challenges of translating not only terminology, but specific measures of health that require both linguistic and cultural equivalence; the experiences of engagement with information in different formats and different clinical settings. Through the chapter, the opportunity is taken to highlight the differences between the oral/aural and manual/visual modalities, both with respect to the physical differences in transmission (see Chapter 1) and how modality impacts the grammatical, neurolinguistic and cultural differences and resources between languages.

3.2 Definitions

Two concepts, as they will be used in this chapter, must be introduced for clarity. *Lexical item* has a narrower, more specific meaning than *word* so is more useful for this detailed discussion. *Word* has broader, therefore vaguer, meaning based on its common use, it has alternative specific meanings within fields of linguistics such as grammar and its common use refers to just one ring-fenced syntactic item i.e. it would be a separate word if in a written language. A lexical item, in contrast, is defined as a single concept so can contain several words e.g. 'final examination', or one lexical item can be represented by different words e.g. (in English) walk, walks, walked or (in BSL) BOOK BIG or BOOK SLIM. Lexical item is therefore more useful within a semantic and pragmatic (Huang, 2007 p2) discussion.

Terminology is used throughout this thesis and has thus far been used with its lay meaning of 'vocabulary found in specific subject areas'. The common usage has sociolinguistic connotations of the subject area being highly technical i.e. synonymic with *jargon*. In this chapter it will be used in combination with the definition above to mean lexical items – i.e. single concepts – within a specific subject area. Its use is not limited to technical subjects, though the published research referred to in the chapter is predominantly health-related and, in the case of genetics particularly, investigating a specific, relatively technical, field of vocabulary.

3.3 Lack of status and visibility of BSL leads to limited opportunities for discussion of technical subjects in BSL: implications for development of terminology

Chapter 1 looked at the position of deafness and signed languages in society. Once the impact of a minority signed language being surrounded by the majority spoken language(s) is recognised, the influences on the language itself can be explored. The following example of a disastrous communication failure made public the lack of power and consultation routinely faced by Deaf people in regard to their language.

On 10.12.13, Nelson Mandela’s memorial ceremony was streamed live worldwide. One common reaction for Deaf people and signers was initial pleasure that a sign language interpreter was placed prominently beside the main speaker’s lectern. The initial reaction was followed by bemusement and anger as it quickly became obvious to Deaf people that the interpreter, Thamsanqa Jantjie, was not signing anything meaningful. Despite the Twittersphere and internet exploding with complaints from Deaf people, no changes were made during the ceremony (itself an indication of a lack of professionalism as good interpreters always interchange with colleagues rather than sign continuously for that length of time). The results of a promised full investigation from the South African Government are still awaited. It emerged that complaints had been made previously that this ‘interpreter’ could not sign, which belied his later claim that his meaningless signing on this occasion was due to a schizophrenic attack – a claim that was robustly addressed by Atkinson (2013), a Deaf clinical neuropsychologist. Swinbourne (2013) summarised the implications of the incident several days later:

> *For me, the controversy also revealed something a little disturbing about the disconnect between deaf and hearing people – simply in the fact that someone could sign nonsense for so long on stage, at an event broadcast worldwide, without anyone realising, let alone stopping him. I believe that Deaf people need to be part of booking signers, giving feedback and helping to evaluate them – so that this kind of thing – on a big or small level – doesn’t happen again. The illusion of access is worse than having no access at all, it’s offensive. It makes a mockery of the language and demeans Deaf people…If nothing else, there is now much more global awareness of the fact that the quality of sign language provision really matters. That wasn’t the case before.* (Swinbourne, 2013)

The lack of understanding of the nature of signed languages continued to be highlighted as the hearing/non-signing media repeatedly asked Deaf people and interpreters to ‘translate’ what Jantjie had been saying – an impossible task as he was not using any identifiable signs, nor any grammatically meaningful facial expressions and body language, both essential components of signed languages (Kyle and Woll, 1985a; Turner et al., 1998 p152). Since the incident, he has appeared in an Israeli advert for a video-sharing app (BBC News Africa, 2014) in which his contribution is based on making a joke of his inability to sign, despite his protestations of competency at the time.

Returning to the barriers to communication access encountered by individual Deaf people, some fall into this area i.e. when a hearing service provider lacks the ability to judge an interpreter’s competency. The distinction between the different types of communication barriers (system-linked, attitudinal and language-linked) has been made in Chapter 2. The example above highlights specifically a link between this issue of mediated communication (i.e. needing to rely on a third person who is in control of the access to information) with the lack of recognition and validation of the Deaf way of being that was explored in Chapter 1. The later sections of this chapter will reference this recognition that the dearth of understanding of Deaf experience and culture extends to signed languages as well. This lack of understanding means that service providers may not take the nature of the language, including the significance of a visual-spatial modality, into account when considering how to make information accessible.

Another aspect where the relative power between languages is important concerns the origins of new lexical items. A minority language is influenced by the majority language around it (Ann, 2001). New concepts are labelled first in the language in which they were first described. English (alongside several other world languages that are the most widely spoken and/or influential e.g. Spanish, Mandarin, German, French) is a highly dominant language within academia, production, education, medicine, science and other areas of technical and terminological innovation, both in the UK and around the world. This means that a technical term for a new concept in a minority language is most commonly translated from an existing lexical item in a majority language such as English, rather than developing separately in the minority language directly from the concept. As will be explored later, the influence of the first language can influence the translation choices which are made.

In addition, Deaf people themselves have historically had few opportunities to take on professional roles (Ladd, 2003) because of a lack of educational opportunities affecting their language and literacy and restricting their routes into employment (Moores, 2010). They therefore have not been in a position to influence development of topic-specific concepts, terminology and information, embedded and understood with true scientific literacy. Those Deaf people who have succeeded in academic and professional settings have often had to work predominantly within the written/spoken language (and culture) of their country because they are working mostly with hearing, non-signing colleagues (O'Brien and Emery, 2014 p33). This creates further bias towards those d/Deaf people who have succeeded in the oral education system that dominated from the infamous Milan conference of 1880 until recent decades (Moores, 2010; Turner et al., 1998 p145) and still influences the education system today. Ironically, those Deaf academics and professionals working as a minority within hearing-dominated fields, perhaps with an oral education and greater access to the hearing world, may not necessarily be seen as the natural leaders in their own community. As discussed in Chapter 1, the legitimacy conveyed by being a native sign language user is powerful when the community looks for its natural leaders (Ladd, 2003; Mindess, 2006). The linguistic innovation and involvement with new terminology of this small professional group may not have many opportunities for wider dissemination in the broader signing community.

As a result of this dominance of the English language and hearing professionals in most situations where there are discussions of technical subjects, there are many areas (genetics being just one of them) which are not discussed widely in BSL or other signed languages. In addition, the opportunities for social interaction in BSL are limited for most Deaf people by the reality of being isolated within the hearing population for the majority of home and work life. These limitations on discussion of such subjects with peers limits the opportunities for terminology and embedded discussion of concepts to evolve naturally and make maximum use of the resources of the language. It means that discussions about terminology often remain within practically-focussed projects which are seeking the best ways to represent concepts in BSL, but are slightly removed from naturally-occurring language. These are concepts already understood by the researchers and/or professionals involved (e.g. Deaf Professionals in Mental Health, 1997; Scottish Sensory Centre, 2014; Genetics and genetic counselling translation team, 2005), but not necessarily the wider Deaf community. Where signs are not in existence or known to conversational partners in a particular exchange, then local agreements have to be made about the signs that will label different concepts, if only as temporary placeholders: for example between interpreter and client (Jones, 2004 p30; Major et al., 2012) or teacher and pupils (Roald, 2002). This brings the discussion back to Deaf people's common experience: of mediated communication, either through an interpreter or by hearing people controlling the flow of information. These factors can disempower the individual and, insidiously, also the language (Temple and Young, 2004), particularly when BSL is not recognised as legally equal to other UK minority languages, such as Welsh (British Deaf Association, 2014b).

To summarise, there are several reasons why the opportunities to develop new terminology in fast-developing technical subjects may be limited in a minority language such as BSL:

- The language is not well understood by most English users and therefore service providers do not appreciate the importance of supporting the use of BSL and ensuring that the quality of interpreting and translating is of a high standard.
- English is highly dominant in technical subjects and, as the majority language, can significantly influence BSL development with much less influence in the other direction.
- Deaf people have historically faced barriers to joining professional and academic fields where they would influence BSL development in technical areas and there are limited opportunities for the individuals working in these areas to share new concepts with the wider community.
- The opportunities for discussion of such technical subjects in BSL is limited by the opportunities Deaf people have to communicate in BSL with peers, rather than the more usual experience of mediated communication. Consequently terms tend not to evolve naturally within conversational exchanges, but rather have to be more formally created.

There are reasons to be cautiously optimistic that some limitations may be easing. For example, a greater number of Deaf people are now working as academics (O'Brien and Emery, 2014), nurses, teachers and in other professions. BSL has been recognised as a UK minority language by the UK Government (Dodds, 2013; Smith, 2003; Department for Work and Pensions, 2004), albeit without legal protection to date (British Deaf Association, 2014b) apart from in Scotland (The Scottish Parliament, 2015). BSL courses are widely available (Signature, 2014) and initiatives such as 'Learn to Sign' week are raising its profile in schools and other settings. However, this does not fundamentally change the points above and their impact on limiting the development of specialist terminology in BSL. There is one factor now having a significant positive impact on the spread of signs, both nationally and internationally: the explosion of digital technologies and social media, allowing Deaf people to communicate easily over long distances. These technologies may not have been developed specifically with d/Deaf people in mind, but this group have been significant beneficiaries of the digital revolution (Belk et al., 2013; Young and Temple, 2014d). Not only has instant written communication (SMS, emails, instant messaging and social network sites) superseded textphones and faxes in many situations, but the speed of broadband connections means that BSL can be used instantly and directly for communication, either as posted clips or through video calling using software such as FaceTime, Skype and ooVoo.

3.4 The strategies used to overcome or bypass lexical gaps or translation challenges

Rather than, or alongside, exploration of specific components of communication such as terminology, some studies have looked more broadly at the strategies used to overcome or bypass lexical gaps or translation challenges. For example, Major et al. (2012) highlighted the lack of an extensive health lexicon in Auslan (Australian Sign Language, which comes from the same sign language family as BSL). Data from their focus groups of Deaf Auslan users found that written notes were frequently tried, but were often inadequate because of time, literacy levels and complexity of information; signs were shared between interpreters and Deaf clients when one party knew of an existing sign, fingerspelling (effectively a use of transliteration) was used to borrow words from English and act as placeholders (i.e. in the absence of another sign), as were signs invented just for that communicative exchange (*nonce signs* (Major et al., 2012)); depiction (pointing/placement in space), gesture and use of pictures were substituted for lack of vocabulary; explanation of the meaning of the missing term without use of a specific word was the most common strategy. Interestingly, this last conflates the separation between linguistic knowledge and encyclopaedic knowledge (Kiefer, 1988) and suggests that, if the aim of a conversational exchange is to transfer encyclopaedic or at least conceptual knowledge, then it may not be enough for the conversational partners to agree on the word or substituting placeholder (e.g. fingerspelt word) because this is only linguistic knowledge and does not consider the meaning. In their conclusion (Major et al., 2012), they stress the important point, discussed in Chapter 1 of this thesis, that ‘Often, deaf people’s preferences for the negotiation of English health terms depend on their level of bilingualism’. For example, if an individual already has a good conceptual understanding of an English term, then they may be quite happy for a placeholder such as a fingerspelt word to be used with no further elaboration of the meaning, whereas a term that an individual has never encountered before is likely to need contextualisation for there to be any chance of accurately conveying the meaning. The aims of particular communication studies may be placed along a continuum from outcomes, as in the paper above, to process, which considers the constituent parts of the communication – or could include aspects of both. As studies are discussed during this chapter, their positioning between process and outcome will be acknowledged.

3.5 The importance of the researchers' choice of consultation method with participants

Aside from a research study's focus on process and/or outcome, there is also the issue of how researchers have consulted with their participants. The epistemological orientation towards this study (see Chapter 4) drew from published work that has explored how the means of consultation affects the outcomes, positively or negatively (McLaughlin et al., 2004; Temple and Young, 2004). During the detailed exploration of language studies in the following sections, it must be borne in mind that how accurately the researcher(s) discerned the participants' experiences of language was likely to depend greatly on whether the researcher(s) had the cultural awareness and knowledge to have tailored their approach in the most appropriate way. Since the start of this research, there are an increasing number of studies which have emphasised this need to take Deaf cultural and communicative mores into account in planning their methodology to maximise their results' validity (Major et al., 2012; Rogers and Young, 2011; Pollard et al., 2009; Johnston and Napier, 2010; Rogers et al., 2014; Rogers, 2011; Rogers et al., 2013a; Rogers et al., 2013b; Young et al., 2016).

3.6 The different approaches to investigating knowledge, understanding and perception of existing terminology

This section begins with a position statement to emphasise that this literature review is not aiming to interrogate existing research into terminology from a particular and detailed linguistic perspective. Rather, it aims to describe the approaches that have been taken and identify the varied aspects of language that can be seen in previous research, including areas of linguistics, psychological impact, effects of modality and challenges of translation between languages. In many of the existing studies, the researchers have taken a service-based practical approach, without necessarily analysing or naming aspects of linguistics that could be discerned from their investigation process. The section considers research focused on BSL, but also in other signed and spoken languages. The one area that is not included in detail here, as being outside the scope of this study, is research related to the pragmatic live decisions made by interpreters.

The list below summarises possible approaches to investigation of terminology that could be and have been taken. It is proposed here so it can be used to structure the following chapter sections. It is not likely to be exhaustive, nor is it presented with any hierarchy in mind. Rather, this list aims to give a framework and stimulate thought about the degree of magnification one could use when focussing in on aspects of existing language and also to point out the different perspectives that could be taken. Each approach is valid in its own right, as will be explored in each section. If there is an issue in a number of the studies that are included in the chapter, it is rather that they are not explicit as to exactly what is being investigated and, therefore, where the limitations are as to what can be concluded. The use of this framework aims to reveal a variety of assumptions that are made about terminology and translation during research.

1. Collecting language in use, allowing a later analysis of variation and change with respect to geography, age, gender or other demographic factors (this is very close to corpus research).
2. Collecting language in use and asking the users for additional information about why they select those lexical items, with respect to e.g. conceptual and encyclopaedic knowledge, emotional impact, sociolinguistic factors. This is a direct request for reactions from participants (though it is not suggested that participants are also asked to label those reactions).
3. Presenting existing terminology and asking participants what it means to them i.e. conceptual or encyclopaedic knowledge of the concept.
4. Presenting existing terminology with the aim of discerning other factors that affect meaning e.g. emotional reaction to lexical items. This could be achieved by indirect means such as presenting synonyms for comparison and/or requesting preferences and reactions to specific terminology.
5. Presenting existing terminology and taking a more direct approach to discerning e.g. emotional reaction by asking people why they like or dislike certain lexical items.
6. Investigating translated terminology and conceptual equivalence.
7. Presenting existing terminology and looking at how alternatives affect outcomes. This is also an indirect approach, but potentially it is more complex to discern whether outcomes are affected by terminology alone or by other factors.
8. A whole additional layer of complexity is added in studies which present existing terminology embedded in a context, be that a sentence, a larger information resource or within a narrative. All of the aspects above (level of knowledge, emotional impact, sociolinguistic factors, modality and language-specific factors) can potentially be investigated and the same approaches (1-7) used. In addition, there are grammatical and syntactic relations to be investigated.

For Approach 8, the impact of terminology (in whatever way – transfer of knowledge, emotional impact etc.) clearly cannot be separated from its context. However, it can be argued that it is impossible to present terminology unaffected by context, even if attempted (Approaches 1-7). Terminology is always affected by the manner in which it is presented, the channel (written, voiced, signed), the setting of the research and, importantly and part of what can be investigated, the background that a receiving individual brings (emotion, fund of knowledge (Pollard, 1998), previous experience of that lexical item). Even if it were possible to present terminology independent of context, are the results obtained as valuable when the terminology is fixed rather than interactive? How transferable are findings if participants cannot engage with the terminology being presented and debate the meaning, either with themselves or with others, in order to understand how the terminology can be applied? This is not to suggest that studies trying to present terminology out of context are not valuable, but simply that their results must, in turn, be interpreted through a lens of potential future contexts.

By considering the published literature through the framework above, this chapter will illustrate that it is impossible for a single lexical item to contain all the information needed to understand its full conceptual and encyclopaedic meaning and that terminology that is out of context or not fully explained is more likely to be understood differently by different people. Before returning to the review of published research, two recent anonymised examples are used to illustrate this point:

1. A link to a report about modified viruses being used in gene therapy was posted online in an open discussion group. In the laboratory setting, a ‘modified virus’ is defined as one which has been attenuated by changing a gene(s) so that they are no longer infectious and no longer cause symptoms of ill health. However, a posted comment in response to the report showed that ‘modified’ had been understood by the poster to mean the virus could be more dangerous and/or more likely to spread as a result of being changed by unnatural means (similar to the debate about genetically modified crops) and therefore was seen as inappropriate to use with humans.

2. During a previous discussion with a health professional, a mother of a deaf child was told that ‘some causes of deafness are genetic and some are environmental’. ‘Environmental’ is used in a medical context to mean any other cause which is not genetic e.g. a viral infection or other problem during pregnancy, a complication during birth or an illness during early childhood. The mother understood ‘environmental’ to

mean something she had done to alter the environment and had worried ever since about what this could have been.

3.7 Research that has investigated knowledge, understanding and perception of existing terminology

Many studies of terminology incorporate aspects of more than one of the approaches suggested in the framework above. Others may not fit neatly into one or other category. For that reason, the following review places studies in a similar order to the above list, but without subheadings.

Research groups within many countries have now produced a corpus of their National Sign Language (NSL), including BSL (British Sign Language Corpus Project, 2012; Schembri et al., 2013), Australian Sign Language (Auslan) (Johnston, 2009) and Sign Language of the Netherlands (Nederlandse Gebarentaal, NGT) (Crasborn, 2010). The corpus is intended to be drawn from all demographic groups within the Deaf population so they can be interrogated later with specific research questions. Slightly differing approaches have been taken. For example, the BSL Corpus Project presented contributors with English words and asked them for their own signs. The Auslan Corpus (‘Signbank’) started with collecting signs direct from participants, but now asks Deaf contributors to upload their own signs for storage. This approach has allowed them to branch out into an area of specialised terminology and recently create the Medical Signbank (Johnston and Napier, 2010) for collection of health signs. It will be valuable to monitor how the latter approach of the Australian group develops.

Pollard and Barnett (2009), building on their previous work recognising lack of access to background health-related information (Pollard, 1998) and existing barriers to healthcare for Deaf people (Steinberg et al., 2006; Barnett, 1999; Barnett and Franks, 1999; Pollard, 1992a), studied the health-related vocabulary knowledge of a highly-educated group of deaf adults (80.8% had a college degree). They found that a third had a score judged as less than 9th grade, which was considered a marker for low health literacy, and that it was not necessarily the vocabulary considered more challenging that deaf participants did not know. However, the authors chose an existing test (the Rapid Estimate of Adult Literacy in Medicine – REALM) which consisted of 66 written English words. With hearing subjects, the object of the test is for each person to read out the words and they receive a point for each word pronounced correctly. Pollard and Barnett amended the test so that the deaf participants were asked to circle the word if they understood what it meant and cross it out if they did not. It can be argued that the test is therefore not measuring the same thing: with hearing participants, the test is clearly measuring cultural scientific literacy/linguistic knowledge (essentially, familiarity with the word) rather than level of comprehension. With the deaf participants, they are asked about their understanding rather than simply familiarity and are free to interpret ‘understanding’ – an interpretation which may vary greatly within the group. Nevertheless, the authors’ conclusion can be accepted that a significant proportion of deaf people do not have good understanding of health terminology in English.

Moving on to think about emotional reactions to specific lexical items, two studies (Condit et al., 2004; Ando et al., 2009) are illustrative of attempts to understand in what way and why individuals (whose hearing status was not mentioned in these studies) have preferences for some terms over others, when the terms have very similar referential (see definition overleaf) meaning. They focus on ‘gene mutation’ and the synonyms ‘variation’, ‘alteration’ and ‘change’ (Condit et al., 2004) and the Japanese words for ‘change’ and ‘lesion’ (Ando et al., 2009). Ando et al. acknowledged the use of Condit et al.’s English questionnaire in their study, but as the former group had translated it into Japanese, it is informative to be able to compare similar studies carried out in two languages. Ando et al. carried out a factor analysis to measure four aspects of the concept and how these varied between the synonyms and between two groups (people who had experienced genetic counselling and university students). The four aspects were whether a gene mutation/change etc. was a favourable event, the effect on development, the rate of change and whether it was intentional or not. The authors noted that ‘the results revealed that situation-specific differences seem to affect term preferences.’ (ibid.), recognising that it was not only knowledge of the concept that affected perception of the word, but also the socially constructed meaning of a synonym as it met the individual’s own experience (or not) of a genetic condition. For example, those who had experienced genetic counselling had lower scores for Favourability and Development, seeing these aspects of a mutation more negatively. An interesting addition to Ando et al.’s research would have been a qualitative investigation of individuals’ own descriptions of their emotional reaction to the terms to contextualise and triangulate with the results of the factor analysis. This was included by Condit et al. through use of focus group discussions. Their key finding was that ‘mutation’ was seen as a ‘scary’ (ibid.) term, conjuring up images from science fiction. In both groups, mutation was seen as the technically correct term, but one which should be avoided in health discussions or public health messages. These results are not surprising, but it is of interest to try and define the underpinning cognitive aspect of language that has been explored here.

In the context of semantics, Crystal (2010a p106) discusses the meaning of ‘meaning’ itself. He explains the difference between two aspects: *reference* (the thing that is being referred to) and *sense* (what it means to the individual), a distinction originally made by Frege (1892). The balance between the sense and the reference of a concept when an individual is choosing how to say something depends on why they are saying what they are saying. If it is more about transmitting facts, then the reference may be more important, whereas if the relationship of the term to the individual is more significant, perhaps the emotional resonance or a metaphoric meaning, then the sense is more important. This may suggest why the meaning of terminology used in a scientific setting was more likely to remain close to the encyclopaedic definition: because the reference tends to be more important in the lexical item’s use than the sense. This could be why ‘mutation’ is suitable for a discussion between scientists, but is one word which, because of its use in literature and media, has taken on negative emotional sense and is therefore unsuitable for discussions involving lay people, particularly in a clinical setting.

Crystal moves on to introduce *semantic space* (ibid. p107) which is defined as the psychological and emotional meaning of a lexical item to an individual. He uses the example of how different animals take up different semantic space in our heads with respect to size and ferocity. He also highlights that semantic space for the same concept can vary between cultures: an important point which will be considered again later in the chapter. The idea of semantic space as the psychological meaning of a lexical item to an individual is very different from, for example, Pinker’s (2008) explanation of what a lexical item is doing to the brain when it is understood with respect to the dimensions of space, time and causality. The semantic space is related to a lexical item’s sense whereas Pinker’s focus is more on the description of the reference. Pinker’s argument backs up the importance of an individual gaining at least conceptual knowledge of a lexical item, if not encyclopaedic knowledge, as it is only with this level of understanding that they would have a clear picture of the reference and be able to develop a sense of what the concept means for them. A slightly separate aspect of language which is given just a small paragraph at the end of Ando et al.’s paper (2009), is the finding that the Suddenness of Change factor for ‘gene mutation’ was higher than that for ‘gene lesion’. They posit the explanation that the Japanese term for ‘mutation’ contains the term ‘sudden’ and that, therefore, the word mutation may conjure, for these participants, a sense of suddenness. This is an example of how an individual lexical item’s sense may vary between different languages, in this case as a result of one word’s meaning influencing another.

Another study that focused on how the sense of lexical items affected preferences (though the concept of *sense* was not labelled) was Abramsky and Fletcher's (2002) questionnaire study with English-speaking participants (whose hearing status was not mentioned). Their 581 participants were asked questions such as 'which sounds more worrying' and 'which sounds less common' for terms that were similar in terms of frequency ('unusual' and 'relatively rare') or referential definition ('a chromosome rearrangement' and 'a chromosome translocation'; 'an extra chromosome' and 'a trisomy'). Like the 'mutation' studies discussed above, they found that the lexical items judged more worrying were those where the meaning had been influenced by other connotations e.g. the word 'syndrome' was more worrying for the majority than 'association'. In addition, however, they suggest a further reason: that more technical and less well-known lexical items were more worrying because people could not gain a sense of their meaning so this worry may stem from a fear of the unknown and the implications for the individual.

The next study to be considered is one focusing on the translation of terminology between languages: Shaw and Ahmed's (2004) retrospective investigation of leaflets in Urdu about genetics and genetic counselling translated from English. As discussed in Chapter 2, many studies have acknowledged the barriers to genetic counselling for people from minority language cultures living within a dominant language culture such as English and the importance of accurate translation and interpretation in overcoming these barriers. However, this study is one of only two known published accounts (Belk, 2006; Shaw and Ahmed, 2004) of specific translation challenges between English and another language in the area of genetic counselling. They highlighted other contributing problems such as difficult technical language in the starting script, content that was insensitively written with respect to specific cultural norms (e.g. around consanguinity) and the absence of dual language leaflets (text available in Urdu and English alongside each other) or contact details for someone to speak to in Urdu or Panjabi [sic] (ibid.). The main focus of the paper was on the problematic translation of specific lexical items and concepts. They found that these were not limited to difficult technical concepts and they grouped the types of error into three main categories. The first was 'Inaccurate information', where the reference or definition of the concept itself was simply wrong. This was sometimes as a result of focusing on finding a lexical equivalent without adequately considering whether the chosen translation was also pragmatically equivalent (Baker, 2011) e.g. 'planning your family' becomes 'family planning', 'tests during pregnancy' become 'pregnancy tests', 'genes' becomes 'inherited cells', 'high-risk screen' becomes 'too much dangerous screen', 'rare' becomes 'precious'. These types of error raise the concern that the translators of these examples did not have adequate conceptual or encyclopaedic knowledge of a lexical item to enable them to translate accurately.

The second category was ‘Difficult or unfamiliar Urdu vocabulary’ where the authors explain that a highly educated Urdu reader may know that e.g. ‘beze’ means ‘ovum’, but this is highly unlikely to be known to a lay reader. Another example is ‘stage of pregnancy’ where a correct lexical equivalent may be found, but it is not a concept that is discussed as widely in Urdu-speaking cultures as in the UK English-speaking health services. The third category was lexical items for which there was no Urdu equivalent known. In the same way that Major et al. (2012) highlighted the use of placeholders in interpreting from English to Auslan, Shaw and Ahmed found common use of English words transliterated into the Urdu Nasta'liq script e.g. ‘genes’, ‘recessive genes’, ‘ultrasound scan’, ‘cousin’ (a particularly important one, given that, in Urdu, ‘apparently equivalent [kinship] terms may denote a wider or a narrower category of kin than in the dominant language’ (ibid.). Alternatively, text was missed altogether e.g. ‘increase in birth defects’ becomes ‘increase of birth’.

All three categories of error highlight that, if the definition and explanation of terminology in an English leaflet is inadequate and relies on the reader having at least a level of cultural scientific literacy for the terms included, then it is unlikely to translate well into a minority language without serious thought being given to pragmatic equivalence and/or the need for additional background to allow the concept to be adequately explained. For the same reasons as discussed at the start of this chapter in relation to BSL, the lexical items needed may not exist at all or may have additional cultural or social meaning that does not exist or is different in Urdu-speaking cultures. Without considering how to overcome these translation challenges, it is unlikely that readers would be able to understand the meaning of concepts from either the referential or sense perspective i.e. neither theoretically nor relating it to their own situation.

The next two studies to be addressed both highlight further that it is extremely difficult, if not impossible, to discover and document all aspects of individuals’ understanding of terminology. As the first few studies have illustrated, there is breadth of aspects of knowledge (referential, encyclopaedic, emotional, social, cultural), but also depth of knowledge from familiarity with the lexical item to the ability to link aspects of the meaning in with existing personal and cultural knowledge. The abstract of Lanie et al.’s (2004) paper said that they sought to find out how individuals used and understood the terms ‘genetic’ and ‘genes’, but the detail of the paper showed that they had made the methodological choice to embed the terms in the questions ‘what do you mean when you say something is genetic?’ and ‘where are genes located in the body?’. Respondents were encouraged to answer the questions in a detailed qualitative way so the data was broad and open-ended. However, the use of the questions does point the respondents’ attention to two aspects of the concept of genetic/gene: the action and the spatial location. These are only two of the dimensions and relationships that are part of the mental representation of that concept in the brain (Pinker, 2008).

Lanie et al.’s (ibid.) aim was not to discover all aspects of the mental representation of the concept in these participants, but to highlight the range of knowledge of the action and spatial location. They showed that many participants had little or no idea of the location or function of genes so pointed out that it would be difficult for individuals to build on these uncertain foundations to understand, for example, genetic counselling discussions or new genetic concepts discussed in the media such as gene therapy. They also introduced the valuable consideration of metacognition: thinking about thinking or, in the context of their study, an individual’s awareness of what they know and what they do not know. They point out that knowing one does not know something is more valuable to the individual than inaccurately believing one’s understanding to be correct. This ‘illusion of knowing’ (Park, 2001) can be one disadvantage of the wide availability of information through media: it is possible to have cultural scientific literacy for a wide range of terms without realising the gaps in knowledge that prevents one making sense of the concepts.

It is valuable to pause briefly here and consider the specific issues for Deaf people compared to hearing people. It could be hypothesised that Deaf people, on average, may have a greater illusion of knowledge because of the fewer opportunities available to them to check and be challenged about their existing knowledge. As discussed in the previous chapters, Deaf people are likely to miss spoken discussions amongst family or colleagues and the opportunity to overhear conversations in public or on the radio. However, the opposite could also be argued: that hearing English speakers could have a greater illusion of knowledge precisely because of familiarity with scientific terminology. This is a key point about sharing the same spoken language: the level of understanding is much less visible than it is in a signed language. A Deaf person may choose to fingerspell an English word to make it clear that they are familiar with the correct term. Although this achieves their aim, the fingerspelt and the spoken English word are then equivalent: little can be deduced about the user’s level of understanding beyond recognising that they have linguistic knowledge of that term. As will be shown later, lexical items in BSL have the capacity to convey different aspects of meaning and potentially to a greater degree.

Returning to the second of the two papers, Christensen et al.'s team (2010) contained several of the same authors as Lanie et al. (2004). Again, they stated their aim as examining understandings of basic genetic concepts. However, although they asked for a simple agree or disagree response, the statements they presented were far more complex than the questions in the previous study e.g. 'two women will always be more genetically similar to one another than a man and a woman' and 'there are different types of genes in different parts of the body'. Although there may be value to knowing what proportion of people can accurately answer those questions and how it varies between demographic groups (one of their objectives), the results could not be used to discern on what level these participants understand the contributing concepts and terminology. The statements require the participants to bring together several genetic-related concepts and make sense of them all. This study is an example of where it could be valuable for the authors to define what they mean by 'understand'.

Lang et al.'s (2007) paper takes us back to working both with a signed language (American Sign Language, ASL) and to a focus on single lexical items. Their study was in the context of terminology in science teaching. They aimed to elicit participants' direct responses to particular terminology and also for participants to elaborate on the reasons for their preferences or objections. The focus was on the 'Classroom of the Sea Project' so all the lexical items were related to sea creatures. The group collected several hundred lexical items from the general science curriculum and sought pre-existing ASL signs. As other authors have also noted (Roald, 2002), the authors comment that classroom teaching in ASL and English is often interrupted by discussions of the most suitable sign to use. They therefore designed two studies: the first presented science teachers (some of whom were experienced ASL users and some not) with signs in three groups: Group 1 were selection errors i.e. the sign itself was morphologically inaccurate to portray the semantic meaning e.g. the sign for DISTANT was used for DISTANCE; Group 2 used correct morphology for the general semantic meaning, but the pragmatic meaning meant that it was a poor choice of sign e.g. RESPIRATION was signed as for human respiration with the chest rising and falling when the signer was talking about fish; Group 3 were signs that had been invented for concepts that did not have a known ASL sign e.g. CURRENT was signed as a flow in one direction. The teachers were asked to comment on the signs, whether they were happy to use them and their reservations. In the second study, the teachers were presented with several variations of sign for five animals (SEAL, CRAB, WHALE, DOLPHIN and SHARK) and asked which they preferred. The comments presented in the results for both studies picked up many of the issues relating to semantic space and sign choices in ASL and other sign languages. For example, one participant commented on CURRENT: ‘This sign for FLOW gets at part of the idea, but there is more to a current. The word current refers to the strongest area of flow. How to show that?’.

This example is a good illustration to introduce one issue about a visual-spatial language: that because the language has access to expression in four dimensions (three spatial dimensions plus time) rather than just one (time alone because English and other spoken languages are linear) (Meier et al., 2002 p11; Meier, 2006), it has very different resources available to it in how it can express space and time. This means that signed languages lend themselves to iconic signs (Emmorey, 2002): signs that are representative of the visual appearance of the concept they encode in the same way that onomatopoeic words are representative of the sound of the concept they encode. As with any language, the resources available to express ideas vary so that some semantic dimensions of the meaning are easier to portray in a signed language than a spoken language and vice versa. So the implications for the example in the previous paragraph (CURRENT) are that the sign is iconic of water flow: therefore giving much more semantic information about the referent through the use of movement and space than the English word ‘current’.

However, an iconic sign can only represent some of the semantic dimensions, as pointed out by the participant commenting on the CURRENT example. Which dimensions should be encoded within a sign may be a source of debate if terminology is being created or several signs are available and this issue will be addressed further in the next section. Although the extra information provided by an iconic sign can aid the receiver in engaging with and making sense of the concept, the RESPIRATION example illustrates that a sign that has evolved in one context may not be appropriate in more general situations. This shows that word meanings cannot be assumed to map neatly on a one-to-one basis between languages: there is a parallel here to the previous example of ‘cousin’ not referring to the same degree of kinship in English and Urdu (Shaw and Ahmed, 2004). It highlights that translating between a spoken and signed language, in the same way as between two spoken languages, can be potentially more challenging than developing terminology or an information resource from first principles: one reason being this potential for giving incorrect information because of the pragmatic meaning of a sign. In this case, the use of RESPIRATION would give the erroneous information that fish breathe using a trachea and lungs.

The other dimension introduced by this paper is the variation in reasons given for preferring certain signs. For example, some participants liked the iconicity of a sign, believing that a visible link to the etymology would make both the concept and the lexical item itself more memorable for their pupils. Others preferred an initialised sign e.g. ‘Dolphin’ signed as a letter ‘D’ plus a classifier (Sutton-Spence and Woll, 1999b p47) moving as a dolphin would swim through the water, because they thought the inclusion of the initial letter would help pupils make the link to the English word (an example of the power relationship between a minority sign language and surrounding majority spoken language which is unlikely to be replicated in translations between two spoken languages). Yet others objected to the use of an initialised sign precisely because it was English-like. In their literature review, the authors (Lang et al., 2007) summarise the, so far, limited evidence of links between the categories of signs illustrated above and how well they were understood and memorised by learners. This review flags up the need to consider the evidence as to how lexical items are memorised and used; and how different languages and modalities differ in this regard. This area will be addressed later in the chapter.

In the final part of this section, several studies illustrate some of the pragmatic factors that can significantly alter individuals’ understanding of the reference of a concept and influence their engagement with that concept and the personalised sense that they take from it. All involve genetic information and genetic counselling, but none were carried out with d/Deaf people. However, the conclusions appear potentially valid for d/Deaf people and sign language users as well.

Roter et al. (2007) focused not on which terminology was used, but on the effect of the complexity of communication in which it was embedded. They recorded genetic counselling sessions with actors playing clients, scored the utterances of the genetic counsellor for use of unfamiliar terminology, complexity, pacing and density and then asked both the genetic counsellor and the actor to give feedback on satisfaction with the session and sense of engagement. They found that a higher proportionate use of complex terminology correlated with several factors including shorter sessions, the counsellors’ speaking turns being fewer and denser, higher readability demand and low interactivity. In turn, greater density of dialogue and less interactivity correlated with lower satisfaction on the part of the actor client and lower ratings of the counsellors’ nonverbal effectiveness. The authors are not explicit about this, but the results therefore suggest that greater use of challenging terminology by a genetic counsellor is only part of their less effective communicative awareness. Conversely, it suggests that counsellors who need to use this terminology should be able to mitigate it and help their client engage with it by use of other communicative strategies and components. If a genetic counsellor is less effective in her communication, is this partly because she is relying on the client's linguistic knowledge of the terminology to transmit the meaning? It suggests that professionals need to check further and/or add to the client's conceptual/encyclopaedic knowledge to ensure the client can make their own sense from referential knowledge of the concept.

Klitzman's research (2010) also did not focus on specific terminology or concepts, but used a method which sought a more context rich understanding of people's knowledge of genetic tests and personal health risks and how they engaged with these. He carried out two-hour qualitative interviews of 64 individuals with a personal or family history of Huntington's disease, breast cancer or Alpha-1-antitrypsin deficiency. His focus was on how 'misunderstandings' is defined: he found evidence of lack of cognitive understanding of, for example, genetic inheritance patterns and the level to which genetic tests can predict level of risk. These are both examples of individuals not having gained a full picture of the reference aspect of the concept. However, as in the studies by Condit et al. (2004) and Ando et al. (2009), he found that his participants' emotional reaction to the information played an important part in the sense they took from the information. In this case, because the participants were themselves affected or at risk for the conditions, with potentially serious implications for their health, the emotional reaction was personalised rather than disinterested. It was apparent from the quotes that the participants varied in their awareness of when there was a mismatch between the reference and the sense they had taken from a concept e.g. 'I look more like my father's side of the family. I think that's stupid, because I don't think it really makes a difference who you look like. But I guess I'm hoping for the best.' (Klitzman, 2010 p436). There are parallels here with the work done by Richards (1996), who was instrumental in bringing 'lay beliefs of inheritance' to prominence in the genetic counselling literature: the concept that individuals use their personal and family history of health and other conditions to draw their own conclusions about how genetics works. The difference is that Klitzman is focusing particularly on individuals at personal risk and concluding that it is not only an apparently logical deduction from the existing evidence of one's own experience, but that the emotional reaction to the threat to self can contribute to a misunderstanding of the referential part of the meaning of a concept.

Work has also been done on cultural factors that impact on the pragmatics of genetic concepts. Shaw and Hurst (2008) studied understandings of genetics, causes of illness and inheritance amongst British Pakistani families who had previously received genetic counselling. They found that individuals had sometimes not been able to engage with the information at all and did not have any referential understanding of the concept. Others held a genetic explanation alongside a cultural belief that made sense of the science for them e.g. a participant reported believing the explanation of a recessive faulty gene as the cause of her son's metabolic condition, but also considered that it had been God's purpose for him to have it. Others had attempted to combine a genetic concept with culturally-shared knowledge (i.e. ways in which a community had made sense of prevailing genetic topics discussed in relationship to their culture) and, as a result, had misunderstood the referential meaning e.g. 'When asked what he understood as 'genetics', Mr B, father of a boy with learning difficulties, said 'genetics is what happens when you marry a cousin' (Shaw and Hurst, 2008 p376). These examples illustrate that it is difficult to draw a clear distinction between individual emotional reactions and cultural beliefs when considering what influences the sense that individuals make of genetic concepts.

Other studies (Nelkin, 2001; Petersen, 2001) have suggested that how genetics is portrayed in the media – a specific area of culture which both reflects and influences wider cultural beliefs – affects how members of the public make sense of the concepts. Specifically, both authors concluded that the media tend to emphasise the 'medical benefits' of genetic research and frame new developments as a 'breakthrough' (Nelkin, 2001). However, Bates (2005) concluded that, although media does influence cultural beliefs, his large focus group study (25 groups with 216 participants) showed that members of the lay public approach news reports about genetics with a critical and questioning perspective and can therefore mitigate the messages they take from it. Nevertheless, there was also evidence in this paper that people try to make sense of the information they are given, but if they do not have appropriate background information with which to assimilate the new concepts and against which to judge them, they are likely to misunderstand fundamental genetic concepts. This may be particularly the case when genetic ideas are presented within science fiction or drama and incorporate elements of conjecture or overextension of current knowledge.

In summary, this section has explored different ways in which existing unfamiliar and complex lexical items and the concepts they represent have been investigated in previous research. These range from consideration of terminology in isolation, with respect to understanding of the referent itself, as well as the impact of factors such as emotional reaction in altering the sense and semantic space for an individual. Different methodological approaches have illustrated that the questions used and the contextualisation of the concepts significantly affect the semantic dimensions to which participants will pay attention.

3.8 The different approaches to creating or collecting terminology in BSL and other sign languages

The focus of this section is on studies which have looked forward to collect, create, formalise or standardise terminology rather than looking back to analyse how existing terminology is used or understood. There is overlap between the corpus studies that have collected a wide range of existing lexical items (British Sign Language Corpus Project, 2012; Schembri et al., 2013; Johnston, 2009; Crasborn, 2010) and the studies in this section that also collected lexical items, but in a more focussed way with the aim of producing specific glossaries, terminologies in a certain subject area or focussed information resources.

As discussed earlier, a minority language such as BSL does not have the same opportunities for terminology to develop at a similar rate and/or with the same level of use as in the surrounding majority or dominant language, either through discussion and/or through practical application of the concept(s) in question. In this case, the issue is often the identification of a lexical item in the minority language that represents a concept already understood and named in the majority language. This raises the challenge of identifying the advantages and disadvantages of different methodologies when considering new or developing terminology. Terminology can develop through different routes, some conscious, some unconscious, some directed by a small group of people and others through growth within a community. The route by which terminology develops is not always made explicit – or, indeed, known – but this section aims to summarise what can be deduced from existing work.

As in the previous section, it is valuable to set out a framework of possible ways in which lexical items are determined, against which the work in this area can then be considered:

1. The lexical item labelling a new concept that does not (yet) have a directly comparable meaning in another language can develop organically or deliberately through any of the normal channels available to it (Sutton-Spence and Woll, 1999b): iconicity; metaphoric borrowing of an existing sign with a related or contrasting meaning e.g. some sign names; simultaneous use of signs; compounding of signs; or a new sign which is not necessarily recognisably influenced by existing signs, but must obey the morphological rules of the language e.g. DEAFHOOD.

2. A lexical item labelling a concept already known in another language can be consciously developed to be representative or iconic of some dimension(s) of the concept's referent e.g. CURRENT in Lang et al. (2007).
3. Lexical item(s) labelling a concept already known in another language can be collected as part of a corpus or smaller project and then the most suitable item discussed and a consensus reached. The discussants may vary depending on their practical availability, starting with those individuals that need to use the signs for a specific purpose. Ideally such discussants should be expert in one or more ways (professionals working in that subject area, Deaf, fluent signers, interpreters, translators).
4. A similar approach to 2 or 3, but with a focus on how the context of lexical items changes both their semantic and pragmatic meaning i.e. how the meaning is modified by the grammatical and syntactic resources of the language and the extralinguistic factors such as culture and relationship to other concepts. This approach may be needed when developing a glossary that works in a specific setting or for a particular information resource.

3.9 Research that has created or collected terminology in BSL and other sign languages

This section now analyses the available work that has looked forwards to gather and create terminology and resources, with reference to the framework above. As with the studies looking retrospectively at existing terminology and resources, not all studies fit neatly into just one of the sections above.

One of the most notable and well known lexicons collated in a specific area is the reference book of BSL terminology in mental health produced by a specially convened group of Deaf mental health professionals (Deaf Professionals in Mental Health, 1997). This was published by the British Society for Mental Health and Deafness and is known colloquially as the big red book. It contains over 130 lexical items for mental health concepts e.g. 'hallucination', 'central nervous system' and related concepts e.g. 'IQ', 'empathy', 'accommodation', accompanied by written English definitions. The book's introduction summarises the aims of the lexicon as firstly to increase access and awareness of terminology and concepts for deaf consumers of services and professionals from other disciplines interacting with mental health services. Secondly, it aims to 'enhance the status and value of deaf people who work in this very isolated field' (ibid.). The introduction also explains that the lexicon was developed from an initial planning meeting, the formation of a working group and then subsequent interviews with mental health service users and Deaf lay people. The signs most frequently used in these discussions were included in the reference book. The authors also state that ‘many other signs were created by the Working Group’, but give no further information about the development process. They do acknowledge that the compilation was ‘simplistic’ (ibid.), but seek to present the signs as only the start of further discussion. There has been no published research from the group further explaining the development of the lexicon or analysing the choices and influencing factors. This would be of great interest as only limited conjecture is possible from viewing the photographs of BSL signs in the book. For example, there are signs which would appear not to portray their full meaning within the two-dimensional photographs e.g. the sign used for AFFECT appears the same as the well-known FEEL. AFFECT therefore does not appear to incorporate the additional dimension of meaning of ‘affect’, that is, feeling or mood, but particularly feeling or mood as manifested in one’s facial expression or body language. Similarly, the photographs of ANOREXIA NERVOSA appear to be the same sign as PERSON GETTING THINNER. However, when signed, these pairs of lexical items are likely to be subtly different from each other as explained below.

The examples illustrate a difference in the language resources (Slobin, 2003) available to BSL signers compared to English speakers. BSL (like other sign languages) has a much smaller ‘established lexicon’ (Sutton-Spence and Woll, 1999b Chapter 11) than English, but makes great use of a ‘productive lexicon’ (ibid.). The latter term means that the core lexical items within the language (distinct recognised signs on the hands) can be modified in many ways to label different, often related, concepts. These modifications can be through degree of movement, variation of sign placement in relation to other signs or anaphora, through use of facial expression (e.g. adverbial manner or emphasis, adjectival variation) and body language (e.g. shift to signal emotional relationship to concept) (Sutton-Spence and Woll, 1999b). It is these modifications of the basic handshape which would distinguish AFFECT and FEEL; ANOREXIA NERVOSA and PERSON GETTING THINNER, though context, grammar and syntax also help to make these semantic distinctions.

The next paper illustrates an alternative way of presenting a subject-specific lexicon. The Medical Signbank established in Australia (Johnston and Napier, 2010) has already been mentioned in the context of sign language corpora. It is also relevant to this section because of the methodology the researchers chose to develop this focused lexicon. Corpora have been gathered directly through videos of narratives, interviews and conversations with invited participants, but Johnston and Napier are clear that they wished to promote ‘cooperative language development’ (ibid.) by encouraging Auslan users (interpreters and Deaf lay people) to contribute their own signs, report successful or unsuccessful use of signs already on the site and report medical/mental health terms in English for which there appears to be no existing Auslan sign. They propose this approach as an alternative to ‘traditional language planning’ (ibid.) which they characterise as tending toward a hierarchical and top-down approach such as that used by the Académie française (2014). They consider language planning as having an important place, but highlight how it can be problematic to define who should be involved in such discussions. It seems clear that their approach can facilitate easier sharing of existing signs and provide a central language repository for the low incidence, geographically-dispersed population of Auslan users. Unfortunately, the Auslan Signbank website (Johnston, 2009) is not currently open for contributions and it is not possible to ascertain from either the website or Johnston and Napier’s paper (2010) exactly how people contributed to the lexicon. The wording of the paper suggests that contributions were through written English only. They report that more than 50,000 users registered on the website over five years, but that less than a 150 Deaf Auslan users took part (of an estimated 7,000 Deaf Auslan users in Australia) and only a proportion of these contributed feedback on signs. This has meant that the innovative potential of the site has translated to only a limited contribution to lexical development, but they report its continued popularity as an online dictionary. All the signs are available as short video clips embedded in a webpage with an English definition of the concept alongside. This technology overcomes the limitations of the photographs as discussed in the mental health lexicon (Deaf Professionals in Mental Health, 1997) and as used in previous paper dictionaries such as the groundbreaking Dictionary of British Sign Language/English (Brien, 1992). This is one example of how rapid computer hardware and software developments over the last two decades are revolutionising the digital resources available for transmission of sign languages. This area is further discussed in Chapter 5.

Johnston and Napier (2010) predominantly presented their methodology so their detailed analysis of the collected signs is not yet known. Given the low levels of feedback via the website, they propose online surveys with Auslan interpreters and focus groups with interpreters and Deaf lay people to collect preferences and popularity of signs. They do not state that they will be asking about underlying reasons for preferences. They plan to carry out a linguistic analysis which looks at ‘language-internal principles of phonological well-formedness or semantic appropriateness and distinctiveness within the existing Auslan lexicon’ (ibid.) and do not explicitly state that they will also seek to identify pragmatic influences such as cultural or sociolinguistic context e.g. English influence. They are seeking statistics on ‘accuracy, comprehension and popularity' (ibid.) of signs and it will be of interest to see how they measure accuracy and comprehension, given the discussion in the previous section.

The BSL Science Signs Glossary (Scottish Sensory Centre, 2014) has been an ongoing project for some years with the aim of increasing the lexicon within the natural sciences and maths for use in schools. They have not published any peer-reviewed or publically available written evaluation of their development process nor the acceptability of their lexicon to Deaf BSL users, though they are well-known for presenting at science festivals (O'Neill, 2008), complete with an excellent live chemistry demonstration for children, and in educational settings. Their approach contrasts with the Medical Signbank in that they have taken a clear language planning approach. O'Neill (2008) explained their development approach for new lexical items and the range of backgrounds of the planning group members: all have undergraduate or postgraduate degrees in a natural science or maths or equivalent relevant experience with wildlife and all are Deaf. In a similar approach to Lang et al. (2007), they begin by identifying the English terms required, collecting existing signs for these terms and then evaluating these signs (they do not specify how they evaluate them). At this stage, the group discuss the definitions of scientific concepts (effectively, the referential meaning) and use morphologically correct features of BSL to create new BSL signs where none exist. After further consideration by the group members (again, they do not specify in what way), they film the lexical item and place the video and a definition in both BSL and written English on their website (Scottish Sensory Centre, 2014). Of particular interest in their approach is a lean towards developing signs clearly representative of an important dimension of the concept's reference e.g. the lexical items EXOTHERMIC (see http://www.ssc.education.ed.ac.uk/bsl/chemistry/exothermic.html#start) and ENDOTHERMIC (see http://www.ssc.education.ed.ac.uk/bsl/chemistry/endothermic.html#start) incorporate the dimension of 'giving out' and 'taking in' during a process of change. In other words, they have decided that a lexical item should not only be a symbolic marker for a concept whose referential meaning would be learned through a definition and/or contextual use, but should incorporate a degree of semantic information. This provides the receiver not only with linguistic knowledge, but with a level of encyclopaedic knowledge incorporated within the lexical item itself. Other newly created lexical items in a variety of languages can incorporate semantic information about the concept's reference e.g. World Wide Web: part of the referential meaning of the whole can be drawn from the referential meaning of the component morphemes giving, in this case, a meaning which is partly metaphoric. In the example of the compound sign EXOTHERMIC, the component morphemes can therefore be identified as CHANGE

START-GIVING OUT-CHANGE END. The facial expression is congruent with the component morphemes in that it supports the dimension of OUT and adds the dimension of QUITE A LOT. This is a good example of the significant use BSL makes of a productive lexicon: the referential meaning of new lexical items can be fully or partly discerned from knowledge of the established lexicon together with knowledge of the syntactic rules of BSL.

Wolverhampton University (2005c) built up a similar online glossary of BSL terminology for science over several years, but aimed at undergraduate level. They used similar presentation and methods to the Scottish Sensory Centre group, with the significant exception that they have not developed new signs for concepts. They state their methods on the website as follows: ‘Signs were not created, but were found by interviewing individual or groups of Deaf professionals, and looking at signed TV programmes (SignZone, See Hear etc.). On completion of the research, a panel of 'critical friends' evaluated them to check that they were culturally and linguistically appropriate. Only those signs deemed appropriate by the panel have been included.’ (ibid.). As with the previous group, they have not published any detailed information about their methods and do not explain how their panel evaluated the signs. They do list the six members of their panel and the more than 40 BSL consultants on the project: these individuals include some of the best known and respected Deaf presenters and Deaf and hearing interpreters/translators in the UK so the project is certainly culturally sensitive and embedded within the Deaf community. The same team have also worked on other subjects with specialised technology including engineering (Wolverhampton University, 2005b) and art and design (Wolverhampton University, 2005a).

In an entirely different subject area, there is an ongoing long-term project to translate the Bible into BSL (BSL Bible Translation Project Team, 2009). The team includes Deaf and hearing theologians, ordained ministers, interpreters and lay people. It is mentioned here because it builds upon the established discipline of Biblical hermeneutics and translation: an article available through the website exemplifies and is explicit about the careful balance they strike between literal and dynamic translation in seeking the ‘closest natural equivalence in BSL’ (Evans, 2014). They start by careful discussion of the semantic and pragmatic meaning of the original Greek texts and use what is known about the cultural and historical environment in which it was written to choose appropriate lexical items for the translation. As yet, there is no published evaluation of the translation process. Members of the team have given examples in personal communication (Hunt and Raistrick, 2012) e.g. when translating ‘they went up onto the roof’, roofs in that time and location would have been flat whereas the standard sign ROOF is iconic of a pitched roof. There are also plants referred to in the Bible which are native only to that area and do not have a specific BSL sign: decisions have to be made about how to sign and explain such concepts.

One of the team is explicit about the need for awareness of ‘the ‘hermeneutic cycle’…we understand the whole by the parts but also cannot understand the parts except with reference to the whole.’ (Evans, 2014). In other words, to gain a full understanding and therefore achieve a meaningful translation of a text/information resource, one must consider the meaning of the individual concepts, but also their context and how the meaning of the whole may be different from the sum of the parts. There are parallels in this approach with that taken in the research summarised in the next paragraph.

This thesis developed partly from a project translating English leaflets about genetic counselling into BSL (Belk, 2006; Genetics and genetic counselling translation team, 2005). The project team comprised a hearing genetic counsellor (Belk), five Deaf translators who were about to complete their final translation/interpretation qualification and their hearing tutor, an experienced interpreter. The aim was to achieve a dynamic translation of an entire leaflet – ‘text’ – so in the process, it was necessary to pull apart the genetic and inheritance concepts and consider not only what lexical items should be used in BSL, but how their relationship might be represented through the productive lexicon and syntactic resources of BSL. Detailed discussions were needed at the start as the translators did not have conceptual knowledge of the majority of the genetic concepts and were not aware of any existing genetic terminology with the exception of the ubiquitous iconic sign representing a double helix (two fingers on both hands twisting whilst moving apart) for GENE, GENETIC and CHROMOSOME: the distinction being made by use of the English mouthing. At that stage, the lexicons discussed in the previous sections had not yet been made available online. Even if they had, one gives the commonly used double helix signs (Wolverhampton University, 2005c). The other has created several distinct signs (Scottish Sensory Centre, 2014), but the dimensions they chose to incorporate in the lexical items were not those that take advantage of the contextual relationships between the signs. For example, GENE MUTATION is signed as classifiers representing paired nucleotide bases (left and right index fingers) and then movement out of alignment to signal a change, but this lexical item is not related to GENE which is a finger drawn across the spatial location of a double helix that has just been signed. During the genetic counselling project, the team discussed a metaphor not uncommonly used in genetic counselling consultations: the idea of a library with shelves analogous to chromosomes and instruction books analogous to genes. DNA is the paper and ink of which the genes/books are made and mutations are the changes in spelling or missing/extra pages in a book. From this understanding, the translators decided upon the use of the double helical sign for CHROMOSOME and lined the genes up using a repeated O hand along the anaphoric location of the chromosome. This then allowed them to ‘pick up’ a gene for apparent closer inspection when discussing MUTATION (signed as SMALL-CHANGE over the location of the gene) (See Glossary at Genetics and genetic counselling translation team, 2005)

The consultation that took place during the development of terminology in these projects means that the terms defined are likely to be acceptable to many Deaf users, even though the routes by which they were developed varied. The studies highlight the diversity of perspectives towards whether lexical items should be collected from language in use, allowed to evolve or created with conscious planning towards aspects of meaning that can be incorporated in the lexical item itself.

In summary, this chapter has taken a close look at terminology and communication of concepts, both out of context (as much as is possible) and embedded in a context. In doing so, it has highlighted a number of challenges and the decisions that have been taken by particular groups. The challenges include potential for influence of a lexical item in a minority language by the majority (source) language; potential for greater understanding or misunderstanding due to dimensions of the lexical item itself or contextual influences on meaning; English-influenced signing such as transliterated fingerspelling or initialised signs. However, the range of approaches also highlights the rich, but varied resources of different languages and how grammatical and modality differences influence terminology development and translation. In terms of the specific focus of this study, the chapter evidences the importance of individuals being able to contribute directly in their own language, so avoiding the even greater communicative complexity and subjective decisions necessary when translating between languages.

Chapter 4: Methodology and Methods

4.1 Introduction

The three literature review chapters have contextualised this research within the complex interactions between identity, society, culture, language and cognition. Chapter 1 argued that perception of the condition of deafness is subjective and affected by multiple factors, not only related to the individual, but to their social and physical environment. Health services, in common with other public services, are provided from the perspective of the dominant culture and, currently, do not adequately meet the varied requirements of d/Deaf people. Chapter 2 illustrated this argument with the specific example of genetic counselling, where these barriers exist despite the ethos within the modern genetic counselling profession to maximise the autonomy of the person attending the service and value the perspectives that they bring to the consultation. A significant proportion of the existing barriers to accessing services are related to communication and language. Chapter 3 investigated the relationship between language, information, knowledge and cognition in order to appreciate the challenge when aiming to communicate clearly and be understood both within a single language and between languages. In the context of this thesis, there are specific challenges of communication arising from the meeting of a dominant and a minority language that have different modalities and where the encounter is often mediated through a third-party interpreter.

This chapter takes the contextual understanding gained through the review of the literature and identifies the problem which the research then seeks to address. The problem is then focused down to the purpose, and thence to the aims of the study. Next, an argument is made for an appropriate ontological and epistemological orientation to inform the stated aims. An ontological position of subtle realism (Hammersley, 1991) and a phenomenological epistemology (Husserl, 1970) are adopted as the lens through which to reflect on the aims. The specific objectives of the study are next identified and the methods associated with each objective are set out. Finally, I address the ethical issues associated with the study and, given a phenomenological orientation has been adopted, a reflexive note is included considering my positioning as the researcher within this study.

4.2 Purpose of the study

The purpose of the study is determined by the delineation of the contributing factors through the literature review, the problem identified as a result and the potential solution that was judged to be feasible within the parameters of the study.

The factors explored in the earlier chapters included the diversity of d/Deaf peoples with respect to identity, language fluency and preferences; the widely dispersed population and low incidence within the general population; the conditions that vary between visual-spatial and phonic (Derrida, 1976) languages with respect to modality's effect on the semantic resources of the language; the barriers to participation in feedback and the limited availability of adaptations to service access other than interpreters.

Health professionals and researchers involved in health services research and development have identified the need to engage more effectively with d/Deaf people. The factors summarised above point to four aspects of the problem with facilitating such engagement. First, that despite the flexibility and reach of online written surveys to overcome geographical barriers, these methods of data collection will not work for d/Deaf people, whose English is not necessarily fluent (Young and Hunt, 2011 p8). Secondly, Deaf BSL users have rights to information provision and access in their own language (NHS England, 2015a) and meeting these rights requires adaptations of methods in order to fit the visual requirements of a language with non-written form (Rogers et al., 2016). Thirdly, if those leading on such data collection are seeking to understand linguistic and cultural preferences, then responses in the participants' preferred language are more likely to allow cultural nuance and other pragmatic factors to be expressed (Young et al., 2016). Finally, although face-to-face methods for data collection e.g. interviews, may be preferred when seeking context-rich and detailed qualitative data, particularly in a visual-spatial language, wide geographical distribution of potential participants and the need for the interviewer to be fluent in the same language may be challenges to the use of such methods.

A potential solution to this problem is to develop a means by which Deaf people can autonomously participate in feedback and research through remote data collection that prioritises use of a signed language, but still gives the option to use the majority spoken/written language instead or as well. As previously discussed in the Introduction and Chapter 1, this is because d/Deaf people display a wide range of bilingual skills in signed and spoken/written languages and will utilise these in making sense of new information and gaining understanding of the unfamiliar (Young et al., 2016). The autonomous participation element is of importance because of the extent to which Deaf people's access to services, self-help and assessment is conventionally mediated through a third party e.g. a sign language interpreter, reducing their experiences of independent participation (Napier et al., 2017). Such a means of participation could be provided through the development of an online tool that works within existing hardware and software platforms. The stated purpose of the study was therefore to develop such a tool as one means of facilitating better engagement for d/Deaf people with service development and research.

Prior to stating the specific aims, the objectives linked to them, and elucidating the associated methods, I first review some fundamental issues of methodology relevant to the study.

4.3 Ontological and epistemological considerations

4.3.1 An ontology congruent with the areas of knowledge underpinning this research

Ontology is defined, within this thesis, as the nature of reality, but not divorced from the experience of that reality. The following brief overview of contrasting ontological perspectives conceptualises how much it is possible to know about the phenomena of interest in this research project and thereby identify an appropriate ontology that is congruent with the methods chosen, the aims of the study and the outcomes sought.

Streubert and Carpenter (1999 p3) highlight how difficult it can be to break away from the concept learnt in childhood that what is known is a fixed certainty rather than open to interpretation. The challenge as one grows up is to free one's thinking to consider alternative ways of knowing. This simplistic positivist way of looking at the world remains the basis of the majority of our day to day interactions with our surroundings and is pre-eminent in the natural sciences. Positivism can be summarised by the belief in an objective reality and relationships between material objects that can be understood and predicted by natural laws such as those discovered by Newton and Euclid (Van Langenhove, 1995 p18). They are not dependent on the enquirer; they exist regardless of being perceived. These relationships can be tested by experimentation that employs a 'logic of enquiry which can, most of the time, disregard the problematic relationship between perception and reality and function as if they had a direct access to an objective material world' (Murphy et al., 1998 p1).

Van Langenhove points out that the positivist approach used predominantly in the natural sciences is also the way in which people tend to experience and make sense of the everyday world (Van Langenhove, 1995). To explain this further and link it to the social sciences, he uses Harré's visualisation of phenomena in the material world as falling into three categories. These are 'objects of actual experience…[such as]…rocks, houses, people and the moon' which are in Realm 1, 'objects of possible experience…[such as]…micro-organisms and X-ray stars' which are in Realm 2 and can only be experienced with instruments such as telescopes and microscopes and 'objects beyond all possible experience [Realm 3]…which will never be experienced because inexperienceable in principle, for instance ensembles of quantum states prior to acts of measurements' (1995 p19). He argues that the developing disciplines within social science initially defaulted to the same positivist approach when considering psychological and social dimensions, placing behaviour in Realm 1, attitudes that can only be measured with the help of 'psychological instruments' like tests in Realm 2 and mind and self as unobservable phenomena in Realm 3.

This delineation of phenomena within the material and psychological worlds highlights the limitations of positivist science to answer all questions. In particular, it poses the question of how one can know that there is an objective reality to all phenomena when some fall into Realm 3 and are unobservable by definition. Nevertheless, one perspective is a belief that there is an objective reality to the world and that all efforts should be directed towards reaching an understanding of this reality (Murphy et al., 1998 p4). These strong realists believe that scientific explanations are literally true. At the other end of the spectrum is the radical constructivists' perspective that reality is what the individual believes it to be (ibid). The extent to which experience is required as well as belief is contested.

Subtle realists believe that current scientific understanding is used to get as close to objective reality as possible, but that much 'reality' is contingent on the observer's perspective, particularly in the psychosocial domains (ibid). In addition, subtle realists recognise that 'material reality can itself be a constraint on the possibility of definition. We can only perceive the world in ways which are in some sense consistent with the immanent organisation of that world' (Murphy et al., 1998 p4). For example, a subtle realist would accept that we are only able to see the world because of the objective existence of our eyes and the neural connections to the brain. The argument that scientific knowledge is developed from unbiased observation can therefore also be countered from a subtle realist's position. The methods chosen by a scientist are based on their background experience: Warburton (1994) argues that it is impossible to be completely unbiased because all observation is based on knowledge of previous theory so the observation will vary depending on the observer. As he says: 'What you see usually depends on what you know, and the words you choose to describe what you see always presuppose a theory of the nature of the thing you see. These are two inescapable facts about the nature of observation which undermine the notion of objective, unprejudiced, neutral observation' (ibid p113). An appreciation of the inevitability of subjectivity can therefore be of value to all researchers, both positivists and interpretivists. Warburton agrees: ‘Even though philosophy does not necessarily affect the way [positivist] scientists work, it can certainly change the way they understand their work’ (Warburton, 1994 p123). However, a social scientist might argue that subtle realism encompasses a wider range of factors that affect the means by which reality is known and, therefore, potentially the nature of reality itself. As well as prior knowledge and the ‘material reality’ of observation (Murphy et al., 1998 p4), these other factors could include the experience and social positioning of the observer.

There are objective realities to deafness such as measurable differences in the extent to which one might have the facility of hearing. The same is true of health service provision, comprising components that can be described in objective terms. However, as Bauman and Murray (2009; 2014) in their exposition of Deaf Gain point out, these are but some components of what are more appropriately described as multiple realities or multiple positions on the nature of reality. By contrast, a measureable reality from a Deaf person’s perspective might be the extent of fluency in sign language that a hearing person might possess. The point is not that multiple realities are measureable, but rather which are given pre-eminence or predominance (Young and Temple, 2014b Chapter 2). A radical constructivist ontology, by contrast, would not accept or necessarily incorporate the significance of the material reality within its purview, although an increasing number of Deaf scholars advocate for this position in research that concerns Deaf signing peoples (Kusters et al., 2017). As this study concerns deafness, Deaf people, health services, and language in interaction, it is also acknowledged that the fact that an individual’s experience of all these areas is highly subjective means also that a positivist approach is too simplistic. Consequently, a position of subtle realism is most appropriate, recognising as it does both the objective and subjective components of the contributing areas of knowledge and, by extension, the area of overlap between them that is addressed in this study.

4.3.2 Identifying an epistemology that provides insight into the orientation of the research

Having considered the possible realities with respect to these phenomena, the next consideration is how they can be known and how knowledge can be (co)created within this project: the possible epistemological positions. If it is accepted that there are multiple realities with respect to a phenomenon, then it is important to facilitate ways in which those realities can be known. What we experience, objectively and subjectively, can be interpreted in myriad ways depending on our cognition and our prior experience, both individual and societal, and the realities of these interpretations are regarded as equally valid if one adopts an *interpretivist* position. Interpretivist epistemologies stress that our realities are not only reliant on our interpretations, but emerge through relational processes, including communication: between individuals and phenomena and within a society. An interpretivist epistemology is therefore consistent with a subtle realist ontology. This section elaborates further the relationships between an interpretivist stance and the focus of this study, including the definition of its aims and objectives.

The identified aspects of the practical problem highlight the underlying epistemological implications. Considering the implications of the problem in this way gives further impetus to the need for such a tool. Firstly, knowing that Deaf people have barriers to involvement in services and research identifies the need to uncover the perspectives of individuals whose contribution has previously been unrecognised because their engagement was not facilitated. Further, these individual perspectives may give insights to a problem that can provide information about the community to which the individual belongs. Considering a problem from this particular interpretivist angle: a *social constructivist* epistemology, recognises that focusing on individuals in isolation is not enough to explain other phenomena such as society and relationships, nor can individuals create their own understanding of the world without being influenced by the societal views that are evident at that time in history. Van Langenhove summarises it thus: ‘The primary social reality can be conceived as a species-wide and history-long network of people speaking to each other in an environment that is socially meaningful’ (1995 p21). Gergen (1999 p33) argues that a social constructivist orientation offers exciting possibilities in adding further dimensions to meaning: it allows a fuller exploration of why and how people see the world differently and why some ways of seeing and knowing are denied or remain unrecognised within dominant social discourse. It particularly allows a focus on minority communities and the validation of their differently-constructed realities.

If developing such a tool can unleash and recognise Deaf ways of knowing, then it is also imperative that the ‘voices’ of Deaf people are not distorted through the process of capture. This imperative relates to the need both to facilitate a means of autonomous contribution by a Deaf person and that the contribution can be given in a non-orthographic way.

One interpretative and social constructivist epistemological orientation is phenomenology (Husserl, 1970). A phenomenological lens is particularly helpful when considering the challenges of communicating within and across languages, and for the researcher to recognise her own preconceptions of the meaning of language and understand the user’s perspective. Particularly relevant are:

- the centrality of language to phenomenology and its close relationship with hermeneutics: ‘it is literally more correct to say that language speaks us rather than we speak it’ (Gadamer, 1989 p463)
- its integral concept of intentionality that makes it very appropriate for the exploration of psychosocial constructs
- the fact that it focuses on the individual’s experience of a phenomenon (*sense*) rather than an objective reality (*reference*) (Gergen, 1999), but recognises that the meaning of a concept is developed through dialogue with others
- the acknowledgement of the potential influences introduced by the researcher so they can be made explicit
- the ability of phenomenology to make visible the taken-for-granted assumptions of everyday life, including the assumption that language means the same to everyone

Given that language does not mean the same to everyone, a phenomenological orientation highlights the perspective of members of a minority culture, making it particularly suitable for work with BSL users whose culture is commonly not recognised and whose language is often, wrongly, seen as based on and secondary to spoken English. Finally, it can be seen that, in order to access the nuanced and complex information that can uncover difference and diversity and enable interpretation, the tool that is developed should be capable of capturing qualitative and narrative comment and not only numeric and quantitative data.

4.4 Research aims and objectives

The overarching research aims were:

1. To develop a means of Deaf people’s autonomous participation in data generation concerning access to and use of genetic information and genetic counselling.
2. Through that process, to engender a generative approach to the communication of concepts and identification of terminology in BSL in this field of interest.

There were four research objectives that operationalised the aims of the study and that, in turn, guided the design and methods. These were:

1. To identify the essential features of an online data capture tool that meets Deaf people’s requirements.
2. To develop the specification for that tool.

3. To explore how the currently available technologies (hardware and software) could be utilised to develop a video presentation and video capture system that incorporated these identified features.
4. To build, test and pilot the tool.

4.5 Methods associated with each objective

The methods associated with each objective are summarised in Table 1 and then, where necessary, described in further detail.

Objective	**Method**
1. To identify the essential features of an online data capture tool that meets Deaf people's requirements.	Review of the available literature about signed language users' engagement with the internet, the exploration and development of relevant technologies in the context of signed language use on the internet, and the ethical implications of working with signed language data collected in this way. The search terms and approach to reviewing the literature are specified below.
2. To develop the specification for that tool.	Development of a use case from which a specification could be derived that meets the requirements of that specific case, but also has generalizable functionality. This development is described in Chapter 5.
3. To explore how the currently available technologies (hardware and software) could be utilised to develop a video presentation and video capture system that incorporates the specified features.	Consultation with software engineers and developers supplemented by available literature. This iterative process of consultation and trial forms a large part of Chapter 5's results.
4. To build, test and pilot the tool.	Software adaptation resulting in proof of concept. The build process is described in Chapter 5, followed by details of the in-house testing and external piloting.

Table 1: Summary of the methods associated with the four study objectives

4.5.1 Objective 1: Review of the available literature

The literature search strategy followed a similar process to that described in Chapters 1 to 3. The literature was found within a wide range of disciplines, with much published in information technology journals. There was a large amount of grey literature, particularly conference reports and abstracts. The search terms included *online*, *visual data capture*, *communication*, *deaf*, *ethics*, *ethical*, *British Sign Language*, *Sign Language(s)*, and *signed language(s)*. Iterative searching yielded the greatest numbers and most relevant material: both reviewing reference lists to search retrospectively and using the newer capabilities of Primo Central (University of Manchester, 2015; Ex Libris Group, 2015) and individual journal websites to prospectively follow newer literature citing papers already reviewed. Primo Central is 'a cross-disciplinary resource that details millions of e-resources, such as journal articles, e-books, and digital collections from sources such as Web of Knowledge, JSTOR, MEDLINE, ScienceDirect and publishers such as Elsevier, Springer and Wiley' (University of Manchester, 2015). It is accessed through the University's single *Library Search* function and therefore captures all material categorised by the leading databases, equivalent to if they had been searched individually. No limit was put on dates, but given the nature of the subject material, the earliest publications captured only began in the mid-1990s (the earliest was 1993). The appraisal of the literature also followed the same approach as the literature review chapters. Recognising the particularly swift changes in information technology, newer publications in the empirical literature were prioritised unless the paper discussed principles rather than specifics. Only literature available in English was included and unpublished dissertations were excluded.

4.5.2 Objective 2: Development of a use case

To bring the identified essential features together within a working tool requires first a use, real or imagined, to which the tool can be put. The features and the intended use can then be used as the goal to guide the development of the structure and functionality of the tool. From this structure and functionality, the specification of the tool can then be derived so the technical development can begin. The formalisation of this process, with a real or imagined use of the end product at its centre, is the core of the use case approach developed by Jacobson since 1992 and most recently presented as Use-Case 2.0 (Jacobson et al., 2011). The use case is formalised story-telling to describe how an IT system will be used to deliver a particular goal and illustrate its outcomes and value.

As well as identifying the lack of opportunities for Deaf people to engage with services and research, Chapters 1, 2 and 3 explored an area little understood: terminology and communication about genetics and genetic counselling in BSL. For the purposes of the study reported here, this subject provides an ideal use case. Such a use case forms the framework within which the features of the tool (outcome of Objective 1) can be realised and demonstrated.

The starting point for development of the content of the use case was previous work carried out in the context of genetic counselling information provision (The London IDEAS Genetics Knowledge Park and The Design Laboratory, 2005; Belk, 2006). The consultation with Deaf and hearing people that had taken place during both these previous projects focused attention on a number of factors, both around the context within which information was structured and presented and the ways in which data collection should be facilitated. These are explained and built upon in Chapter 5.

I had support from and consultation with two other sources throughout the development. The first was my ‘Deaf mentor’: Valerie Leach, a retired social worker who is Deaf herself with a wide and deep knowledge of Deaf culture and language through both personal and professional experience. The second was my advisory group for the project, comprising a mixture of Deaf and hearing professionals and lay people (see Appendix 2). Consultation with and feedback from Mrs Leach and other Deaf members of the study advisory group were particularly valuable at this stage of development.

4.5.3 Objective 3: IT consultation process and building of development team

The IT experts were identified through a discussion chain that started with an approach to the web team within the Faculty of Medical and Human Sciences University of Manchester. Their primary responsibility was webpage development to provide online presence for faculty research groups and teaching and to support software that had faculty-wide application. They did not have substantial staff resources to support research for individual projects using online methods and therefore predominantly provided a primary point of contact for this study and introductions to other IT professionals within the University. The discussion chain description is integrated in Chapter 5 within the description of the options that were explored and trialled.

4.5.4 Objective 4: To build, test and pilot the tool

A full description of this process forms, in part, the results presented in Chapter 5.

4.6 Statement on ethics

This thesis reports the development of a tool from identification of essential features, through specification, build, feasibility testing and piloting. All consultation and piloting was with individuals who were either staff members within the University of Manchester working on the project, members of the study advisory group, individuals external to the University employed to work on the project, and/or colleagues or personal contacts, both Deaf and hearing, who gave feedback at the pilot stage.

No ethical permission was therefore required at this proof of concept stage. Of course, if work were extended to involve individual participants then ethical permission would be required and sought. Nevertheless, an important aspect of the development was to consider the ethical implications of working with this type of data and this method of data collection.

4.7 A reflexive note

Reflexivity is the conscious recognition of how the researcher's background impacts on research methodological choices and process (Finlay, 2003). There are several features of who I am which were immediately obvious as relevant to how my approach to the study may be influenced, how I perceive myself, and how others perceive me and the process and products of my work. I am hearing and have lived within the dominant hearing culture all my life, though I have had Deaf friends and colleagues for twenty years. I have been a counsellor for 25 years which has developed my natural tendency to respect others' perspectives and continually question my preconceptions as a core part of that role. I have a strong personal commitment to diversity. My pathway to my orientation towards this research is as someone who trained first in a highly positivist subject (genetics), who then moved to become a genetic counsellor as a result of wanting to work within the communication and personalisation of this information for individuals. In this role, I have extensive experience of working cross-culturally and cross-linguistically with deaf people, Deaf people who are BSL users and people who use other minority spoken languages. My perspective and approach prior to this research was influenced, I believe, by this background as a geneticist with a liking for positivist facts, but I have challenged myself to remain constantly aware of that tendency in order to remain open to constructed realities understood through qualitative analytical approaches.

In terms of my strengths and weaknesses and their impact on methodological decisions, I am not Deaf nor a native BSL user, but I do understand genetic terminology in English well, am experienced in communicating about genetics in BSL and came to the project with extensive practical understanding of the difficulties experienced by both d/Deaf and hearing people in understanding complex information. Indeed, it was this understanding and recognition that was the impetus to develop this study.

I recognise that it is not only my self-awareness, but the attributions that I am given by members of the Deaf community that can influence the study. My positionality has some power as an NHS professional, with the resulting risk of being viewed as the expert rather than a seeker of others' perspectives and knowledge. I also acknowledge my own discomfort as a hearing researcher and genetic counsellor working in this field: the former because I fear others may see me as an interloper in a field where there is debate about whether hearing researchers have a right to contribute (Sutton-Spence and West, 2011; O'Brien and Emery, 2014; Kusters et al., 2017) and the latter because of the controversial nature of genetics for many in the community and therefore my expectation that my aims and motives may be questioned by some. This understanding of myself as a researcher external to the community is important. The personal attributes mentioned here are those which I judge have the greatest influence on my orientation towards the research. There are likely to be others including my gender, age, sexuality, personality traits and patterns of working and I have aimed to remain open to learning about these influences as an integral part of my development as a researcher.

4.8 Conclusion

This short chapter has identified the problem which the study sets out to address and considered an ontological and epistemological orientation towards it which recognises and values diversity in engagement and communication, specifically focusing on the perspective of Deaf BSL users. It has then operationalised the challenge through stating the aims and objectives, has set out the methods and my positioning as the researcher within a study which considers the needs and preferences of a minority group to which I am an outsider. The next chapter now moves to the detailed process by which the tool was developed.

Chapter 5: Results

5.1 Introduction

The results presented in this chapter are divided into four sections corresponding to the four research objectives. The sections therefore comprise:

1. The detailed identification of an online data capture tool's essential features meeting Deaf people's requirements based on the reviewed literature [Objective 1]
2. The incorporation of these features into a use case setting out a specification ready to be operationalized [Objective 2]
3. The exploration into the feasibility of the different technologies available at that time to realise such a video presentation and video capture system incorporating the identified features [Objective 3]
4. The development of the tool, with the chosen technologies, through an iterative process of building, internal testing by the developers (myself, the IT specialists and other contributors), and external piloting with a small group of individuals. [Objective 4]

It is important to note that the iterative development of the tool meant that, in practice, I moved back and forth between the stages. For the purposes of this thesis, the objectives are presented in a linear fashion. To locate these results in time, the initial scoping discussions reported here took place in Summer 2008, with the detailed development taking place between February and November 2009. Given the pace of change in hardware and software development, it is acknowledged at the start that some of the technical details presented below will now be redundant and have been replaced by newer solutions. It is contended, however, that the *principles* and *identified features* underpinning the tool itself remain of significance regardless of subsequent developments in digital and online environments because they are bespoke solutions in a highly specific context – that of bilingual and bimodal language use online. This contention is further examined in Chapter 6: Discussion.

5.2 Objective 1: Identify the essential features

5.2.1 Introduction

The method used to identify the essential features of an online data capture tool that would meet Deaf people's requirements was a literature review (Section 4.5.1). The essential features would need to meet identified challenges and opportunities related to online data capture, Deaf people and signed languages. The literature could also shed light on solutions that are being explored. The review therefore focused on three areas:

1. What is known about the engagement of sign language users with the internet.

2. How relevant technologies have been explored and/or developed in the context of signed language use online.
3. What has been considered in relation to ethical issues which are unique or similar when data is collected in a signed language and/or online.
The review is predominantly focused on the context at the time this online tool was being developed as that explains the choices that were made in tackling the subsequent objectives. However, where subsequent and contemporary developments are now changing the landscape and available options, this is indicated.

5.2.2 Engagement of signed language users with the internet

5.2.2.1 Internet structure and navigation remains text-based, although other media formats are increasingly available online

Published research into the use of computer technology with D/deaf people can be grouped into several distinct areas. One area addresses accessibility or lack of accessibility to the internet. Some authors have argued that the internet enables more equal access to information and education for deaf people by overcoming geographical barriers for those living in isolated communities with few deaf peers (Belcastro, 2004) or by the fact that the internet is a leveller between deaf and hearing people because language becomes visual i.e. written rather than aural (Barak and Sadovsky, 2008). However, the internet is still structured as a text-based medium and Deaf people have the recognised barrier of a lower average literacy level for English compared to hearing peers (Mayer, 2007).

Some authors have looked specifically at the impact of lower literacy levels on internet use within deaf populations. For example, Smith (2006) observed 22 Deaf adolescents as they used Google to attempt to find answers online to questions presented to them in ASL and/or Signed English. He questioned them one-to-one as to why they had selected particular search terms and chosen particular webpages, concluding that they struggled to find efficient search terms and filter returned information that was predominantly available only in English above the average reading level. Zazove and colleagues (2004) gathered demographic information and asked questions about computer use through a two page paper questionnaire within a larger study investigating information provision on cancer prevention behaviours to deaf people. They reported this data separately to their main study, finding that two-thirds of their 227 participants, all aged 18 years and older, used computers, but that computer use was statistically significantly associated with participants' English use and Signed English use in a number of settings e.g. at home. Both these studies support the supposition that good written English facilitates internet use, whilst lower literacy is a barrier to engagement and to finding specific information. In contrast to Zazove et al (2004), Bowe (2002) found that almost all of his 884 Deaf and hard of hearing respondents used email and instant messaging from home in the United States and a quarter (a significant proportion for 15 years ago) had broadband internet. However, his study was conducted online so there was a significant potential biasing factor in the recruitment.

Many articles were published in the first decade of the millennium, leading up to this study, that considered online hosting of other types of media, particularly for general education (rather than specifically for d/Deaf users), and the use of software such as Camtasia to screen-capture lecture presentations for later display on the internet (Joukov and Chiueh, 2003; Kameda et al., 2003; Blezu and Popa, 2008; Dickson et al., 2008; Malanik et al., 2008; Ramani and Sirigiri, 2008; Conlon and Pavlika, 2009). This literature predominantly considered the use of this technology to tackle geographical challenges in the delivery of information rather than in the context of accessible information provision for d/Deaf people.

Since the development stage of this study, there have clearly been great developments in increased broadband speeds and the fourth generation of mobile networks (4G) with 5G on the horizon. These changes have facilitated the move from desk-based computers to mobile technology, have brought down the cost for an individual of connecting to the internet and allowed widespread use of multimedia in one-way and two-way communication. The potential implications of these developments are discussed further in Chapter 6: Discussion, but not addressed here, given the development of this tool pre-empted these changes.

Despite great strides in technical developments that, serendipitously, have enabled greater use of signed languages online, there remains a reliance on text for movement around the internet: even when signed language videos are embedded in a webpage, it is usually still necessary to understand a written language in order to navigate between webpages and search effectively for relevant content. Prior to and contemporary to this study, a couple of groups have explored a particular aspect of internet use by Deaf people with the specific aim of improving access for those who experience the world visually and use a visual-spatial language. For example, Fajardo and colleagues have studied how pictorial icons substituted for text (Fajardo et al., 2006) and signed language instructions embedded as videos (Fajardo et al., 2010) affect accuracy and speed of internet navigation. This is something our research group (Social Research with Deaf People) has also explored recently: the option of using GIFs (Graphics

Interchange Format which can present as short videos) or photos as hyperlinks to navigate around our webpages. However, this is not straightforward when working within an institution which is primarily geared to work in English and other languages that can be represented orthographically. Standards of accessibility for the creation of web-based content, whether within higher education or other public bodies, also typically do not address the issues of visual navigation for sign language users. Considering a separate aspect, that of clarity of signed language within an online video, Muir and Richardson studied eye gaze in Deaf participants whilst they watched presentations in American Sign Language (Muir and Richardson, 2005). Their aim was to determine which areas of the body and therefore the video screen were the most important in understanding the information. By so doing, they set out proposals for which areas of the video screen needed to be higher resolution and, by limiting higher resolution to those areas, reduced the data volume that would be needed and allowed more of the capacity to be directed towards video speed – an equally important component in the transmission of information in a signed language. Development and research directly about signed language use online was limited to these few examples at the time of the technical development of this study.

5.2.2.2 Bilingual and bimodal presentation and response through online platforms

At the time this tool was built, only a small number of published studies had used computer-based signed language questionnaires. An early example was Lipton and colleagues (1996) using an innovative videodisc reader system to present signed information to their participants. Berman and colleagues (2000) developed a computer-based questionnaire in American Sign Language (ASL), English and Signed English (ASL signs used in English word order) to investigate tobacco use in young Deaf people. More recently, Gerich and Lehner (2006) have presented several health assessment measures (the World Health Organisation Quality of Life inventory, the Brief Symptom Inventory and the General Health Questionnaire) in Austrian Sign Language. Both these latter studies accessed the questionnaire video clips and software on a standalone computer rather than via the internet. The questionnaires were quantitative and asked for responses via click buttons onscreen. Since then, the major development has been the wider availability of broadband internet connections, allowing the option of placing online questionnaires that include videos. Studies shortly following this project have been exploiting online questionnaires for the first time, including reliability and validity testing of a number of psychometric tools following translation to BSL (Rogers et al., 2013a; Rogers et al., 2014; Rogers et al., 2016). All these studies have presented their information and questions predominantly or entirely in a signed language, are asynchronous, and required online participation via a website by click button responses.

Chapter 1 discussed that Deaf people vary greatly in their communication history and preferences and many are bilingual in English to a greater or lesser extent. Recognising this fact, some published work exploring signed languages online chose to give the option of accessing the content in the national written language as well as in the signed language, but prioritised presenting the information separately in the two languages, rather than alongside each other (Berman et al., 2000). Other studies were specifically validating a BSL version of a psychometric tool so it was essential that only BSL was used during the online data collection. However, given the wide variation in communication preferences, it is well known anecdotally that, in a complex subject area, Deaf people may wish to refer to the English alongside BSL in order to make use of both of their languages in understanding concepts. At the time of the study, there was little published work to support this anecdotal knowledge. It was supported, however, by a recent study (Young et al., 2016) where focus group participants discussed the meaning of concepts such as ‘informed consent’ using their knowledge of the English lexical items ‘inform’, ‘consent’ and ‘informed consent’ as well as the semantic morphemes of the BSL lexical item. Bilingual presentation can also potentially address some of the ‘unique problems’ of signed language videos when compared to English text (Zazove et al., 2004) such as not being able to show questions and answers simultaneously, longer completion times for surveys and difficulties in translation of certain lexical items (Rogers et al., 2013b). Recognition of the latter issue was, of course, the primary focus of Chapter 3.

5.2.3 Exploration and development of online technologies of particular relevance to signed languages

Developments that make feasible an online solution to signed language data capture can be separated into technological and social. Broadband internet connections are now reliably fast enough, in many settings, to allow streaming in real time of video files, both for download and upload. Affordable webcams are available with fast enough frame speeds and resolution to use with signed languages. Data storage has increased to a level where large video files are manageable within a university central server. Software and programming languages are capable of creating questionnaires, building websites without recourse to first programming principles, capturing streamed video data, hosting videos and editing digital video.

From a social perspective, these technological advances have facilitated the massive growth of social networking with a widespread use of webcams, familiarity with blogging and posting of video clips online, particularly through user-generated content sites e.g. Youtube. Members of the Deaf community now use these tools for visual communication using signed languages through instant messaging and internet-based videophone connections using software such as FaceTime, Skype, vlogging (video blogging) as well as written English on emails and SMS texting. This increasing familiarity with internet-based communication means that, while the data collection tool to be developed was new, it brought together existing technologies and therefore would not be entirely alien to potential users.

I was concerned that a technologically complex interaction may be off-putting to some people. However, it may attract others because of its novel approach. Young Deaf people, in particular, have been enthusiastic adopters of internet technology because of the potential to supersede the restrictions of communication using telephone lines (Swinbourne, 2016). Since this study was conducted, there has been a sea-change in the way in which people engage with computers, with the majority shifting from accessing the internet using desktop and laptop computers to using smartphones and other wearable technology. Coupled with the ability to stream online content via 4G as well as wifi, this allows individuals to be online, with increasingly fast and robust broadband, at a place and time of their choice.

5.2.4 Ethical implications of online collection of signed language data

When considering collection of data in a signed language, there are, firstly, the ethical priorities common to any data collection. There are the same principles requiring security of data collection and storage, informed consent to be established with study participants, confidentiality and anonymity. It was envisaged, however, that there would be differences in how these requirements should or could be addressed. At the time of developing the tool, there was little literature considering the ethical differences in working with a language with a visual modality. There was literature from an ethnographic perspective which discussed the ethics of working with and alongside minority cultures, rather from an outside perspective, to identify research priorities and make visible their culture. Notably, Pollard (1992b) discussed the ethical requirement to consider the whole minority community and not only individual members in issues of consent. Since this work has been undertaken, there have been a range of additional texts discussing ethical issues in data collection with and amongst Deaf people who use signed languages: examples include Harris et al. (2009), Young and Temple (2014c) and O’Brien (2017).

There was no literature available prior to the development of the tool that considered how practical differences with signed language data impact on ethical imperatives. There were some guidelines available that discussed the ethics of visual methods (Prosser et al., 2008; Wiles et al., 2008), an area of methodology developing in parallel with data in signed languages, and also at a much earlier stage of development a decade ago. These guidelines focused predominantly on image and video data and signed languages were not mentioned. They were still valuable and relevant in their consideration of anonymity and confidentiality, highlighting the difficulties in balancing anonymity for participants against loss of data in terms of nuance communicated through body language and visual contextual information. In addition, the point was made that some participants may actively wish to remain visible in the presentation of the research, and this may be even more the case when the individual is a member of a minority population who are less often visible in the broader sense. This last issue was subsequently discussed in greater depth and related specifically to sign language-using populations (Young and Temple, 2014c).

As a research group, we were already discussing the challenges of how to present BSL data on video in results and dissemination. It was clear that not only additional nuanced information is lost when attempting to anonymise a BSL user e.g. by obscuring their face, but the fundamental semantic meaning given by facial expression. This solution could not be acceptable, potentially shifting the debate from the necessity to anonymise to the necessity for improved confidentiality and informed consent about where and how the data would be shared. The first paper that clearly debated these issues was published a year after the tool was developed: Crasborn (2010) highlighted the impossibility of anonymising the video content of the Sign Language of the Netherlands (Nederlandse Gebarentaal or NGT) corpus and discussed the difficulty of consenting participants to making the corpus publically available online when it could not be predicted how the data could be used or individuals identified e.g. through facial recognition software, in the future. This debate is, of course, also relevant to how data collected through the online tool developed in this study might be (re)presented in the results, but there was an additional dimension for this study, in the consideration of security and confidentiality during the internet-enabled data collection itself.

Online data in a written language has the advantage that it can be anonymous to the researcher at the point of collection, unless a participant chooses to give identifiable information about him or herself. This is not the case with video data of a signed language. Anonymity could not, therefore, be an obligatory condition at the point of data collection; rather, an approach was required that actively specified who would see the potential participant in viewing the data (typically the researcher and their supervisor) i.e. the limits of confidentiality would need to be addressed explicitly with participants as part of the consent process.

The remaining consideration related to confidentiality and non-anonymised video data is to ensure an extremely secure method of data capture. Again, no published literature could be found at that time that discussed security of online video data capture and its implications for research. This was therefore an area that would have to be addressed by consultation with the IT team during Objective 3.

Since the development of the tool, the most comprehensive discussion about Deaf people, signed languages and research ethics has been Young and Temple's chapter on ethical research practice (2014c).

5.2.5 Distillation of required features of the tool from the literature reviewed

In summary, the literature reviewed supported the following factors to be considered during the development of the tool:

1. Deaf BSL users are geographically widely dispersed.
2. Deaf BSL users, by definition, need to be able to watch information in BSL and respond in BSL, as their first or preferred language.
3. Broadband connections are now fast enough within the UK, in the majority of situations, to stream videos, as both downloads and uploads, fast enough for signed languages to be comprehensible. However, the video resolution will affect the data volume and therefore the bandwidth required.
4. Deaf BSL users, on average, have a lower literacy level for written English.
5. Deaf BSL users, as a group, have enthusiastically adopted online means of communicating in BSL through posted videos and video communication software. However, the orthographic structure of the internet causes a barrier for some individuals to find accessible websites in the first place.
6. The majority of Deaf BSL users are bilingual in BSL and written and/or spoken English to some extent. They may therefore use both languages to make sense of presented information, especially in subject areas that are complex, fast-changing and little discussed in BSL. They may prefer to respond partly or completely in English as well.
7. Data in BSL cannot be anonymised with current technologies without losing essential semantic information from the communication.

Taking these factors above into account, the tool would need to incorporate the following features:

1. Presentation of content bilingually and bimodally i.e. mirrored content in BSL on video and written English (although whether this should be presented sequentially or simultaneously was not resolved).
2. Deliver streaming video in BSL where the resolution and frame rate is high enough for the content to be seen clearly.
3. Deliver video that will stream fast enough over the majority of broadband connections.
4. The option to respond in BSL and/or spoken English and/or written English (although whether a requirement to be consistent in language and modality choices throughout was not resolved).
5. Ensure that upload of streamed video data captured is also of suitable resolution, frame rate and speed.
6. A reliance on written English instructions alone to navigate the data collection tool and within the information content should be avoided: instructions should be available in BSL as well or primarily.
7. Participant access to the tool should be:
    a. Confidential: no digital trace left on the computer used;
    b. Flexible: accessible at a time and place convenient to the participant;
    c. Easy: the technical steps required to watch and respond using the tool should be minimised and consistent with usual technology and device use in everyday life.
8. Data should remain confidential during its capture and only accessible to essential members of the research team during storage and those people should be specified.

5.3 Objective 2: Develop the specification for the tool

A use case approach (Dunstan Thomas, 2017) was adopted to meet Objective 2, as has been used in several published health informatics development projects (Löffler et al., 2010; Zampognaro et al., 2017). The use case provides the framework within which the features of the tool (Objective 1 outcomes) can be realised and demonstrated. The use case requires aims and objectives, structure and content which the data presentation and collection tool can then be built to deliver.

5.3.1 Developing a use case

The choice of use case responds to the two overarching aims of the research design (see Chapter 4 Section 4.4), namely that the finished tool should (a) facilitate ‘Deaf people’s autonomous participation in data generation’ and (b) that it should ‘engender a generative approach’. These aims also set out the imperative that the tool should enable qualitative and narrative data to be gathered if nuanced insight into culture and language is going to be possible. The functionality of this first iteration of the tool was conceived of as possibly also including the ability to collect a range of other types of data, including numeric and quantitative. However, the greatest challenge was identified as developing a capacity to collect data that captures highly personal perspectives and is as little influenced as possible by the practical framework of the data capture. In other words, the means of data capture must preserve, as far as possible, a participant’s natural and preferred communication, without them modifying their language to meet technological restrictions of data collection method.

5.3.1.1 Questionnaire as structure

A second source of potential restriction was the linguistic capabilities of the recipient of the online data capture. My intention was to design a use case that, in the first instance, would address the issue of online data generation and capture without the communication and language fluency of the researcher being a dependent variable, given it was the functionality of the technology and tool that was the focus. Therefore, I opted for an asynchronous approach to data generation and capture as the first step and built it around a questionnaire structure. I decided to incorporate a pre-recorded guided interview in BSL on the website, presented on video by a Deaf narrator and with the written English translation alongside. In effect, this was a qualitative questionnaire where the questions replaced the prompts that would be used during a semi-structured synchronous interview. The tenor of the questions posed, nonetheless, would encourage participant reflection and allow open-ended consideration.

5.3.1.2 Focus of use case

The content and focus of the use case was developed from previous work on genetic counselling information resources in BSL (The London IDEAS Genetics Knowledge Park and The Design Laboratory, 2005; Belk, 2006). The consultation with Deaf people that had taken place during both these previous projects focused attention on a number of factors, both around the context within which information was structured and presented and the ways in which feedback should be facilitated. The use case would be built around key questions that emerged from this work about what mediated Deaf people’s access to and understanding of genetic information. These questions were:

1. What are the advantages/disadvantages of information about genetics and genetic counselling delivered online and in these formats?

2. To what extent are any barriers to understanding associated with the linguistic content and to what extent are other barriers identifiable?
3. How could information delivered in BSL be further designed/enhanced to maximise understanding of medical and genetic counselling information?
4. For Deaf lay people, what are the effects of additional conditions (e.g. receiving information indirectly through an interpreter or through a secondary medium such as a DVD rather than face-to-face) on the development of understanding of information and accessibility of service provision?

In terms of the objectives associated with these questions, they are:
1. To collect and, where necessary, develop information resources to present genetic concepts and terminology with varied contexts, content and formats.
2. To facilitate the collection of open-ended qualitative data asking questions about preferences for terminology, content and format, understanding of terminology and concepts, and factors affecting communication and understanding.

5.3.1.3 Structure of the materials used as the means of data generation

The materials presented showed contrasting means of acquiring knowledge about genetic counselling and contrasting scenarios of being involved as a deaf person within a genetic counselling situation. Data would be collected by participants responding to questions, posed in BSL, that were linked to the alternative presentations and required reflective responses. The range of content, formats and structures included:

- Live acted scenarios showing direct communication in fluent BSL, BSL as a second language, and communication between BSL and English using an interpreter
- Direct presentation of information to camera in BSL
- Information leaflets in written English
- Animation
- Fundamental genetic concepts e.g. genes, chromosomes
- Modes of inheritance e.g. recessive, dominant
- Examples of genetic, or possibly genetic, traits e.g. cystic fibrosis, deafness, tongue-rolling
- Illustration of genetic counselling consultations

5.3.1.4 Structure of the questions posed to elicit autonomous responses (data)

The outline structure of the questionnaire is presented in Figure 1 overleaf for reference during the explanation of the individual sections that follows.

Many of the design considerations for the questionnaire were common to those in any language. For example, the questions needed to be tailored to address the aims of the study, to be accessible and understandable in order to encourage respondents to cooperate, and to elicit responses that are accurate (Robson, 2002 p242). Further considerations for a topic as emotionally and technically complex as genetics included:

- Questions that would elucidate in-depth information about personal understanding and preferences;
- Questions that were not unnecessarily intrusive in the amount of personal information requested while still assessing participants' self-identity sufficiently;
- A logical structure that led participants through a complex subject and allowed them to build up an increasing knowledge rather than jump from one area to another;
- A structure which, given the complexity and length of the finished questionnaire, allowed participants the option of choosing a shorter version;
- Content that presented the terminology, as the subject of study, in a context that made it understandable and that could stand alone as future valuable information resources.

The final point above considers that abstract concepts that could not be directly experienced, such as genetic inheritance patterns and definitions of genes and chromosomes, could be engaged with through anchoring them in a context to which an individual could relate, regardless of personal experience of that context. My initial ideas had included asking potential participants about previous experiences of communication in a medical setting, but I judged this may be too intrusive and personal, especially early in the data collection process and given it was online without them being able directly to engage with me as a researcher. Instead, I used a scenario about genetic counselling that related the concepts to questions being asked by the illustrative Deaf couple attending. The format of the questions included some fixed choice and Likert scale responses, though the majority of questions were open and would permit signed responses in BSL and/or a response in English. Where closed questions had an 'other – please tell us' option or an invitation to elaborate on the reasons for the choice made, the additional response would also permit responses in BSL and/or written or spoken English.

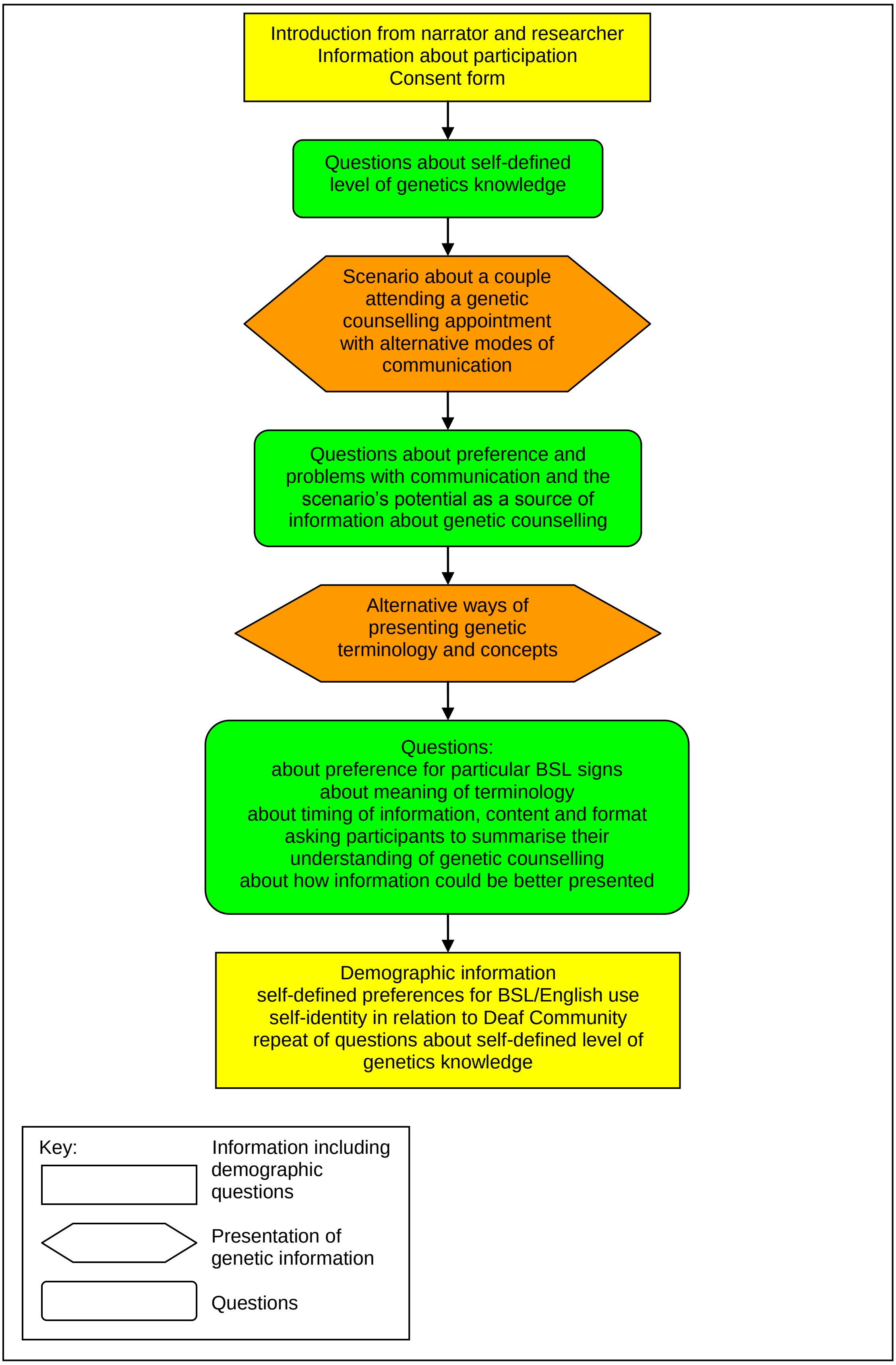

Figure 1: Summary of structure of questionnaire

5.3.1.5 Questionnaire content

The content of the scenario was based upon a scenario developed in a pilot collaborative service development project. The team working on this project included me, genetic counselling colleagues at Great Ormond Street, London, researchers at the London IDEAS Genetics Knowledge Park, animators at the Central St Martins College of Art and Design and a Deaf academic whose work centres around theory of mind in Deaf children, but whose professional and community experience is particularly around services for Deaf children and young people. The project followed on from a previous animation produced by the London-based partners to explain the genetic condition Fragile X to patients (The London IDEAS Genetics Knowledge Park and The Design Laboratory, 2005).

In the second section of the questionnaire, I used other embedded multimedia resources as follows:

- Information leaflets entitled ‘Recessive Inheritance’, ‘Dominant Inheritance’ and ‘What is Genetic Counselling’. The leaflets are in written English with diagrams and pictures and available in PDF format. They were developed by the North-West Regional Genetics Service (Department of Genomic Medicine, 2016)
- BSL translations of the above information leaflets (Genetics and genetic counselling translation team, 2005; Belk, 2006)
- Animated sequence to illustrate genes and chromosomes, used with permission of the team at Great Ormond Street/Central St Martin’s and extracted (removing the audio track) from their patient information animation about Fragile X syndrome (The London IDEAS Genetics Knowledge Park and The Design Laboratory, 2005)
- Description of genes and chromosomes taken from the family information booklet on genetic counselling published by the National Deaf Children’s Society (Belk, 2008)

The content taken from pre-existing resources had been stored on DVDs. To remove information from a DVD in a format compatible for online streaming, freeware called Handbrake (http://handbrake.fr/) was downloaded and used to rip data from the DVD.

The scenario storyboard was ready for production as an animated film, but our collaboration had been unsuccessful in securing further funding. With the permission of those involved, the scenario’s adaptation for use here meant that it was valuable to this study and the previous work put to good use. The storyboard was developed in strip cartoon format to maintain a focus on the visual, although a script was also recorded in written English.

Another key advantage of a scenario-based questionnaire was the flexibility it facilitated to explore different language conditions for the same story/context: one section could be repeated three times to illustrate three different modes of communication: hearing health professional working with third party BSL/English interpreter, Deaf health professional using BSL and hearing health professional using less than fluent BSL. Questions could therefore be asked about preferences around mode of communication and language in context.

Another design consideration was to avoid a focus predominantly on the genetics of deafness. Given the historical relationship between medical/genetic services and the Deaf community, I wished to illustrate that genetic counselling for Deaf people is not only about the cause of deafness. Rather, Deaf people have the same chance as anyone in the general population of having a family history of a genetic illness such as cystic fibrosis or inherited breast cancer. The service development project, from which the scenario had been taken that formed the basis of this questionnaire, had already chosen cystic fibrosis to illustrate recessive inheritance and I added a neutral trait that could be illustrated visually within the scenario (tongue-rolling) to illustrate dominant inheritance.

The scenario in BSL can be viewed at the following locations on Vimeo (password for all clips is: geneticcounsellinginBSL):
Genetic counselling in BSL - Part 1 ‘Recessive inheritance and different ways of communicating in clinic’ https://vimeo.com/67572696
Genetic counselling in BSL - Part 2 ‘Dominant inheritance’ https://vimeo.com/67572694
Genetic counselling in BSL - Part 3 ‘Genes and chromosomes’ https://vimeo.com/67572692.
The English translation of the scenario script is available through a link on page 10 of the legacy website[1].

5.3.1.6 BSL-English translation and consultation for cultural acceptability

The choice to present the questions and resources bilingually and bimodally meant a high quality translation was required. The interpreter I worked with to achieve this is a

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[1] A legacy version of the full questionnaire is available at this hyperlink: Genetic information in British Sign Language (legacy for demonstration) and URL: https://apps.mhs.manchester.ac.uk/surveys/TakeSurvey.aspx?SurveyID=n2LHno56. The legacy version does not have full functionality because of the change in technologies predominantly supported by the University since 2009-2010. For example, the embedded video capture tool will link to the webcam connected to a viewer’s computer, but is not currently linked to a streaming server so will not capture and play back a video clip. Some of the tables holding a number of the videos for playback are not currently functional, though they could be reactivated.

Deaf qualified BSL-English interpreter and translator registered with NRCPD (The National Registers of Communication Professionals working with Deaf and Deafblind People). The translations focused on pragmatic rather than textual equivalence in their preparation (Baker, 2011) and she and I collaborated at all stages of development. We worked from the prompt of the written English script for both the questionnaire and the scenario, but used the storyboard and her community and linguistic expertise to focus primarily on the BSL. She remained involved in discussions about how to represent the scenario throughout the filming. After filming of both the questionnaire and the scenario, she back-translated the BSL to English, meaning that there were subtle changes in the plain English script that would be made available online alongside the BSL.

A further principle was to work closely with Deaf colleagues to plan and execute the filming and production of the BSL resources with attention to the language and content. This approach was effectively a community participatory one, though my work took place prior to the majority of the increasing number of publications which have used and championed the benefits of such an approach, including work with Deaf communities (Pollard et al., 2009; Graybill et al., 2010; Barnett et al., 2011; Young et al., 2016). I recruited Deaf actors for the scenario production. It was important that their BSL was smooth and clear, that they were believable in their scenario role and that they would be recognised by Deaf people as being from their own culture. Following initial content and structure discussions with my supervisory team and project advisory group, the actors and narrator were fully involved in discussions about the translation and representation of specific concepts in BSL. Some of these discussions resulted in slight changes to the content for clarity and cultural acceptability.

For example, the scenario began with a couple attending for genetic counselling and volunteering near the start that the male partner’s brother had died from cystic fibrosis. The actors felt that this jarred so early in the session and questioned whether a couple would disclose that so early in their first meeting with the genetic counsellor. Although mine and my clinical supervisor’s professional experience would suggest that people do disclose very personal information early when in a safe setting and the information is of clinical relevance, I was prepared to accept the suggestion of changing the background to the brother being ill with cystic fibrosis rather than having died.

This section has addressed two key considerations: linguistic equivalence and cultural acceptability. A formal translation and back translation process as used by, for example, Rogers and colleagues (Rogers et al., 2016; Rogers et al., 2013b; Rogers et al., 2013a) was not followed because this questionnaire was not a standard instrument where the linguistic equivalence was vital to establish its psychometric properties and validity. Rather, this was an exploratory questionnaire. None the less, for Deaf people choosing to access in English, BSL or both, face validity was important: were the questions in each language asking the same thing and likely to elicit the same kinds of information? This was the basic question to be answered in piloting the linguistic equivalence aspects and was addressed in the pilot study, detailed towards the end of this chapter.

5.3.1.7 Developing the BSL content: the filming and editing process

There were two areas of new content to be developed and filmed in BSL. The first was the narration, comprising the explanation of the study to potential participants, the consent process and the questions. The professional translator discussed in the previous section filmed this in her business filming studio, edited it digitally and supplied the completed clips on DVDs. As well as the in-depth discussion already highlighted about content, terminology and cultural acceptability, there were other concerns relating specifically to the use of a visual-spatial language when embedded in a website e.g. placement of specific signs, and also technical aspects of the file format and the website itself. These issues are expanded upon in the next section.

The second area of content was the scenario for which four Deaf actors and one hearing BSL/English interpreter were recruited. I coordinated the filming and set up and operated the video cameras. The filming took place over two and a half non-consecutive days in two locations: an office setting made to look like a genetic counselling clinic and a home setting. I used three fixed cameras filming concurrently to capture camera shots face on to the actors (particularly important for clarity with a visual spatial language) as well as a wide angle shot capturing all actors. A substantial amount of time was spent on discussion and preparation during these days, partly to agree the clearest signs and ways of explaining concepts and partly to supply the actors (particularly the actor playing the Deaf genetic counsellor) with additional background genetic information and understanding.

I edited the digital video recordings produced during filming on a dedicated high-specification desktop computer using AVID digital editing software. This computer was situated in the Media Centre of the University of Manchester so I was able to draw on the expertise of the staff there to assist with technical aspects of the software use. The filming and editing both highlighted considerations that differed between an oral and a visual-spatial language and these are discussed further in section 5.5.1.

5.3.2 Specification

The requirements of the use case and its content were a means to develop the specification of the online tool, in terms of its technological functionality as well as functionality for its users, whether researcher or participant. Making an idea practical by ensuring it meets the requirements of a clearly delineated ‘use’ enabled this next step. The tool was required to have the following functionality. It should:

- Allow remote access by participants at a time of their choosing (asynchronous to the placing of the questionnaire/interview material online)
- Present the information and questions bilingually and bimodally (as BSL videos and in written English)
- Enable the presentation of contrasting materials, whether static or active (information, animation, filmed scenario content)
- Facilitate responses in BSL (as video clips) and/or spoken English (also using the video capture tool) and/or written language (expected to be English, but theoretically not confined only to that written language)
- Automatically store the captured responses securely on a University remote server for later access
- Enable the video clips to be either streamed and watched from the remote server or downloaded for purposes of analysis
- Leave no identifying information on the participant’s computer

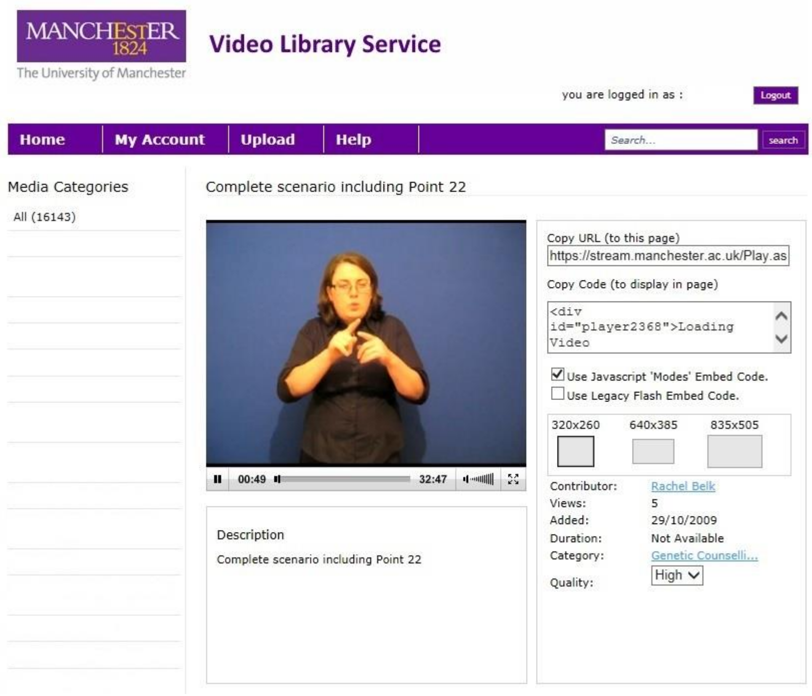
5.4 Objective 3: Explore the available technologies

My initial discussions with the Faculty of Medical and Human Sciences web development team in August 2008 concluded that a custom-built website would be needed and that one or more web development team members would develop this from scratch. This would present each question video clip alongside the English translation and allow participants to record their response in BSL or spoken English from their webcam or type a written language response. It was immediately clear that people from other IT specialist teams within the university would need to be consulted and involved. There were two main areas of development required, as follows.

5.4.1 Hosting video clips

It was possible that the questionnaire video clips could have been hosted on a departmental server and embedded in the website as needed. This would have required a significant amount of storage space. However, this hurdle was overcome easily because the university was, at that time, developing its own version of a user-generated content site. For ease of reference, the best known user-generated content site was then, and still is, YouTube. The university call their site the Video Library

Service (VLS https://stream.manchester.ac.uk). It stores video clips produced university-wide on a particular format of secure server (a ‘streaming server’) and allows controlled access to the clips for use in webpages. MP4 files were the preferred format as any other file formats would be converted to MP4 by the VLS in any case. The great advantage of this service development was that it is very user-friendly: the service automatically generates a Javascript embed code for an uploaded video which a relative novice can then easily copy and paste into a new webpage. Several years on, use of the VLS is now ubiquitous for hosting and streaming information videos and lecture recordings into University webpages. Figure 2, below, illustrates the appearance of one of the question videos when accessed through the VLS to set up options and embedding tools.



Complete scenario including Point 22

Copy URL (to this page)
https://stream.manchester.ac.uk/Play.as

Copy Code (to display in page)

```
<div
id="player2368">Loading
Video
```

☑ Use Javascript 'Modes' Embed Code.
☐ Use Legacy Flash Embed Code.

Contributor: Rachel Belk
Views: 5
Added: 29/10/2009
Duration: Not Available
Category: Genetic Counselli...
Quality: High

Description
Complete scenario including Point 22

Figure 2: Screenshot of questionnaire video clip within University of Manchester Video Library Service

5.4.2 Capturing video clips

The second major area of development required was to develop a means of capturing video from the participants. The challenges related to video data capture that had been identified could be grouped into three areas: ethical, data security and technical. The ethical challenges related to online data collection, generally and specific to this project, have been addressed in section 5.2.4. Some of the data security considerations are similar to those for other research projects dealing with visual data e.g. the location of data during storage and analysis. However, there were additional data security considerations specific to online collection addressed during the development of the tool: the security of data during its passage through the internet and at/after the point of capture. During these initial technical explorations, the underpinning principles were explored further: the requirements for individual access, security, asynchronous access and access independent of a facilitator.

Several possible means of data capture were initially discussed in August 2008 between myself and the University IT specialist with whom I was working. The first was to use Adobe Connect 7, desktop video-conferencing software that had recently been adopted by the university. This was effectively a virtual meeting room. Clarity of the video picture is limited only by specification of the webcam being used and the internet connection speed. The software already contained the function to record a ‘meeting’ and the appearance of the screen can be customised. This would mean that the screen could be set up with instructions as to how to respond to questions, background information and the questions themselves. A hyperlink could be emailed to potential participants that would allow them to access this pre-designated customised ‘meeting room’.

The advantage of this solution was that the software allows a meeting room to be accessed by all invited guests, both internal and external to the university, even though it is controlled internally: this would therefore be suitable for the lay participants sought outside the university. All that a potential participant would require was the private hyperlink to the meeting room, which could be emailed to them.

One major disadvantage to this system was that a moderator (in this case, myself) either needed to be present to enable and disable the recording or the recording had to be running continuously. The former would negate the advantage of a website allowing access at any time convenient to the participant and asynchronous to the placing of the questionnaire: the time of a meeting in Adobe Connect would have to be prearranged by email between the researcher and participant. The latter would mean that huge amounts of video-recorded data would be generated, taking up excessive storage space and significantly increasing the later work in identifying the participants’ responses within the video files.

Of greater concern ethically was that, if a single meeting room was set up for the study, there was no immediately obvious way of preventing more than one potential participant accessing the room at the same time, so making it impossible to guarantee anonymous participation. This was clearly unacceptable. The solution would be to set up a separate meeting room for each participant, but again, this would be unwieldy and only possible if I emailed a different link for a separate meeting room to each participant. This would similarly delay and limit a participant’s freedom of access to the questionnaire.

We briefly considered a second alternative: whether Skype online chat could be recorded remotely. Skype is freeware used for instant messaging and to make free video and audio calls via the internet. This idea was dismissed quickly as not being secure enough in how to control who could access the conversation. As with Adobe Connect, there was also the barrier of not being able to facilitate the recording without a prearranged time.

A further solution suggested by the IT specialist was for him to look at the potential of the VLS. His initial thoughts were that I could set up a private folder for clips generated by participants. This would entail participants using the software that came with their own webcam or other software such as Skype or MSN to record their responses. They would then need to access the user interface of the VLS to upload their videoclips to the private folder. There were two major hurdles to this solution. The first is that the VLS is only intended for use by members of the university and there is currently no facility for external people to access it without a university username and password. The other was that, even if a folder were created that was private to the study, there may not be a way of preventing participants accessing the other videoclips already uploaded, so risking removing the anonymity of other participants from each other. As with the Adobe Connect shared meeting room, this would clearly be ethically unacceptable. A further disadvantage of this potential solution was that the participant would have to record their video clips to the computer they were using prior to uploading them. Although this may not be a problem for some participants, it does increase the risk of identifiable material not being adequately deleted and being found on the computer at a later date by a third party, so compromising someone’s anonymity. This would be a particular problem if a participant was using a public computer, for example in a Deaf club or a library. Finally, a system which required participants to record and separately upload their video clips incorporated a greater number of technical steps and may therefore dissuade participation.

This exploration of existing video capture technologies concluded that there was no pre-existing software that could be customised for use in this setting and would meet the requirements. A solution that had the novel addition of capturing responses directly via the study website was therefore sought. It was necessary for the IT specialist to custom-build this capture solution and how this was achieved is addressed below in section 5.5.2.1.

5.5 Objective 4: Build, test and pilot tool

Having resolved the specification and the requirements of the use case that would be the means of testing the functionality of the tool, the next steps concerned the development of the BSL content, the building and testing of the technology and the piloting of the tool. This was approached both in terms of discerning best practice for these elements (given the previous review of available technologies and also of Deaf people's engagement with online materials) and in terms of piloting whether the tool would actually work in being able to deliver material to stimulate data generation, engage participants in processes of autonomous data generation and be acceptable to them, capture data online in multiple languages and modalities, and remain secure.

5.5.1 Developing the BSL content: lessons learned through filming and editing

With any filming, it is necessary to position the actors so that they do not impinge on another camera shot if more than one camera is being used. For example, the filming of the genetic counselling scenario was set up with two close-up cameras at right angles to each other and a third central camera filming a wide-angle safety shot. Care had to be taken that another person does not come into frame, for example when moving a foot. This is possibly a little more difficult with a signed language because people are necessarily moving their hands more than an oral speaker. While the occasional appearance of a hand or foot in shot is not a major problem for the standard of filming needed for this study, it would be distracting if it happened repeatedly. The solution was to place the actors further apart than in real life. This could look unnatural so a careful balance had to be kept. For someone not familiar with a signed language, it might be expected that just the one wide angle shot would be very adequate. However, this would mean that the actors are being viewed almost side on throughout. Whilst a signed language is understandable from this angle, it is certainly easier to understand if closer to face on, hence my decision to use three cameras. When several cameras are used, the use of a clapperboard (or a ruler on a book) in front of the cameras can be used later, visually and/or aurally, to cue up the parallel shots in the editing software.

When editing the close-up shots on an individual in a signed language, I was aware of a different complexity from spoken language editing. Editing to a soundtrack of a spoken language which runs over the visual allows the editor to cut between two or more cameras matched on exactly the same timeline (if he/she wishes – in fact different visuals are often interchanged while the same soundtrack continues). In contrast, in a visual language, one person often starts to sign as the conversational partner is just finishing and their end of their utterance would have been clipped if I cut direct from one camera to another. It was therefore necessary to extend a shot by a few frames to allow a phrase to be finished before cutting to the film of the other speaker.

During discussion and negotiation of the narrator's sections to be filmed, she and I agreed on a strict editing and numbering of clips in the script in advance. This made subsequent reference by email between us far easier.

5.5.2 Building of the integrated tool

5.5.2.1 Development of Flash video capture tool

The video capture application was customised by the IT specialist from a basic tool available online as freeware. It was written in Flash programming language and set up a video screen showing a participant the dynamic image of themselves captured by their webcam. Under the screen is a single [Record] button. When the participant clicks on [Record], the video clip is streamed remotely and directly in FLV (Flash) file format to a university server to which access is password-controlled. No information is captured by the computer used for the recording. During recording, the button appearance changes to say [Stop] and the participant clicks the button again when they have finished talking. There is a short pause whilst the upload of the clip to the server is completed and the button changes to [Wait] so the participant is aware of this process.

Alongside the record screen is a preview screen. When the upload is complete, the clip starts to play back automatically on the preview screen. The participant can watch it all or stop it using the [Stop] button under the screen. They can replay it if they wish – the [Stop] button changes back to [Preview] once the clip has finished. If they wish to replace what they have recorded, they click [Record] again and it copies over the original file. If they wish to append extra information, they mark the checkbox next to the record button that says 'add more to my answer' before clicking on [Record]. The entire Flash app could then be embedded within each webpage for which video capture was required.

5.5.2.2 Building the primary structure and layout of website

As this complex video capture solution was being discussed, the web development team's remit was being re-evaluated. When I returned to them several months later with the website structure and English content ready for development and the BSL filming and editing timetabled, they were no longer able to dedicate the time to developing a custom website from scratch.

This was a serious concern. However, the web development team had carried out a significant amount of work in the meantime on SelectSurvey software for use across the Faculty. This software (http://www.classapps.com/selectsurveynetoverview.asp) is used to produce online questionnaires with powerful flexibility and functionality. The team had branded this to match the University website so that individuals could develop their own questionnaires without the need for further cosmetic adjustments. I could therefore use this software to develop most of the questionnaire independently. There were some questions and information content which SelectSurvey could not tackle without further modification and these specific adjustments are explained in the next section.

Some of the SelectSurvey integral features that I particularly valued for this study included a marker for progress through the questionnaire. I could access all responses to multiple choice questions or written English responses within SelectSurvey itself. Because the video capture tool would be embedded within the SelectSurvey pages rather than being an integral part of the software, the two types of responses (multiple choice/written English and BSL videos) were captured to different secure servers within the University IT infrastructure. However, with the correct permissions set up in SelectSurvey, participants could choose to respond in a mixture of BSL and written English, even within the same question i.e. there was no stipulation that they must complete every video response or text box.

5.5.2.3 Information in a signed language on a website

One principle within the tool specification was to present the information in BSL and English on the same page. As well as meeting Deaf people's varied communication preferences, this bilingual presentation can be used practically by a participant to shorten completion times if they also have good written English skills and wish to read ahead faster than the video. The written English can also be useful for marking points in the questionnaire in case someone wishes to go back to a previous page and find a video clip.

Although a further principle was to provide a choice of responding in any combination of BSL, spoken English or written language, the content encouraged participants to respond in BSL as it was nuanced information about language in use, culture and context that was particularly sought through the use case. In addition, given the internet is still currently a predominantly text-based medium (section 5.2.2.1), I prioritised BSL within the tool by embedding the clips at the top of the page, in prime position, rather than the BSL being secondary to the English content.

Although I was already aware in principle of the points in this section, the experience of developing the website pressed home to me their importance. It was crucial to break up instructions and questions into short BSL videos and arrange them within menus so that people can see them in manageable chunks. It is easy to stop and start reading within a longer written document, but not easy to find a specific point in a long video. A further advantage of the English translation being alongside is that it can help with this bookmarking process.

Working between any two languages requires careful checking that any changes made later are also made in the other language and this is no different when working between a signed language and a written language. Careful checking between the BSL narrator and me continued throughout production.

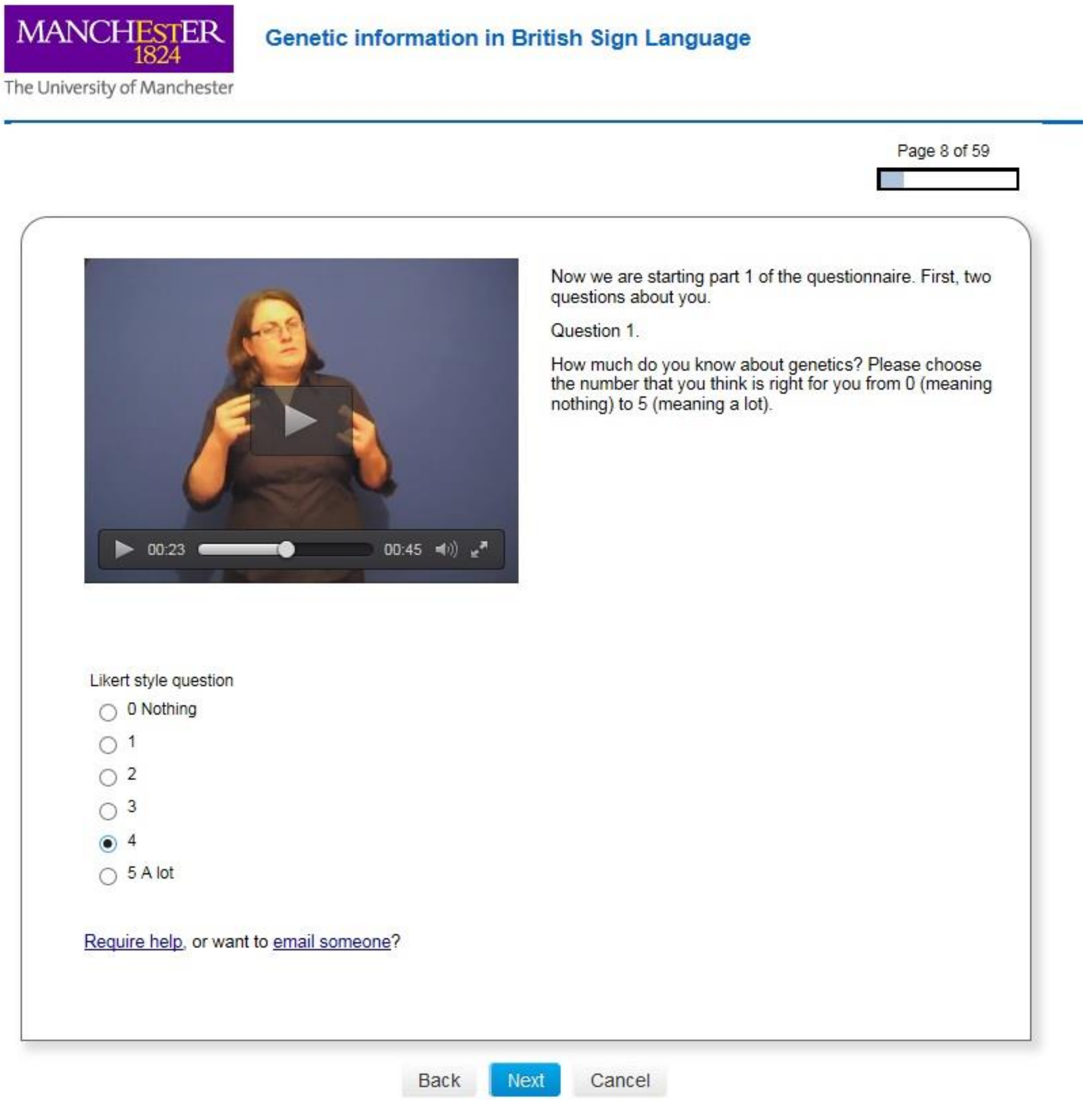
One of the most important points throughout and one which had the potential to significantly delay production of the BSL sections of the website is the highly specified nature of signed language. As discussed in Chapter 3, this refers to the fact that physical placement of a sign in three dimensions is a crucial and integral part of a signed language. What this means in practice when a website is being constructed is that the structure of webpages must be known in advance of filming the video clip. This is particularly true for any instruction page. For example, if the narrator signs ‘click the buttons in the menu to play other information about the study’, the language also specifies where the menu is in relation to the signer i.e. on their left and slightly higher. When there were delays earlier in development with the technical aspects of website construction, this meant that filming also could not start because layout could change and make a previously-filmed section defunct. Whereas a written language could be changed slightly, a filmed section would have to be redone entirely.

When the narrator was ready to film, it was necessary to confirm the format for video files with the specialist who worked on the VLS. WMV, AVI or MP4 format were all acceptable, but it was important that the computer used to edit them had the correct codecs (compression and decompression software) installed. The only potential restriction was that AVI files are huge compared to the other two types and the maximum file size for upload to the VLS at the time was 2Gb. However, this file size roughly equates to 30 minutes of film so was unlikely to influence format decisions.

5.5.2.4 Customisation of proprietary software

Construction of the questionnaire in SelectSurvey took a very long time – much longer than would be imagined for most software applications. The reason for this was that every change made had to be rendered (incorporated into the software) in real time, meaning that I had to wait a couple of minutes even after every small adjustment to wording. This was an important lesson for future projects and time-planning, though developments in the SelectSurvey platform since then mean that rendering changes is becoming quicker and therefore less of an issue.

Some questions had response buttons which allowed a single choice to be made (see Figure 3 on the following page for an example) or several buttons could be checked if the question required e.g. a number of different formats in which information could be provided.



Genetic information in British Sign Language

Page 8 of 59

Now we are starting part 1 of the questionnaire. First, two questions about you.

Question 1.

How much do you know about genetics? Please choose the number that you think is right for you from 0 (meaning nothing) to 5 (meaning a lot).

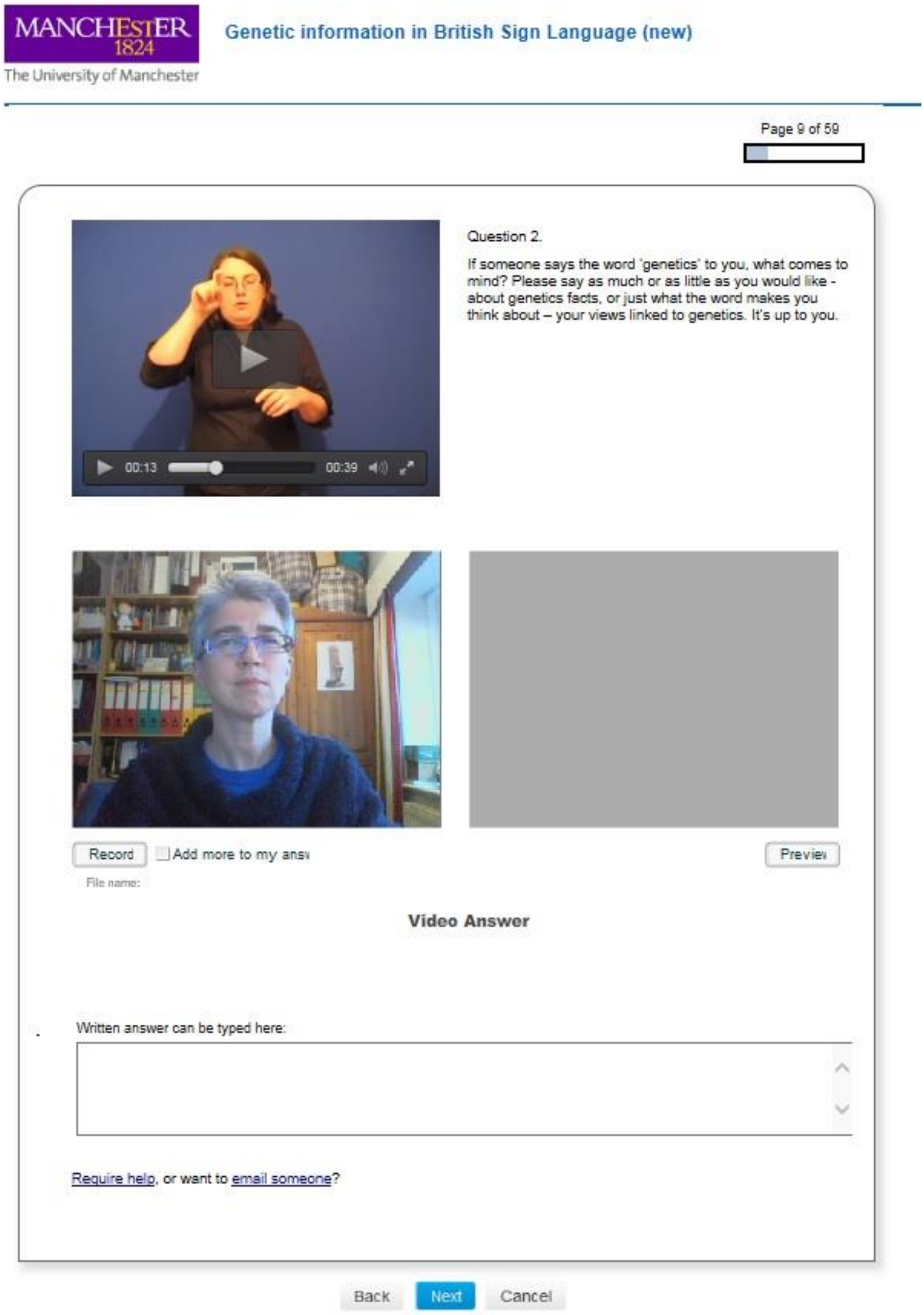
Likert style question

○ 0 Nothing
○ 1
○ 2
○ 3
● 4
○ 5 A lot

Require help, or want to email someone?

Figure 3: An example of a Likert style question

Most of the questions were answered with the choice of video or written text – or both if the participant wished. Figure 4 overleaf shows a typical layout for one of these questions.



Genetic information in British Sign Language (new)

Page 9 of 59

Question 2.

If someone says the word 'genetics' to you, what comes to mind? Please say as much or as little as you would like - about genetics facts, or just what the word makes you think about – your views linked to genetics. It's up to you.

Video Answer

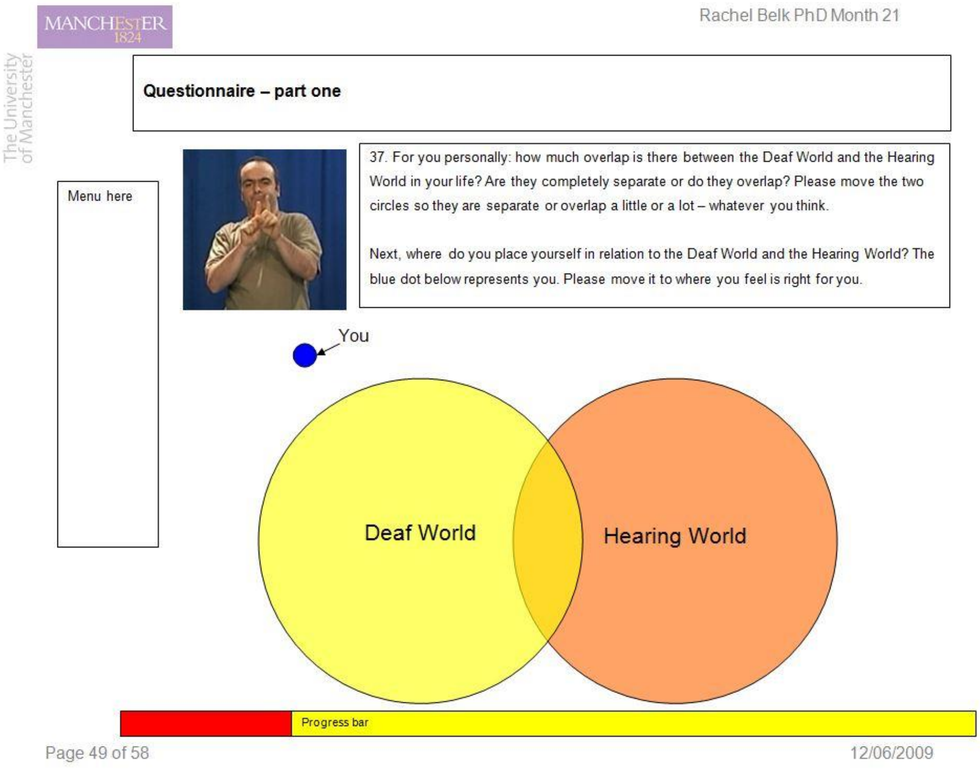
Written answer can be typed here:

Require help, or want to email someone?

Figure 4: An example of a question answered with choice of video or written text

The majority of planned questions could be accommodated within the functionality of SelectSurvey, either in a straightforward way or with only small modifications. However, the tool was developed by bolting together different software already available within the University using in-house expertise to build the links and pass on the skills to me so

I could build the questionnaire. This meant that there were limitations on what development was possible and justifiable within the time available. An example of a question which could not be realised is shown in Figure 5. Because it was visual, it would have required an additional small Javascript or AJAX programme to capture the data using spatial coordinates.



Rachel Belk PhD Month 21

Questionnaire – part one

37. For you personally: how much overlap is there between the Deaf World and the Hearing World in your life? Are they completely separate or do they overlap? Please move the two circles so they are separate or overlap a little or a lot – whatever you think.

Next, where do you place yourself in relation to the Deaf World and the Hearing World? The blue dot below represents you. Please move it to where you feel is right for you.

Page 49 of 58

12/06/2009

Figure 5: A proposed visual question

The question had to be changed to two questions which instead required video or free text responses:

- Q37. How important to you is your identity as Deaf? Please choose the number that you think is right for you from 0 (meaning not important at all) to 5 (meaning extremely important and central to who you are). Please tell us more about why you chose this number.
- Q38. How much time do you feel you spend in the Deaf world? I mean time spent with Deaf friends, family members or colleagues where there is a shared understanding of Deaf culture, identity and communication? Please choose the number that you think is right for you from 0 (meaning no time at all) to 5 (meaning all of your time). Please tell us more about why you chose this number.

This change adequately sought similar information, but the original question format would have represented the concept more visually.

SelectSurvey is not as flexible as a custom-built website, so part of the challenge was thinking of ways of getting round the limited integral options. It involved either directly writing and inserting short pieces of HTML code to tell the software how to format the page, or using more user-friendly software like Dreamweaver to produce the HTML code. For example, the existing page set-up in SelectSurvey put the BSL video screen at the top with the English version underneath. This made the page too long so the viewer had to scroll down and it also wasted screen space alongside the video screen. Dreamweaver was used to programme an invisible table with two columns so that the screen could be put in the left-hand side and the English text in the right-hand side.

HTML code was needed to specify carriage returns in blocks of text, to add scrollbars and frames around text and to construct the menus which host and display the video clip options on each page e.g. where there is a choice of several clips to play within the same page (see Figure 6 on the following page). The embedded video player allowed the clips to be enlarged to full screen.

Hello. My name is Rachel Belk and this is my sign name. On the last page, Jen started to explain the research in depth. When I've finished, on the next page she will carry on explaining in depth. I wanted to introduce myself so everyone can see what I look like.

This research started in 2007. My background up to now is working as a genetic counsellor. I started doing that 15 years ago and continue up to the present. Genetic counselling means I meet people who have a condition in their family that is definitely or possibly inherited. People have very varied questions or worries.

For the last six years, I have focussed on access to services for Deaf and deafened people. Particularly, I think about access to genetic counselling services. I think about how people working in the NHS could improve information and communication. We know that there are barriers to accessing medical services for Deaf and deafened people. Some of these barriers are because of things to do with communication and information.

If you want more information, click on the other two options on my left.

When you have finished watching, click [Next] at the bottom of the page. On the next page, Jen will explain more in depth about the research.

How could this research be useful?

The aim of this research is not to persuade people to come for genetic counselling. I know that genetics is a sensitive and controversial subject area, particularly in the Deaf community. At the end of the research, we hope that we will have improved information about genetics and related areas of medicine. This means Deaf people can have access to better information.

This could mean that people have a better understanding of genetics and this could be useful for two reasons. Firstly, for an individual person so they can decide for themselves if genetic counselling would be useful or interesting for them personally. Secondly, so that someone can decide if they want to be more involved in discussions about genetics in the wider community and in the media.

Who else is involved in the research?

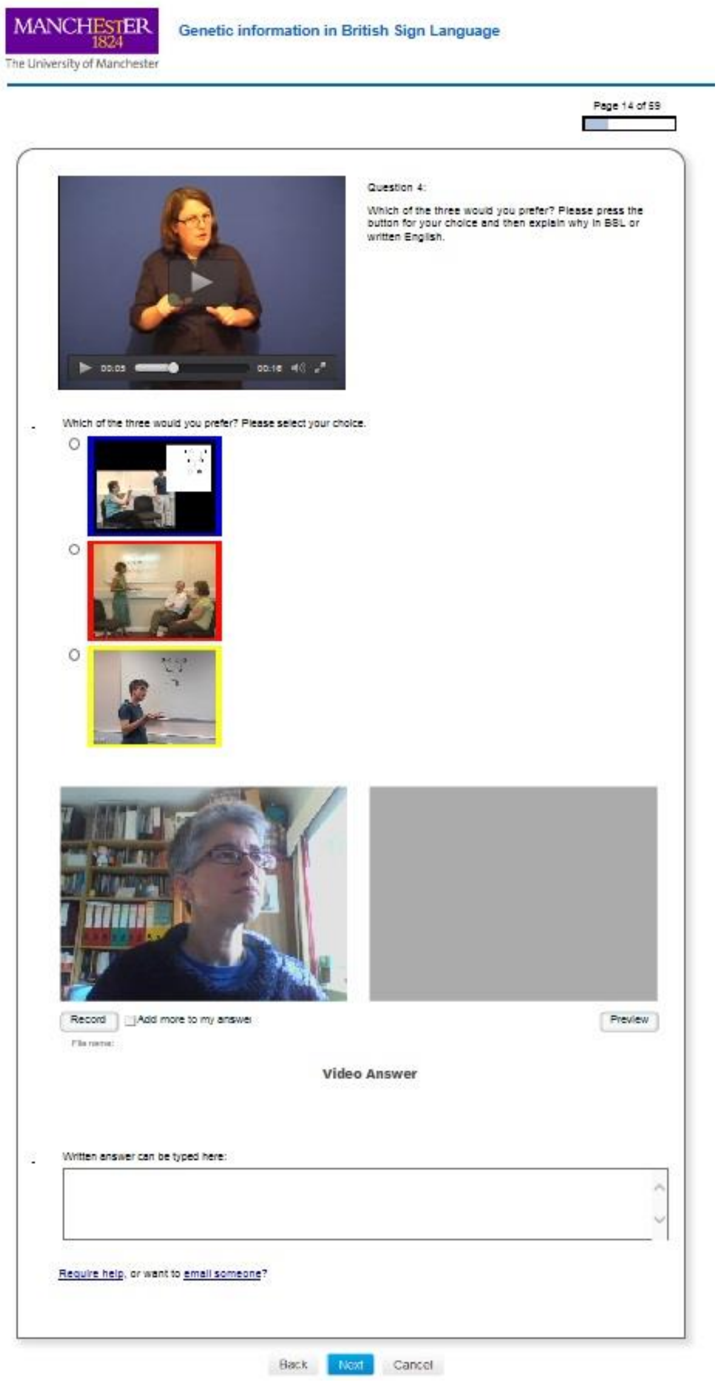
I am studying in the University of Manchester. I have two supervisors who are both professors: Alys Young and Dorothy Trump. I also have a mentor, Valerie Leach. If you want to see what they look like, their photographs are below.

Also, there is a project advisory group. The group is a mixture of professionals and lay people and a mixture of Deaf and hearing people.

Require help, or want to email someone?

Figure 6: An example of videos embedded in a table

SelectSurvey allows only written text to label question responses, but there were some questions where I wanted to refer back to video clips using a screenshot from the clip – in other words, labelling a response button with a picture. Javascript programming was needed to allow picture buttons to be inserted into the questionnaire and, once done, that question could not be edited further. An example of this use was in asking for relative judgements of preference between different language/ communication options in context. Figure 7 shows a question where the screenshots have been colour-rimmed for easy reference e.g. ‘the red option’.



Genetic information in British Sign Language

Page 14 of 59

Question 4:

Which of the three would you prefer? Please press the button for your choice and then explain why in BSL or written English.

Which of the three would you prefer? Please select your choice.

○

○

○

Written answer can be typed here:

Require help, or want to email someone?

Figure 7: Labelling of response buttons with images

A minor concern was that there was no integral way of stopping someone from scrolling through the entire questionnaire out of curiosity when they had no intention of completing it. SelectSurvey options can be chosen so that a checkbox must be ticked or something typed into a text box before a participant is allowed to move to the next page, but there is no means of doing the same with the video response, given that the video capture tool is essentially a separate program inserted into SelectSurvey. I considered adding a check box at the bottom of each page of the questionnaire that a participant had to tick to confirm they had completed their answers on that page. A participant could actually still leave the other response options on that page unanswered, but the intention was that it would reinforce the message that the questionnaire was only intended for use by people who intended to complete it. However, in discussion with one of the IT specialists, we agreed that this extra non-essential step could be off-putting to a participant, given the length of the questionnaire and the complexity of the subject.

One of my biggest concerns during building of the questionnaire was that both the narrator and I found we were regularly timed out of the survey. In discussion with the IT team, I discovered that the timeout period was set for 20 minutes. While 20 minutes is a reasonable time if someone is temporarily taken away from the computer during completion, the main videoclip that participants need to watch early on is 30 minutes long so the questionnaire would time someone out even whilst watching that clip.

Related to this issue is the fact that, when it times out a viewer, they get the rather worrying pop-up message that they have to save what they have done before it times them out - but no instructions are given as to how to save. In fact, on checking, it is not actually necessary to save anything as SelectSurvey automatically saves any response with each page turn.

Of greater concern was that, after being timed out, one would follow the same hyperlink to the questionnaire, but be given an error message saying that the questionnaire could not be reached. There seemed to be no way round the error message other than finding a new link through to the questionnaire from, for example, a different email and starting over again. The IT team’s explanation was that the system can get ‘confused’ after a timeout or when the [Back] button or [Cancel] button are used at the bottom of a page. This is due to it being a dynamic system that renders on the fly, which means that the whole questionnaire is actually a single webpage and has to update itself every time a participant moves to a new page. The error message is due to the URL being adjusted in the address line every time one moves page.

The IT specialist was not greatly concerned by this happening and thought that those people editing were much more likely to get this error than a participant who is moving more quickly and linearly through the website – he had not been previously aware of a participant in SelectSurvey having this problem. Nevertheless, it was a concern because of the potential to lose a participant if they received the error message and then could not access the questionnaire again or simply lost interest because of the difficulty. To try and minimise the chances of this happening, I asked if the timeout function could be removed. This is not possible because a master setting like this would have the same effect on all the 500 or so SelectSurvey surveys open across the university at that time. The IT team did agree, however, to increase the period before timing out to an hour, which I was confident would significantly reduce the chances of this problem happening for a participant.

The next development step was to work out a way of generating a unique filename for each videoclip generated through the study. This was not only one for each participant, but one for each question response for each participant. Given that there were 50 pages in the questionnaire that gave the option of a video response and, if using the questionnaire, 25 participants might be a reasonable estimate, this number would generate 1250 separate clips so a logical and identifiable label for each was essential. The technical description of this step was to pass a variable that identified the specific questionnaire page to the fileName variable that was waiting for it in the Flash programme file.

At this stage, the Flash specialist liaised with a specialist in a separate IT team within the University. The latter specialist understood the structure of the webpages containing the questions: this understanding was specific to the platform within which the questionnaire was finally hosted (SelectSurvey) so this problem could only be solved once the decisions about the platform had been made. His solution was to write Javascript code which captured the unique respondent number and page number from the URL (the address line of a webpage). In the website that was being created (as with most others), the URL would change with each click through to the next page. The Javascript code inserted the unique respondent number and page number into the filename for the videoclip recorded on that page. Importantly, this meant that the Flash recorder could not be placed on the first page of the questionnaire as it needed a ‘page turn’ in order to generate the filename. The filename was created from the two Flash variables (or FlashVars) in the format ResponseID_QuestionNumber.

Another minor issue was that SelectSurvey generates an odd and unmemorable web address which is a random mixture of letters and numbers – so the direct address for this study questionnaire at the time was: http://www.mhs.manchester.ac.uk/surveys/TakeSurvey.aspx?SurveyID=742L4m2. This was not a problem if a potential participant was clicking directly on a hyperlink of this URL. However, it was not accessible if used on a study advertising poster. I therefore set up another webpage with a more memorable address: http://www.mhs.manchester.ac.uk/geneticsigns and this page redirected the viewer to the SelectSurvey page within a couple of seconds. A similar issue was to request the set-up of a new study email address (genetic.signs@manchester.ac.uk) that would keep study-related questions separate from my personal University email account and would also reduce the potential for spam mail because the email address was on a public website.

The final points relate to issues that arose for specific questions. I wanted to give participants the option to print out a completed copy of their own consent form. I set up links within the questionnaire pages to other documents, such as the information sheet and English translation of the acted scenario, so they could be accessed as PDFs by the participants. However, this could not be done with a page that had just changed by addition of personal information so the solution was to give instructions to simply print out the form using the browser toolbar at the top of their screen.

Because of the length of the questionnaire, participants are given the option partway through to miss out the second part and jump directly to the final demographic questions. This page therefore needed two option buttons which were programmed to move to two different pages. SelectSurvey has this function already installed, but testing was needed to check that jumping in this way to another page did not cause problems with the generation of the video capture filename on the subsequent page: it worked without a problem.

A further request that I made to the IT team was to have a link on each page to access the participant instructions about recording video clips. I thought this would be valuable to avoid a participant having to scroll back through multiple pages if they forgot something. One of the inflexibilities of SelectSurvey is that sidebar menus, common throughout the University's webpages, cannot be easily added. The solution was to add a pop-up window, though even this had potential problems as some participants may have had pop-up blockers in place. As a safety net, I therefore also added a link on each page to allow a one click option for a participant to email me about any problems.

One of the final steps in development was the adjustment of the technical instructions for participants because of the changes that were made during development. These instructions were drafted in English, but translation and filming in BSL was left right until the end. I explored several options for the clearest illustration of the instructions. These included the addition of still screenshots to illustrate specific points or Camtasia software (which captures dynamic recordings of the computer screen) to capture video clips of mouse movements around the screen showing how to operate the Flash recorder. These could have run after the narrator had explained it in BSL. A detailed version could have been used for the first explanation together with a short reminder version for the pop-up on each page. However, I decided, in discussion with the narrator, that I would use only the BSL instructions because the introductory information was already lengthy to watch. As BSL uses placement as an integral part of the language, it was already more specified and therefore clearer than spoken or written language and naturally incorporated information about the location of click buttons and page layout.

5.5.2.5 Management of captured video data

The captured video was saved in a folder on the Faculty of Biology, Medicine and Health (at that time the Faculty of Medical and Health Sciences) media server. However, this was not the same as the video having been processed by the VLS so it was not possible to link directly to the VLS to view the videos. An alternative solution would have been to use an application which, given the names of all the videos, allowed one to cue up a series of videos for later playback and stream them out of the server for viewing. However, the disadvantages of any mechanism for online viewing was firstly, that the researcher would always have to be connected to a fast broadband connection and, secondly, that the files could not be manipulated, edited or tagged in any way if this was required for analysis of the data. For example, the most recent versions of NVivo CAQDAS software (Computer-assisted qualitative data analysis software) allow video files to be directly tagged and coded, whereas this was only previously possible with text files, perhaps of transcribed interviews or other written data. In order to use such software, it would be necessary to download the video files.

To achieve this, it was necessary for me to install File Transfer Protocol (FTP) software (Filezilla freeware https://filezilla-project.org/) on my computer. FTP software allows the secure server to be seen and sets up a standard network protocol to access and transfer files to my encrypted computer. It was suggested by one of the IT specialists that the clips could, at this stage, be re-uploaded to the VLS if online analysis was decided upon. However, in addition to the option to use standalone CAQDAS software, there were confidentiality concerns about VLS as IT staff could potentially view the videos, even if access permissions were otherwise limited to me.

Given that the capture software was written in Flash, the captured videos are in Flash Video format (FLV). As a less common file format, this was not (and still is not) one that would play on the majority of software commonly provided with Windows operating systems. To view the files once downloaded from the server, I therefore downloaded the freeware VLC Media Player (http://www.videolan.org/vlc/index.en_GB.html), not widely used in 2009, but now installed on all University of Manchester directly-managed desktop computers because of its flexibility in playing a wide range of video formats. One final step, however, was that FLV is not a format accepted by NVivo so the video files would need to be transcoded from FLV to MP4 format using Handbrake freeware, whose use was mentioned in section 5.3.1.5 for ripping files from DVDs.

5.5.2.6 Summary of challenges found during development and their solutions

The development issues addressed in sections 5.5.1 and 5.5.2 are summarised in Table 2 below.

Page	**Challenge**	**Solution**
138	Actors may impinge on another camera shot	Actors position themselves a little further apart than in real life
138	Matching the timelines of recordings from multiple cameras when working in a signed language	Use of a clapperboard (or ruler on book) to later cue up the parallel tracks visually and/or aurally within editing software
141	Difficulty for viewer of returning to a specific point in a long video	Break up content into short BSL videos and arrange within menus Use English translation alongside as an aid to bookmarking
141	Accommodating highly specified information in a signed language during filming	Finalising webpage structure in advance of filming so placement can accurately reflect layout
142	Compatibility of video file format with editing software	Check codec installation in advance
145	SelectSurvey not able to accommodate complex visual questions	Potentially could be solved with additional coding to capture spatial coordinates, but not pursued in this project
146	SelectSurvey has fixed page	Dreamweaver and HTML text added to alter

	layout not ideal for prioritising BSL videos	page layout and add tables of nested videos
148	SelectSurvey only allowed words as answer labels	Javascript code added to enable images as answer labels
149	SelectSurvey has a default timeout period of 20 minutes for any questionnaire	Agreement from IT team to increase timeout period to 60 minutes
150	Large number of video clips may be captured so logical and identifiable labels needed	Javascript code written to capture the unique respondent number and question number (page number) from the URL and generate a filename in the format ResponseID_QuestionNumber
151	SelectSurvey generates a long and unmemorable web address for the questionnaire	A redirect to the questionnaire was set up from a memorable web address
151	Participants potentially having problems during completion of questionnaire	Link to completion instructions and email link to dedicated study email address added to each page

Table 2: Filming, editing and technical development challenges and their solutions

5.5.3 Testing of the integrated tool

5.5.3.1 Scope of testing

The primary technological build issues have been outlined in the previous section. In this section, I outline the field test concerns and procedures. I tested the online tool widely e.g. from the University, from home and from other domestic internet connections; on desktops and laptops; within different browsers e.g. Internet Explorer, Firefox, Chrome; and different operating systems (Apple and Windows). This allowed testing of a number of variables:

- Broadband speed, computer specification and webcam
- Testing on common internet browsers and operating systems
- Firewalls that may not allow video streaming
- Settings that may mean pop-ups are blocked

No problems were found with any of these, other than those expected e.g. a pop-up blocker would not allow PDFs of written leaflets to open, strict firewall settings (such as those found within hospitals) would not allow video streaming. I was aware of the importance of a reasonable broadband speed, but this was not possible to influence given a participant’s choice of location. I therefore included the option to contact me to discuss technical problems and the option for participants to provide their email address so I could contact them in the event of technical problems.

One major area that was not as prevalent in 2009 was the use of smartphones and tablets. I therefore did not test the website on these platforms. This has been a huge shift since that time and it would now be unthinkable to launch an online questionnaire without testing on all mobile platforms as well. For example, I have since worked on a project where we used a similar website to test the validity of the BSL version of the ED-5D-5L health questionnaire (Rogers et al., 2016). We realised during testing that the videos embedded in SelectSurvey would not stream on mobile devices and the embed codes had to be updated to accommodate this requirement.

5.5.3.2 Challenges found during testing and their solutions

The main issues of concern raised during the test phase centred on participant navigation and use of the tool and whether actions that a participant might reasonably take would be problematic for the underlying programming. For example, one issue about the Flash program’s interaction with individual webcams was found immediately during testing. A participant cannot have any other webcam software open at the same time as the Flash program otherwise the program will not recognise the webcam’s existence. This was addressed by an addition to the participant guidelines on the use of the questionnaire, explaining that all other webcam software must be closed in advance.

It is easy when using written responses to give the option of previewing an answer before submission. This same option is not as straightforward with a video response, but I liaised with the specialist building the Flash program and it was possible for him to build in the preview screen to automatically play back their response.

Some webcams did not record sound as well as video through the Flash program. This would only be a problem if the participant wished to respond in spoken English. I envisaged this would be a less popular option, but the principle was in place to provide the choice to respond in spoken English and therefore a solution was sought. In addition, this could be useful for future applications of the technology with populations whose preferred language was spoken rather than signed. The specialist found that the cause was more than one microphone being available to the computer and the computer defaulting to, for example, the internal microphone. The Flash program stores a list of available microphones in an array e.g. item 1 = USB microphone, item 2 = integral computer microphone. To fix this problem, he wrote an additional small script to make the program find the microphone. The command was ‘Microphone.get()’, which causes Flash to access either the only available microphone, or the first one in the list, or it returns Null (which means there is no microphone and the application does not crash).

The following illustrates the complexity and innovation of the type of solutions tried during the testing: the specialist also found two alternative methods to specify which microphone in the list to access. The first of these uses Microphone.get(0) to access the first microphone in the list or Microphone.get(1) to access the second microphone in the list and so on. The problem with this method is that it is not automatic that a USB microphone (which would be integral to a plug-in webcam) would be the first in the list so it is not possible to know which number to use.

A slightly more complex method is to use the System.showSettings(2) command which opens up a window asking the user to select the microphone they want to use. This is then followed by Microphone.get() to set the one they have chosen. The problem with this solution is it expects participants to understand more about the technology behind their computer and it may not be obvious which microphone to select.

This is an example of the type of hitch that had to be negotiated between the IT specialists who knew the limits of the hardware and software and my understanding of the requirements when working bilingually and bimodally. In this case, when presented with the options, I recommended that we use the first and simplest method, judging that, with this population, the number of people wanting to use spoken English would be small so any additional technological choices would be off-putting rather than helpful for the majority. For participants that did want to use spoken English, the first method would pick up the correct microphone in most cases. The safety net put in place was to add a line to the instruction page asking anyone who wanted to use spoken English to record a test video and then email me so I could check it had recorded sound. If it had not, it would have been possible to negotiate checking microphone settings by email or videolink conversation in BSL.

There was a second issue with webcams. It was found during testing that, with some webcams on some internet connections (lower bandwidth), the webcam ‘zoomed in’ on the participant. The specialist realised that this was due to particular cameras trying to compensate for the internet speed not coping with the amount of data by prioritising the frame rate per second over the image size. This is a tricky trade-off as frame rate is crucial for signed languages as too low a rate will cause a loss of clarity and a swooshing or blurring effect. However, the zoomed-in image size meant that it was now far too small to capture the signing frame and this is an even more important factor with a signed language.

The specialist made a further change to the program so that image size was given priority over frame rate. He also lowered the frame rate so that the Flash program was less likely to manipulate the video settings due to limited bandwidth. He commented in doing this that he had gathered from a number of online discussion forums that the new frame rate of 15 frames per second (fps) was thought to be more suitable for internet streaming in any case. This highlights another important issue: that the majority of discussion by information technologists about video capture and streaming is around appropriate settings for spoken language. At the time this issue signalled the importance of developing experiential guidelines for website settings in signed languages, which may well be different to those for spoken language. For the purposes of this study, I tested the different frame capture rates and agreed that 15 fps gave a clear and understandable recording of BSL.

As the specialist commented, the one issue it is really difficult to overcome is very low band width. Video streaming really requires reasonable bandwidth. The issue of video clips pausing partway through on a slower broadband connection also arose with the clips from the VLS embedded in the questionnaire. The VLS settings do give the option of embedding a lower resolution version of the video and this solved the problem in most cases. However, it does mean, as with the capture settings, a slight compromise on clarity of picture and therefore clarity of signing. Other options that may have been possible to develop, if needed, include buffering prior to playing the video (i.e. downloading the start of the video file to a temporary location on the computer so giving it a head start as it begins to play), giving participants the choice between the high and low resolution versions depending on their connection speed or giving them the option of fully downloading the clip and then playing it from their hard drive. None of these were available as part of the standard choices within the VLS at the time of developing this questionnaire.

Given the questionnaire length, I was offering participants the option to stop at any stage and return later. Having identified the problem of timing out during development, I was concerned that the system may also see a break as a timeout, generate an error message and prevent a return. In addition, I needed to test whether it made a difference if the questionnaire was closed down using the [Cancel] button at the bottom of the screen or by using the X in the top right-hand corner of the browser window. The IT team thought neither of these were likely to be problematic and I found that they could be overcome. However, to allow a participant to return to a survey in progress, it was necessary to choose a specific setting during Selectsurvey development that allows a return at any time within the following 30 days.

Returning to one of the bespoke developments – the Javascript which captures the URL from the address and creates the file name for the video being captured on that page – this only works if a participant has just moved to that page from the previous page. Another concern was therefore whether this process would fail if a participant returned to the questionnaire partway through - would they have to go back a page using the [Back] button and then forward again in order to generate the filename or would it have already happened when they accessed that page the first time round? Related to this issue, if a participant took a break, would the filename contain the same participant number on their return or would the website generate a different one?

To ensure the same filename was kept, the answer was that a participant would need to bookmark the URL (i.e. save to ‘Favourites’) and return to the survey using this link. If they did this, the URL would contain the responder ID and page number, so they would both return to the same place and retain the same generated filename. However, if they returned to the questionnaire from the original hyperlink and were using the same computer (specifically, the same IP or Internet Protocol address which is the numerical label identifying a computer on a network), the SelectSurvey setting I had chosen would enable a return to the same page, but the first page turn would generate a new responder ID as part of the Flash filenames. If they used a different computer or location (perhaps using a laptop computer, but linked to a different wi-fi network), the hyperlink would take them to the start of the questionnaire, SelectSurvey would register this as a new response and the Javascript would generate a new responder ID for the Flash filenames. The participant information and instructions for completing the questionnaire were already long so I decided not to include the information about bookmarking the URL, making the decision that the additional technical steps may be off-putting to participation. I judged it would not be a problem as it is still possible to link the SelectSurvey number to the number on the video files, even if a participant had two or more different responder IDs. My only remaining concern was whether, if a participant returned to the last page they had accessed and tried to record there, without a page turn, whether the video would be captured correctly. I tested this by returning to the questionnaire a number of times, which gave me, correspondingly, several different responder IDs, but there were no issues with labelling of the clips.

Table 3 below summarises these challenges found and fixed during the testing process.

Page	**Problem**	**Solution**
155	Simultaneous use of Flash capture tool and other webcam software not compatible	Point added to participant instructions asking them to close down any other software using the webcam
155	A written response can be viewed before submission: seeking equivalence for a video response	Addition of a preview window to the Flash video capture tool
155	Webcam not recording sound	Addition of script to the Flash capture tool to force the program to find the microphone
156	Slower internet speed causes webcam to ‘zoom in’ to a smaller picture	Addition of script to the Flash capture tool to prioritise image size over frame rate

Table 3: Challenges found during field testing

5.5.3 Piloting of the tool

Once the tool was functioning, there were three key considerations that required a pilot evaluation. These were: linguistic equivalence, cultural acceptability and technical aspects of using the website. For the latter, this included particularly:

- Layout of the BSL within the webpage, using menus, editing and timing issues
- Prioritisation of frame rate over image size in the data capture tool
- Timing out of SelectSurvey

The pilot was carried out with six Deaf people. Three were members of the project advisory group and three were other Deaf colleagues. There was one other hearing colleague who additionally offered to test the technical aspects.

None of the pilot users raised concerns with the layout of the webpages, the linguistic equivalence of the questions and resources, nor the cultural acceptability. Given the information resources were predominantly a vehicle for the use case, I was not asking those who piloted the website to comment on this content. However, one issue was raised: an important principle with a visual language is managing the number of different things happening on the screen at the same time. This is because there are considerations of divided attention when all language and information is being received visually rather than through visual and auditory channels. I had edited in recessive inheritance diagrams alongside the acted scenario and had cued them to appear when the BSL interpreter signed about them. However, two of the Deaf pilot users advised that it was better to have them appear when the genetic counsellor spoke about them so that viewers have the chance to look at them before moving to look at the explanation given by the interpreter.

There was one concern with the technical aspects identified by the hearing colleague. She found she was able to record her voice, but the image did not record. I tried to reproduce this problem elsewhere, but could not and it was not a problem for any of the other participants. I had to therefore conclude it was relatively unlikely to happen again.

The concerns about connection speed were borne out with one user, where the frame rate was significantly reduced due to a slow connection, but the fix put in place during testing had maintained the image size as planned. In this case, the videos were blurred and more difficult to understand, but could still be deciphered.

Table 4 summarises the challenges found during piloting

Page	Problem	Solution
159	Considerations of divided attention for signed language users	Careful pacing of diagram introduction to minimise overlap with important utterances by the scenario participants
160	Tool recorded a video file, but only voice was transmitted and no image was recorded	The problem could not be reproduced so no cause or solution could be found
160	Frame rate was significantly lowered due to slow internet connection	Image size was maintained so the content was understandable, though blurred and less easy to watch Slow broadband cannot be fully overcome

Table 4: Challenges found during piloting

5.6 Conclusion

This chapter has described the parallel development of questionnaire content, filmed content, video display mechanism, video capture tool and the linking of these building blocks to realise an integrated tool that met the specific needs of this project. During the development process, smaller challenges arose from issues of clarity, ethical management and technical hurdles. Many of these challenges were impossible to anticipate until the process was underway and the recording within this chapter of the principles underpinning them will allow their use as a starting point for future online data capture, particularly that which seeks to prioritise visual data capture. The study therefore met its stated aims and objectives. Any development in data capture such as the one described will have its own technical shelf life, but the principles remain innovative in their application and their implications for today’s users and future research challenges are discussed in Chapter 6.

Chapter 6: Discussion

6.1 The existing tool

This project has met its aims of developing a means by which Deaf people can engage independently, remotely, at a time of their own choosing and in their preferred language. Its testing and piloting showed that its use as a means of data collection is feasible.

The existing tool certainly has limitations and challenges as discussed in Chapter 5, not all of which could be overcome. However, the use case approach, now widely used for software and IT system development, specifically sets out a process that delivers a new system in increments. Under this model, this iteration would be acceptable as a first release with testing and reconstruction allowing improvements in the next release. Jacobson states that ‘Each increment provides a demonstrable or usable version of the system.’ (Jacobson et al., 2011 p10), which this tool has achieved.

Although technologies are developing quickly, the tool, as developed, could still be used. However, hardware and software changes mean it is inefficient to develop it with the same technologies each time it is needed. Selectsurvey has been updated so the coding used to modify it needs to be updated each time. In addition, each Selectsurvey questionnaire that required video data capture needs to be individually adapted which is time-consuming. There are also concerns within the University about server space so space on a streaming server for research use has to be negotiated. At this time, Selectsurvey remains the preferred solution for questionnaire creation. Free applications such as SurveyMonkey are commonly used, but captured data is held by the company owning the software and there are therefore data security concerns, specifically the need to meet Principle 7 of the Data Protection Act (1998).

This bespoke solution for an online questionnaire that allowed video presentation coupled with video capture was original and innovative at the time of its development. However, because it used pre-existing proprietorial software as major components, it was not possible to protect the IP of it. In effect, anyone could have used the same software to come up with this kind of solution. The novelty at the time was such that, along with six colleagues working in similar areas, we succeeded in gaining an ESRC Digital Social Research Community Activities Funding Scheme grant (Young et al., 2012) in order to widely disseminate the solution and its usage to other researchers in the field. As a result, the development process and the computer code have been shared with other research groups through knowledge transfer workshops and material transfer agreements. Resulting interest in the tool has shown that there is a demand from researchers working in Deaf studies and linguistics of signed languages and I discuss the possibilities of further research and development below.

6.2 The principles identified

Separate from the direction of future research and development of such a tool, and despite the swift technological advances, from the perspective of 2017, the key principles that underpinned this development still have high contemporary relevance. These principles are summarised as follows:

- Presentation of the information in both BSL and English so that Deaf people with some degree of bilinguality could choose to access either or both languages
- Facilitation of responses in any combination of BSL (as video clips), spoken English (also using the video capture tool) and written English, also to maximise choice and access for Deaf people
- Provision of website navigation instructions in BSL as well as English in an attempt to mitigate the structure of the internet as a predominantly text-based medium
- Use of information structure and content that:
    - provided a context that made it understandable and that could stand alone as future valuable resources
    - facilitated exploration of different language conditions for the same story/context
    - enabled a logical structure that led participants through a complex subject and allowed them to build up an increasing knowledge rather than jump from one area to another
    - used content that kept the participant’s focus on the subject of the research
    - avoided questions that were unnecessarily intrusive in the amount of personal information requested
- Use of a translation process which achieved linguistic equivalence and face validity, necessary for an exploratory questionnaire
- Functionality of the technical aspects of the tool that:
    - provided participants with flexible and easy access
    - captured and store their responses securely
    - provided easy and secure access to the responses by the researcher
    - left no identifiable information on a participant’s computer

6.3 Limitations of tool and development process

This was a feasibility study built upon the potential that could be seen from previous studies that had started to look at digital methods for research and service provision. It was possible to hypothesise some of the potential benefits by extrapolating from at least anecdotal evidence that Deaf people have whole-heartedly embraced digital technologies (Swinbourne, 2016). However, it has not yet been explored whether this was a resource that Deaf people would want and use. There is certainly the concern that use of such technology could introduce a recruitment bias towards users who are more comfortable with it, as may have been the case with some published studies (Bowe, 2002). However, by developing such a tool and proving that it could work, it does give another avenue to explore.

A further concern is in what way the data collection method will influence how participants respond and therefore the information they provide. This will always be the case (Robson, 2002), though new technologies such as the online tool perhaps focus the attention to a greater extent on this issue. Two potential issues related to this challenge can be considered from the use case developed here. Firstly, participants are asked to work alone on a complex topic about which many may have little experience. This may have benefits in that they can consider the content with fresh eyes and fewer preconceptions so could give a more objective view. However, data collected through interaction with either an interviewer or, for example, in a focus group, may have generated more insights or in-depth reflection as a result of challenge to a participant’s views or exposure to others’ ideas. In the recent RCT terminology paper (Young et al., 2016), focus groups had discussions about how the lexical item used for randomisation could be modified to show, for example, randomisation of individuals or groups; the number of trial arms to which the individuals/groups were randomised; who was carrying out the randomisation. It is debatable whether such complexity would emerge without interaction with others.

The second issue is whether using the video capture tool itself may influence how a participant responds. Not only are they responding alone to a complex subject, as detailed above, but are signing to the blank wall of a camera lens, not knowing how their response would be received. This could potentially make someone self-conscious or less willing to say everything they thought.

One particular limitation during development was the pilot, given that the six Deaf colleagues who assisted with this were all University graduates with good bilingual English skills and familiarity with computers. They were therefore not representative of the range of backgrounds, communication preferences and skills, and education in the broader Deaf communities.

6.4 Further research on and using the online data collection tool

So far, I have pointed out the technical challenges, the rate of change of the technologies, but also the fact that such a tool is feasible and apparently in demand. Taking these issues into account, there are three potential directions in which the development work on the online data collection tool could be taken.

The first is to develop an off the shelf tool that is futureproof for a longer period. Some methods of collecting qualitative data may be preferable in terms of real life interaction e.g. interviews, focus groups. However, as well as Deaf BSL users, there are other groups where an online tool may be a valuable addition to the options available. Such groups may have one or more of the following characteristics: a low incidence of the inclusion criteria across the general population, geographically widely dispersed, very comfortable with online technologies e.g. children and young people, situations where non-written data is sought e.g. languages without a written form or participants who do not have good written language skills.

Given the technical advances, a better approach may be to use the principles identified and start again with new constituent components. This approach could aim to produce a standalone tool rather than a research group having to adapt it each time within their IT environment. A streaming server would be required, located ideally in an organisation with high level security already in place, such as a University or NHS institution. Such a tool would encompass the fundamental functionality of questionnaire creation, presentation in a signed language alone or with a written language, response capture in a signed language alone or also in a written language, and multiple choice responses. An early step would be to identify any additional useful functionality. For example, one researcher asked if data presentation and capture could be synced so that a single button started playback and recording simultaneously. This addition would allow psychological research into speed of information processing and reproduction. Another group wondered about using the data capture tool to facilitate an online discussion thread that allowed video comments throughout. Although the speed of IT development is such that a new tool would have to work hard to remain current, the fact that there is not yet any commercially available or published tool with similar functionality suggests it would be a valuable investment of time.

It has not yet been explored how the means of data capture and the remove from the researcher may affect the data content. The second suggested direction for research is therefore to investigate this relationship. Such research may not only include participants' objective views, but a qualitative evaluation of how participants present themselves to video in response to questions, to an interviewer in real life or to a video diary. For example, factors such as whether a participant is inhibited or liberated by presenting themselves direct to camera could be explored.

Linked to the above, the third area to explore would be how such digital and online technologies are impacting on Deaf BSL users. There is still little published work about the Deaf community and these technologies, though there is an increasing body of work about young people and digital media as a means of meeting preferred communication modes and styles (Gainer, 2010; Pirbhai-Illich, 2010; Bowers-Campbell, 2011; McLean, 2010) and there may be transferable insights.

6.5 Contribution to practice

This tool was developed in response to a perceived need to broaden the routes by which Deaf BSL users can engage and contribute. It demonstrates that some potential preferences (in this case, online engagement in a preferred language) may not be available for Deaf people in some circumstances. It was solved here with respect to a research task, but could also be solved with respect to clinical engagement. For example, providing a means of live data capture in BSL when contacting a hospital could be a type of engagement that utilises Deaf people's strengths and preferences and is complementary and parallel to the recent rise in visual relay services (Mager, 2014) and next generation text (Next Generation Text Service, 2016). Such an online tool could, for example, act as a BSL equivalent of an answerphone or a 'contact us' page on a website – allowing BSL users to leave a message in their preferred language rather than in spoken or written English.

6.6 Contribution to education

The online tool also has potential for use in an educational setting. During the development period, the IT colleagues, all of whom spend much time developing web-based teaching and online hosting of lectures, were excited by the idea of embedding the Flash capture tool in a website for students (hearing as well as Deaf) to record presentations for later review by their tutors. It could also be used for evaluation questionnaires, for example. As an alternative to an online questionnaire in written English, a similar tool could administer the questionnaire in a spoken (or signed) language and a student could respond likewise. This modality would have the advantage of giving extra information between lecturer and student through body language, tone of voice and facial expression. The previous discussions about changing technologies still apply and it may be necessary to return to the identified principles and re-evaluate current technologies at the time.

6.7 Contribution to policy

The rights of an individual to equal access to information within health services, both fixed information and through consultation, are supported by legislation (e.g. UK Government, 2010; UK Government, 2011; United Nations, 2006) and guidance (NHS England, 2015b; NHS England, 2015a). The latter two documents are particularly relevant to this thesis: the Accessible Information Standard (NHS England, 2015a) includes requirements for health and social care providers to ensure that ‘patients, service users, carers and parents with information and / or communication needs related to or caused by a disability, impairment or sensory loss have these needs met.’ (ibid. page 28 requirement 26) and ‘patients, service users, carers and parents with information needs (a need for information in a non-standard print format) are sent or otherwise provided with information, including correspondence, in formats which are appropriate, accessible and that they are able to understand.’ (ibid. page 28 requirement 27). These requirements have been in place since the end of July 2016. The tool developed here and the principles underpinning it may therefore be of value to policy makers considering how health services can meet these obligations.

6.8 The effect of technical developments on similar studies

6.8.1 Internet speed

Many issues tackled through this development e.g. the trade-off between resolution, frame rate and frame size, arose because 2009-2010 was a time on the cusp of the potential of video-streaming being exploited, so broadband speeds were still a major limiting factor. In many UK settings, they now no longer exist as essential barriers in this kind of development, though in more rural locations, home broadband connections may still struggle to stream video. In addition, such barriers may well still exist in some international settings e.g. in some low and middle-income countries. Internet speed should therefore still be a consideration during similar development work.

At the time this issue signaled the importance of developing experiential guidelines for website settings in signed languages, which may well be different to those for spoken language. For the purposes of this study, I tested the different frame capture rates and agreed that 15 fps (frames per second) gave a clear and understandable recording of

BSL. Despite advances in internet speed, this issue remains of relevance and should be tested before further use of a similar tool.

6.8.2 New hardware and software

Given their ubiquity now and users' familiarity with them, new technologies likely to feature in future tools are applications allowing visual conversations (in signed or spoken languages) that are incorporated as standard in smartphones and handheld devices. Some have inbuilt ways of recording their conversations. In other situations, screen recording software such as Camtasia (Techsmith, 2016) could be utilised in a research setting. Any new technology would still need to be considered with respect to the security principles identified in this study and backed up by the need to meet legislation, such as, in the UK, the Data Protection Act (1998).

6.8.3 Security and confidentiality

There continue to be concerns about the security of data transferred via the internet and this is especially pertinent when video data, essential for transmission of a signed language, removes the possibility of anonymous participation in an internet-based study. The data capture method developed here is likely to be one of the most secure solutions currently possible, but there would be a need for the current position with respect to security (both in increased security technologies, but also in the abilities of hackers to potentially access such data) and data protection legislation to be revisited each time a similar tool was used.

6.9 Conclusion

This study has presented the underlying principles and the development of a remote, asynchronous, secure, online questionnaire tool that can allow BSL users to contribute to research or service development or, indeed, to engage with services for other purposes, in their preferred language(s). There are technical and ethical challenges to such an approach, but it has been proven to be feasible and have potential for future development.

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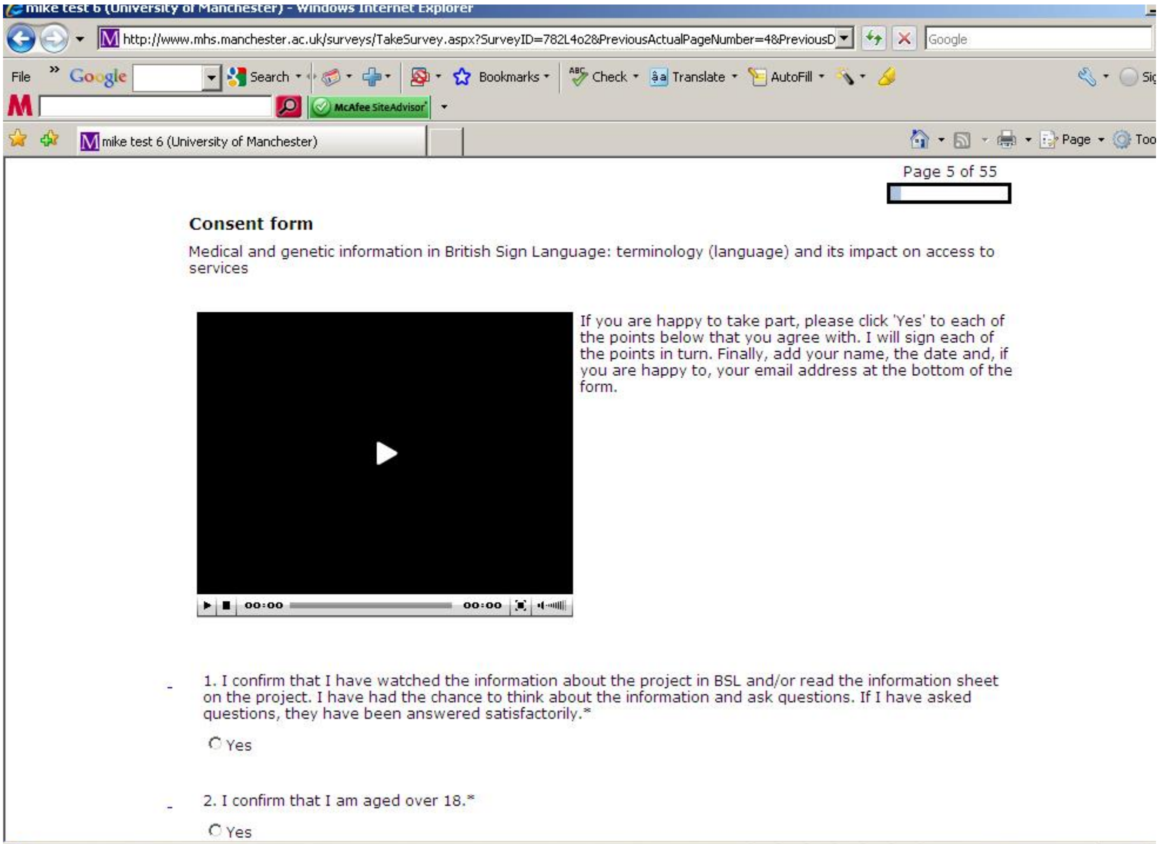
Appendix 1: Online consent form and English translation of instructions for questionnaire completion



Page 5 of 55

Consent form

Medical and genetic information in British Sign Language: terminology (language) and its impact on access to services

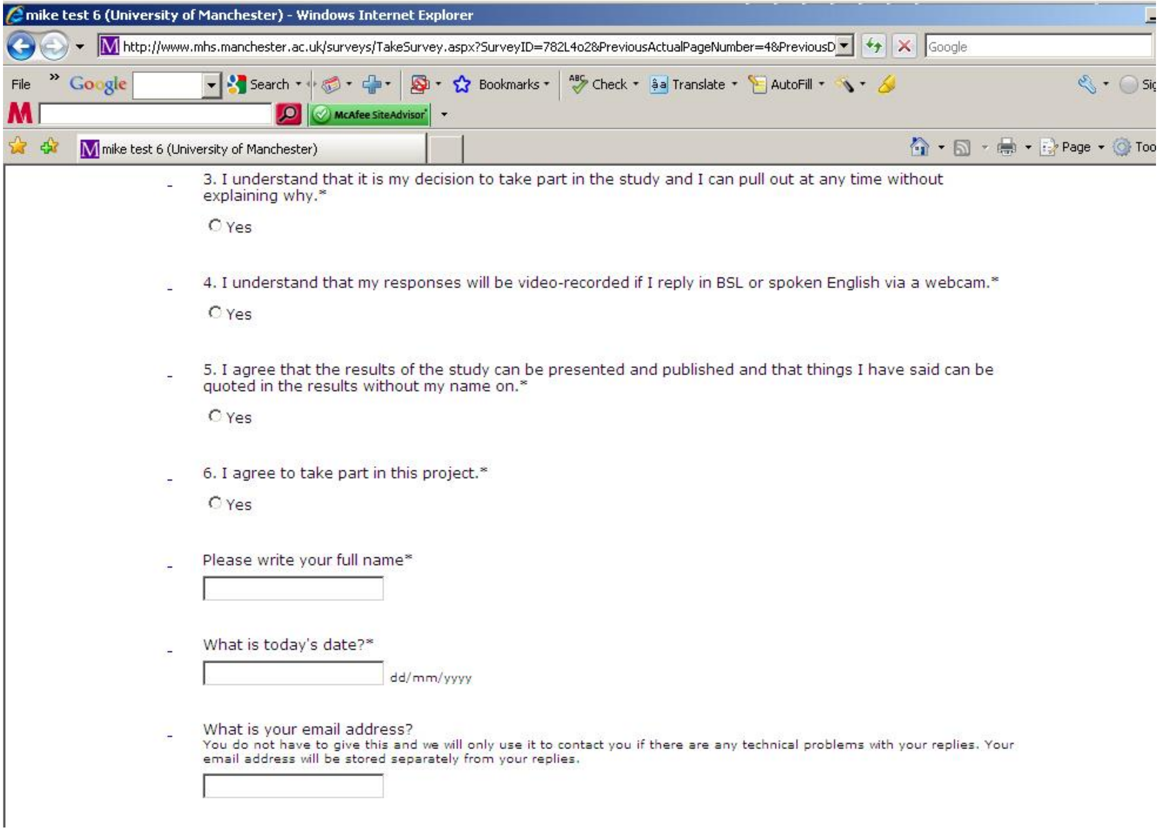
If you are happy to take part, please click 'Yes' to each of the points below that you agree with. I will sign each of the points in turn. Finally, add your name, the date and, if you are happy to, your email address at the bottom of the form.

1. I confirm that I have watched the information about the project in BSL and/or read the information sheet on the project. I have had the chance to think about the information and ask questions. If I have asked questions, they have been answered satisfactorily.*

○ Yes

2. I confirm that I am aged over 18.*

○ Yes



3. I understand that it is my decision to take part in the study and I can pull out at any time without explaining why.*

○ Yes

4. I understand that my responses will be video-recorded if I reply in BSL or spoken English via a webcam.*

○ Yes

5. I agree that the results of the study can be presented and published and that things I have said can be quoted in the results without my name on.*

○ Yes

6. I agree to take part in this project.*

○ Yes

Please write your full name*

What is today's date?*

dd/mm/yyyy

What is your email address?

You do not have to give this and we will only use it to contact you if there are any technical problems with your replies. Your email address will be stored separately from your replies.

[Participant would be able to print off consent form at this point]

How to record your replies to the questionnaire

Page layout

You can print out a written English summary of these instructions by clicking on the link under this screen. This might be helpful for you to refer to at any time.

Throughout the questionnaire, the page layout will remain similar. So there will be this video screen at the top with me, the narrator, explaining things and asking the questions. Sometimes the screen will show other information about genetics or scenarios to explain more. Next to the screen will usually be the English translation of what I am signing. Occasionally, the English translation will be written below the video screen because there are other video clips to play in a menu to the side of this screen. This page is like that because the extra clips on my left give you more information about recording your replies.

For the scenarios, you can access a PDF of the English translation by clicking on the link under the screen. You can print this out if you want. On some pages there is other written information that you can access as a PDF by clicking on the link under the screen.

Below this section of the page with presented information are two more screens side by side. Look lower down this page to see what I mean. These are the reply screen and the preview screen. Underneath them is a box you can type in.

How to connect your webcam and record your replies

If you are going to reply in BSL or in spoken English, your webcam needs to be connected to your computer now. Do not switch on your webcam in another programme at the same time – it will not work for this website simultaneously. When you plug it in or if you click on the [Record] button underneath the reply screen, a pop-up box will ask you for permission to access your webcam. Click on [Allow]. If this box does not pop-up, use the [Back] button at the bottom of the page to go back a page, plug in your webcam and then click on the [Next] button to come back to this page.

When you have clicked on [Allow], the webcam picture should appear in the left-hand screen. The picture should automatically be the right size. If there are any problems with the webcam picture, please email Rachel at the address genetic.signs@manchester.ac.uk before you go to the next page and start the questionnaire. She will try and reply very quickly.

If you want to reply in spoken English, it should automatically pick up your microphone. If you are not sure if the microphone is working, please do a test recording – next I will explain how – and email Rachel before you start the questionnaire so she can check your test recording.

To record yourself, just click the [Record] button under the screen. You will know it is recording because the button will change to a [Stop] button. When you have said everything you want, click on the [Stop] button. That is all – it has automatically been saved securely on the University of Manchester server. If you want to check what you have recorded, you can click on the [Preview] button under the righthand screen and it will play back what you have recorded. The [Preview] button will change to a [Stop] button while it is playing. You can watch it all or you can stop it.

If you want to reply in written English instead or if you want to add some extra information in written English as well, you can type in the box underneath the video screens.

If you are happy with your video reply, that is all. If you want to re-record it and copy over your first recording, you can do this by clicking on the [Record] button again. Alternatively, if you want to keep your first recording, but add some extra information to it, click on the little box that says ‘Add more to my answer’ before you click on the [Record] button. This will then add an extra bit to the end of your first recording.

If you would like to, have a try with your webcam and the controls now. Rachel will not use the practice recordings that are made on this page.

Some other things to know about

At the bottom of every question page, there is a line that says 'Please click here when you have finished your answers on this page’. You have to click [Finished] before you can click [Next] to go to the next page. You can use the [Back] button to go back to a previous page if you want to watch something again or change anything about your answers on that page.

A few of the questions have different sorts of answers where you have to click on a choice from several options, but these are clearly explained when you get to these questions.

If you need to stop halfway through the questionnaire, your answers will already be saved. When you come back to the questionnaire later on the same computer, click on the link you originally used to access the questionnaire and it should take you back to the page you had got to as long as you come back within 30 days. To be very sure you can easily get back to the same page, you can use your internet browser to save the page in ‘favourites’. Don’t worry if you leave the computer for a few minutes and it logs you out or if you click the link to come back to the questionnaire later and it does not take you back to the same page. Your answers are already saved. It just means that you will have to click [Next] at the bottom of each page to go through to where you were before – you would have to fill in the consent form again and click [Finished] at the bottom of each question page you have already answered.

Appendix 2: Members of Research Advisory Group

Mushtaq Ahmed is a Principal Genetic Counsellor for the last 16 years in Leeds. He has predominantly worked with Pakistani families with recessive conditions. Deafness in the Pakistani population has been his major workload. He has a research interest in the psychosocial aspects of genetics, particularly in the Pakistani population. He is currently conducting a DoH-funded questionnaire study with Pakistani parents of children with genetic conditions and their relatives. This study is looking at people's attitudes towards carrier and prenatal testing and termination of pregnancy for thirty genetic conditions, including deafness.

Maria Bitner-Glindzicz: I am a clinical and molecular geneticist with an interest in genetic deafness. I hold a weekly genetic clinic for families, based at Great Ormond Street Hospital and the Royal National Throat Nose and Ear Hospital, as part of the North East Thames Regional Genetics Service. I also hold a monthly multidisciplinary Dual Sensory Impairment clinic, together with hearing, balance and ophthalmology specialists, at the National Hospital for Neurology and Neurosurgery. I work together with the North East Thames Regional Clinical Molecular Genetics diagnostic laboratory to provide a quality assured molecular testing service for deaf individuals and their families. I also lead a group researching causes of genetic deafness, both syndromic and non-syndromic.

Lynne Clarke: BSL/English interpreter

Steve Emery: Research Fellow, Heriot-Watt University. Steve was awarded his PhD in Citizenship and the Deaf Community in 2007, and his BA(hons) in Cultural Studies in 1992. He is also a qualified counsellor and has held many posts in the Deaf Community. He is currently working on research into the attitudes of Deaf people to genetics and genetic counselling and the mental health needs of Deaf people from Black and Minority Ethnic Communities in Glasgow.

Claire Haddon (BSL/English Interpreter): Qualified BSL/English interpreter with a first degree in Experimental Psychology. Work primarily as a translator/interpreter and have particular interest in language/translation issues and interpreting in academic settings.

Valerie Leach is a qualified Social Worker who has worked in local authorities with all client groups for many years. She has extensive experience working with deaf children and adolescents in mental health settings. She is currently working part time with the in-patient unit at Springfield Hospital, London. Her special interests are Child Protection and Communication issues at places of work. Her leisure pursuits are rambling, visiting historical places and flyfishing!

Marion McAllister is a genetic counsellor and MRC Post-doctoral Research Fellow at the University of Manchester. Her research interests focus on outcome measurement in clinical genetics services, and her present research is to develop a measure of "empowerment" for use as an outcome measure for clinical genetics services. Marion's methodological expertise includes qualitative methods (grounded theory) and psychometrics / development of health measurement scales.

Katherine Payne: I am a senior research fellow in Health Economics, Health Methodology Research Group, The University of Manchester. I currently hold an RCUK academic fellowship in health economics to focus on the economics of genetics technologies. I have particular interests in the evaluation of genetic-based diagnostics, including pharmacogenetic tests, and the valuation of preferences.

Hilary Sutherland: Freelance Researcher Consultant for the past sixteen years working on various projects concerning deaf children and their families. Currently I am working on NDCS Deaf Role Model project covering Northern Ireland, Wales, North West and South East of England.

Noel Traynor: I have a wide range of experience working with the deaf community from young to older generations. I currently work as Chief Executive at Manchester Deaf Centre and am particularly interested in partnership working with other organisations to look at research that will identify gaps within the deaf community and influence changes needed for improvement.

Dorothy Trump: Prof Trump has a longterm interest in genetic conditions affecting vision and more recently in those causing deafness. She runs specialised regional paediatric eye, retinal and deafness genetic clinics in collaboration with opthalmology consultants and audiological physicians. In the past her research has concentrated on molecular analysis of genes and proteins involved in genetic retinal conditions and more recently Prof Trump has initiated a project investigating the characteristics and genetic input to auditory neuropathy, a recently recognised form of congenital deafness. Her research group comprises a university Research Fellow and six research workers (post doctoral, graduate students and undergraduates). Current funding is from the MRC and Wellcome Trust and there are close links with the clinical genetics department and Nowgen, the North West Genetic Knowledge Park. The development of the genetic service through the genetic deafness clinic and links with audiological physicians led to the DVD translation project and the newer programmes of research such as auditory neuropathy.

Jenny Wilkins (nee Beech): Jenny has worked in the Further and Adult Education sector for the past 10 years delivering training courses such as Literacy and Numeracy to Deaf students, NVQ Levels 3 and 4 BSL, NVQ 4 BSL / English Interpreting workshops and Deaf Awareness, and is a qualified Assessor and Internal Verifier. She is a native BSL user from a Deaf family and has been involved in various posts in the local Deaf Community both professionally and socially, and has worked for and with a wide range of services, forums and user groups such as the NDCS, MRAD, and Birmingham Deaf Forum. She was involved in the pilot translation of written genetic information into BSL onto DVD (as one of the translators working with Rachel Belk). Prior to working in the FE sector Jenny worked as an Assistant Forensic Scientist in the DNA Database and holds a degree in Biomedical Technology. Currently Jenny is a Deaf Community Fieldworker on the British Sign Language Corpus Project, DCAL, and a part time Lecturer at Walsall College.

Alys Young: Prof Young's current research programme has 3 major strands: families, deaf children and associated services; the evaluation of professional practice with particular emphasis on service user involvement; multi professional working; theoretical

work in methodology and D/deaf studies. Current grants held: DfES funded national evaluation of the Early Support
Programme (PI); DfES/NDCS funded project on Informed Choice and families with deaf children (PI); Health Foundation funded project on service users with aphasia working as service providers (PI); Big Lottery funded longitudinal outcomes study of early identified deaf children and families (CA); HTA funded RCT of intensive speech and language therapy for patients with aphasia (CO). Other recent projects include: NDCS funded study of parenting and deaf children (PI); DH funded study of the implementation of best practice standards in social work with D/deaf adults (PI); DH funded national evaluation of the implementation of universal newborn hearing screening in England (CA); JRF funded project on Deaf/hearing professional practice (PI). Prof Young is on the editorial board of two journals 'The Journal of Deaf Studies and Deaf Education' and 'Deafness and Education International'. She is currently writing, under contract with Oxford University Press, a book on methodology for their Deaf Studies Handbook series. She was previously visiting international scholar at National Technical Institute for the Deaf in the USA.

Maria Gascon-Ramos, Research Associate, Audiology and Deafness Group, University of Manchester
Yasmin Kovic, Safeguarding Advisor for Deaf Children and Young People, NSPCC
Anna Middleton, Consultant Research Genetic Counsellor, University of Cardiff
Graham Turner, Chair of Interpreting and Translation Studies, Heriot-Watt University