Considering consent: an analysis of factors influencing parental perceptions of decisional quality in the context of newborn screening

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Abstract

This thesis explores decision-making and perceptions of decisional quality in parents whose children have undergone newborn bloodspot screening. Newborn bloodspot screening is the programme through which newborn babies are screened for a variety of conditions shortly after birth.

In the UK babies are screened for phenylketonuria (PKU), congenital hypothyroidism (CH), sickle cell diseases (SCD), cystic fibrosis (CF) and medium chain acyl-CoA dehydrogenase deficiency (MCADD), with Duchenne muscular dystrophy (DMD) offered as additional screen in Wales. Much has been written about the applicability of consent to newborn bloodspot screening, yet research has tended to revolve around parental knowledge and information provision. These studies say little in terms of actual or perceived decisional quality or whether parents are making an informed choice. Taking an exploratory sequential mixed methods approach, the present study aims to identify and model factors that influence parental perceptions of decisional quality within the context of newborn bloodspot screening.

The thesis draws on two studies; an exploratory study of parental experiences of newborn bloodspot screening using semi-structured interviews, and a subsequent quantitative phase which analysed data collected through a postal questionnaire. The results of these studies provide significant insights into parental decision-making. Attitudes toward medicine were shown to have a significant causal influ-

ence on perceived decisional quality through its indirect effect on parental attitudes towards screening. Through the disaggregation of these general and specific attitudes, the significant role of perceived choice is identified. Perceived choice is demonstrated not only to be a significant contributing factor to the perceived quality of decision made, but is also shown to have a strong influence on attitudes towards screening through an indirect and positive relationship with perceived knowledge of screening. Both of these elements suggest that the context of screening and its presentation are key determinants of parental decisional quality.

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Declaration

I confirm that this thesis is the sole work of Stuart Nicholls.

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Mairi Levitt

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Chapter 1

Contexts and concepts

In 2003, and following the euphoria generated by the human genome project, the UK Department of Health White Paper *Our Inheritance Our Future* was published. This paper detailed numerous projects to support the development of research into medical genetics. The paper also documented support for the expansion of newborn bloodspot screening. This was soon followed by reports from the British Medical Association (BMA), Human Genetics Commission and the Nuffield Council on Bioethics, all of which supported the notion that newborn screening was an area of increasing interest. Since the publication of the Department of Health White Paper screening has been expanded within the UK and internationally. Recent reports have also suggested that private companies are now considering the possibility of using newborn bloodspots to analyse whole genomes, with one company predicting that this will be feasible by 2019 (Henderson, 2009). So what is newborn bloodspot screening, and why the interest?

1.1 Newborn bloodspot screening

Newborn bloodspot screening (herein newborn screening) is the programme through which newborn babies are screened for a variety of conditions shortly after birth. Screening differs from a programme of targeted testing on the basis that there is no known risk of disease within the defined population (UK National Screening

Committee, 2000).

The advent of newborn screening is credited to Dr. Asbjørn Følling who discovered and defined Phenylketonuria (PKU), the first condition for which screening was conducted (Paul, 1997; Centerwall and Centerwall, 2000). PKU is an autosomal recessive disorder resulting in phenylalanine hydroxylase deficiency that in turn leads to an increased amount of phenylalanine in the blood. If left untreated this carries a high risk of mental handicap (Anon, 1999). Early testing in the UK used ferric chloride to test urine from babies and soon became known as the "nappy test" (Sharrard and Pollitt, 2007). In 1959, the health visiting service in Birmingham undertook a limited programme to test the urine of all babies. This revealed four positive tests in 19,000 babies. Following on from this, health visitors in Birmingham continued to test all babies in this way (Raffle and Gray, 2007).

Urine testing was inefficient and improvements were sought. In 1961 Dr. Robert Guthrie published his results of the agar diffusion microbial assay, a test that improved both the speed and efficacy of screening through the use of blood as opposed to urine. However, the UK continued to use urine testing until 1965 when, in the South East of Scotland, ferric chloride testing of urine was replaced by the taking of blood via the heel prick and the application of the Guthrie test (Douglas et al., 1968). At the same time the Medical Research Council set up a Working Party to consider the issue of newborn screening and in 1969 the Department of Health and Social Security issued a circular endorsing the use of the Guthrie test for a national programme of newborn screening for phenylketonuria (Simpson et al., 1997).

1.1.1 Screening in the UK after Guthrie

Despite the 1969 Department of Health and Social Security circular, newborn screening for conditions other than PKU continued in a piecemeal fashion. In

1981 the Department of Health and Social Security added congenital hypothyroidism (CH) to PKU as part of the national screening programme. At this time it also recommended that a national committee be established to co-ordinate the regional screening programmes for CH (Joint Standing Sub-Committee on Screening in Medical Care, 1981).

There was still a lack of co-ordination, with regional health authorities implementing their own programmes in an ad hoc manner. In 1996 the Department of Health established the National Screening Committee (NSC) to oversee all national screening programmes, including the newborn bloodspot programme (The Nuffield Council of Bioethics, 2006). Its remit would be to advise on whether programmes should be initiated, continued or withdrawn.

In 2001 it was announced that national screening for cystic fibrosis would begin, 25 years after it was first introduced in Leeds, with screening for sickle cell anaemia to begin in 2004. Alongside this, in 2002, the Department of Health funded a UK Newborn Screening Programme Centre (Holland et al., 2006). Today the UK National Screening Committee recommends that all babies are offered screening for phenylketonuria (PKU), congenital hypothyroidism (CH), sickle cell diseases (SCD), cystic fibrosis (CF) and medium chain acyl-CoA dehydrogenase deficiency (MCADD) (UK Newborn Screening Programme Centre, 2008b, p2).

1.1.2 UK Policy and Practice

As part of the national screening programme every resident newborn baby and those under the age of one who enter the UK, should be offered blood spot screening (UK Newborn Screening Programme Centre, 2008b). This achieves almost universal uptake (Bradley et al., 1993; Simpson et al., 1997; Parsons et al., 2002; Pollitt, 2004). The process through which this occurs for newborn screening is shown in Figure 1.1.

From the schematic one can see that in all cases a record should be maintained regarding the parents' decision and that this can be one of three options; to consent to all, some, or none of the screening tests. As can be seen, informed choice is an integral stage (UK National Screening Committee, 2000; Campbell and Ross, 2004; Kenner and Moran, 2005). This is spelt out clearly in the National Screening Programmes own literature which states that:

> "It is important to offer parents an informed choice about screening for their baby, to gain consent and to prepare them for the blood sampling procedure." (UK Newborn Screening Programme Centre, 2008a, p2).

Consequently, screening can only proceed on the authorisation of the parents and this authorisation should be made on the basis of an informed choice.

1.2 Consent and decision-making

In the newborn screening context the term informed choice appears to be used somewhat liberally and may be equally referred to as "informed decisions" or "informed consent". Although definitions do vary (Gillon, 2001; Beauchamp and Childress, 2001; del Carmen and Joffe, 2005; Dawson and Spencer, 2005; van den Berg et al., 2006; Elwyn et al., 2009), the core aspects are often very much the same. Firstly there are the requirements that the individual giving the consent must be competent to do so; secondly, the consent shall be given voluntarily; thirdly, the giving of consent must be based on relevant information, and finally a decision is made. Consequently the process of decision-making and what informs this process is central to any concept of informed consent.

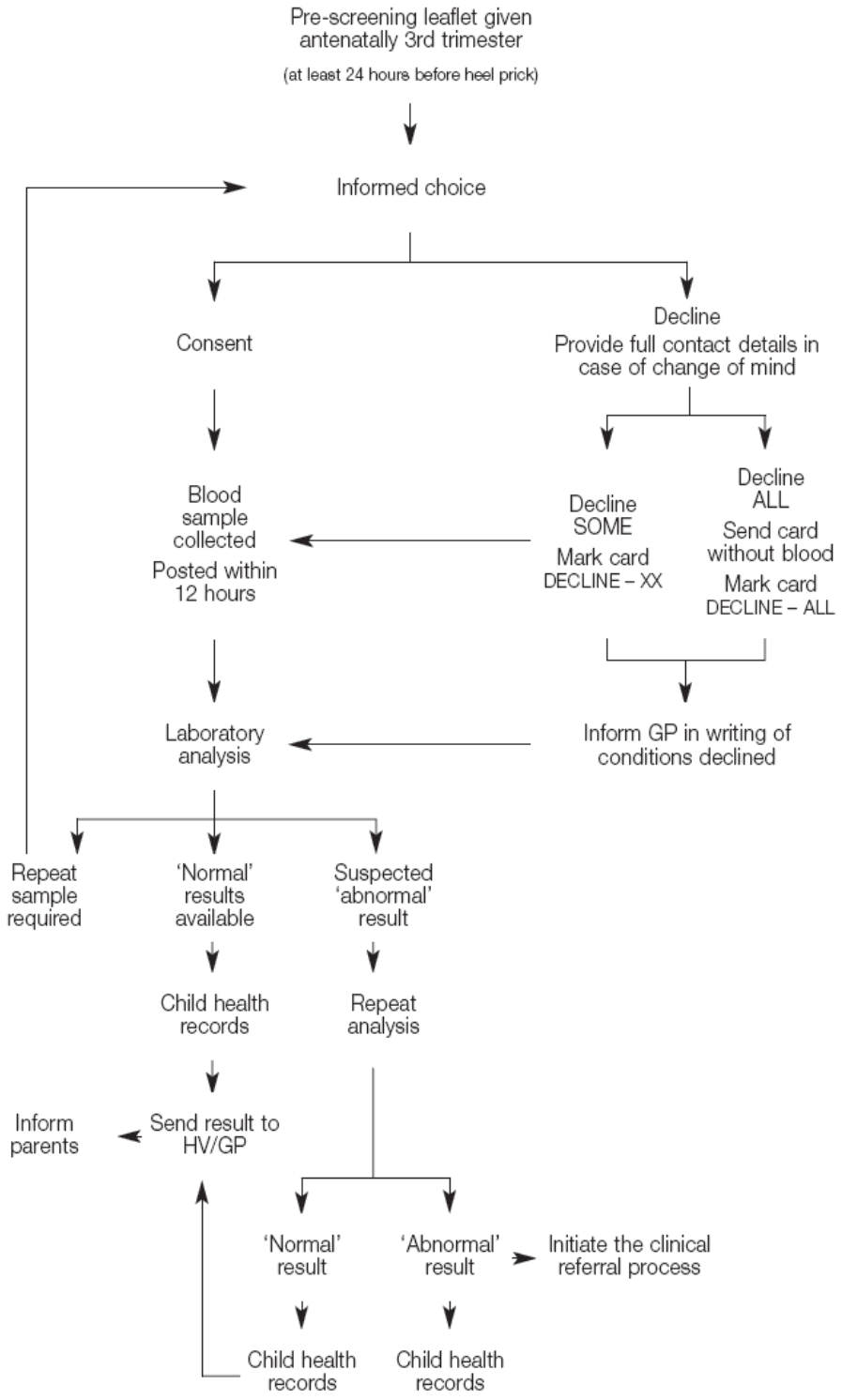

Figure 1.1: *The newborn bloodspot screening process in England, taken from the Newborn blood spot screening in the UK Health professional handbook, April 2005, p18*

1.3 Informed choice for screening in England

Whilst newborn screening has been in operation for a substantial period of time and has a high uptake rate, little is known about the decision-making process of parents when accepting newborn bloodspot screening. Research has tended to revolve around parental knowledge (Smith et al., 1990; Tluczek et al., 1992; Zeuner et al., 1999; Tluczek et al., 2005; Lang et al., 2009) and information provision (Hargreaves et al., 2005b; Fox, 2006). However, these findings say little in terms of parental decision-making or whether parents are making an informed choice.

Assessment of knowledge based on recall at best serves only to give evidence of the individuals' memory (Beauchamp and Childress, 2001); indeed the converse may be true. The ability to recall information, says nothing as to whether that information was used in the decision-making process and therefore whether the decision was informed (Bekker et al., 1999). Research into leaflet content has found that many "did not facilitate realistic expectations or support the informed choice agenda" (Hargreaves et al., 2005b, p116). Yet this again says nothing as to whether the leaflets are used by parents when making decisions. The dearth of research means that there is a lack of empirical data about how parents make decisions regarding whether or not to consent to newborn bloodspot screening. The implication of this being that:

> "There is a general lack of both procedures, and research to inform the development of such procedures, for:
>
> - providing parents with information about the newborn bloodspot screening
> - inviting informed consent for newborn bloodspot screening
> - routinely informing parents of the results (positive or negative)
> - explaining to parents the need for further tests

- addressing the potential for misunderstanding by parents of the test results; and
- understanding and addressing the particular difficulties raised by revealing carrier status." (Stewart and Oliver, 2003, p2)

Confirming this, a systematic review of the psychosocial implications of screening found that whilst newborn screening for phenylketonuria is an international test that has been established for over forty years:

> "...we found no studies which looked at parents' responses to the process or results of PKU screening and only a very small number looking at knowledge and attitudes to Guthrie testing in general" (Green et al., 2004, p57).

As such we know little as to how parents feel about the decisions they made, whether they feel they have made an informed choice, or what factors affect these assessments.

Measures such as the Multidimensional Measure of Informed Choice (MMIC) (Marteau et al., 2001) do exist in other contexts but suffer from methodological or theoretical issues. The MMIC, which has been developed for prenatal screening, contains recall-based knowledge components and so may fail to appropriately capture parental understanding. The findings within the prenatal screening literature may not be relevant to newborn screening. A systematic review of different screening programmes found that the important determinants of uptake varied depending on the type of screening test (Jepson et al., 2000). Termination of pregnancy, for example, has been found to be a highly significant influence in the context of prenatal screening (Potter et al., 2008), yet is not a salient factor in the context of newborn screening.

Audit data may be suggested as a proxy for explicit research data; for example, audit data may include factors that are associated with the level of completeness of coverage of newborn bloodspot screening. This may provide useful insights into factors that affect uptake of newborn bloodspot screening but provides no information about actual decisions taken by parents. Instead they may be documenting administrative failings as much as actual refusals. Equally, the low level of refusal - which may constitute only a handful of parents - is detrimental to any statistical modelling or accurate quantitative analysis of actual uptake. Instead a more appropriate process is to consider the reflections of parents on the decisions taken. This not only provides insights into the decision-making process but potentially generates variability in response allowing statistical analysis and the identification of significant variables which may impact on these decisions.

1.4 Motivation

Thus, whilst there is an increasing interest in and expansion of, newborn bloodspot screening, there has been little research focusing on the effects on parents or the factors which influence parental decisions. Furthermore, research in other contexts may not be relevant to newborn screening. There is, therefore, an opportunity to create a better understanding of not only whether parents are making high-quality decisions but also the factors that influence these decisions.

1.5 Aims

This research aims to identify and model factors that influence parental perceptions of decisional quality within the context of newborn bloodspot screening (NBS).

1.6 Objectives/Research questions

- What are the factors that parents identify as affecting their decision to consent to newborn bloodspot screening (NBS)?
- How are these factors related to their decision-making?
- To what extent do these factors influence the perceived quality of the decision?

This research will provide an insight into whether parents feel they are making high-quality decisions when accepting or declining newborn bloodspot screening and the factors which influence these perceptions. The results of this work will be valuable in developing informed consent practices.

Chapter 2

Decision-making and newborn bloodspot screening: what do we know?

In the opening chapter newborn bloodspot screening was introduced together with the motivation for this study; a lack of information about whether parents are making informed choices or the factors that affect parental decisions to accept newborn bloodspot screening. Informed choice, within the documentation provided by the Newborn Screening Programme Centre, can be equated to informed consent which itself is often decomposed into a number of elements which can broadly be described as either informational or decisional aspects (Beauchamp and Childress, 2001; Gillon, 2001; del Carmen and Joffe, 2005; Dixon-Woods et al., 2007). In the remainder of this chapter I shall review the literature on newborn bloodspot screening through these informational and decisional lenses and appraise the current state of the literature regarding the existing evidence about parental decision-making.

This literature review has been confined to empirical studies within the context of newborn screening and as such will not be reviewing the existing, and voluminous,

literature regarding prenatal screening. This literature, to which reference was made during the introduction, has been extensively reviewed elsewhere[1]. Whilst it is acknowledged that this literature may overlap with that on newborn screening it was felt that the two contexts raise differing issues, particularly in relation to termination following prenatal diagnosis.

Papers were sought through a narrative review of bibliographic databases such as EMBASE, MEDLINE, and the Applied Social Sciences Index and Abstracts (ASSIA). In addition references were reviewed and author specific searches conducted in order to identify texts that would not be located in these repositories.

2.1 Informing and influencing decision-making

Decision-making is often linked to the process of information provision and collation. Research relating to information provision has suggested that parents use a range of sources to inform their decisions. A recent pan-European survey of information for cystic fibrosis newborn screening found that 53% of the programmes that responded provided information by a booklet with others providing information via a pediatrician, nurse or midwife (Munck et al., 2007). Other studies support this, in particular noting the role played by the midwife (Hargreaves et al., 2005a; Parsons et al., 2006, 2007). Other, non-medical sources of information are also sought with some research finding that prior personal experience and the experiences of family and friends were second only to the advice given by the midwife (Tymstra, 1986; Davey et al., 2005). In a Welsh study, personal experience was felt to be an influential source to such an extent that one family's negative experience of screening for Duchenne muscular dystrophy (DMD) was viewed to have been the cause of a much higher refusal rate in one particular area (Bradley et al.,

[1]A comprehensive review by Green and colleagues identified 52 papers regarding understanding and decision making for prenatal screening, the majority of which related to Down syndrome screening but few regarding newborn screening. For more information see Green J.M., Hewison J, Bekker H.L., Bryant L.D., Cuckle H.S., (2004) Psychosocial aspects of genetics screening of pregnant women and newborns: a systematic review. *Health Technology Assessment*, 8(33)

1993). Additional qualitative work found that the advice of others, including the midwife, was a key factor that affected parental decisions (Parsons et al., 2006). This information seeking from non-medical sources was emphasised in a study from the Netherlands where it was noted that

> "Most of what they had heard came from other parents, for example, during visits to see newborn babies and their parents. Several respondents said that they had read about the test in a book about pregnancy or seen something on television. They had seen little or no "official" information." (Detmar et al., 2007, p241)

Despite not receiving information through medical sources, participants in all of the groups thought that the midwife played a key role in the provision of information. This may suggest that the reference to "official" sources relates to written information, with the midwife occupying a position not explicitly linked to information provision.

Written information, whilst being received, is rarely cited as a primary source, with some studies suggesting that instead it plays a greater role as a reference material as opposed to influencing the initial decision-making (Hargreaves et al., 2005b). The low priority placed by parents on written information serves to emphasise the potential role that personal interaction plays in the information gathering process.

The reliance on midwives is an important finding if one wishes to identify the best methods of service provision, yet in a survey of four maternity services across England, midwives were found to be "not at all confident" in discussing with a couple the genetics of cystic fibrosis and their risks of having an affected child. Twenty nine percent of these respondents felt that genetics was a specialist area and should be carried out by others (Metcalfe et al., 2008). Whilst this survey was carried out prior to the development of training as part of the expanded national newborn screening programme, a lack of confidence or knowledge by midwives could lead

to misinformed or uninformed decisions being taken by parents.

While interesting from an information provision perspective the implications for parental decision-making are limited as they tell us little about what informational aspects parents act upon. For this one needs to consider the content of the disclosure and the information that parents take utilise when making their decisions.

2.1.1 Content of disclosure

Despite an extensive search of the literature, no studies were found which explicitly explored the communication process that occurs between parent(s) and their midwife or parents and other parents in terms of collecting information about newborn screening. Nor were any studies identified which questioned parents as to what information they sought. There are, however, a limited number of studies that have assessed written information provided to parents. Studies that have assessed written information provided by healthcare organisations have found that there is variance in terms of the details included and the way this information is portrayed (Loeben et al., 1998; Hargreaves et al., 2005b). One review of over 100 patient information materials from the UK, US and Australia found that there was an inherent bias towards the presentation of the benefits of newborn screening compared to the limitations (Hargreaves et al., 2005b).

If this finding is replicated in the parent(s)-midwife discussions then there may be a biased and less than optimal disclosure of information that precludes parents from making an informed decision. This, however, is impossible to assess as the published studies regarding parental experiences of newborn screening consistently fail to provide evidence about what is disclosed to parents. As a result the information giving process and the subsequent discussions between parents and professionals remains opaque. None of the studies outlining information provision

described standard practice in any great detail. In those cases where there is a brief description of the process e.g., parents are provided with a leaflet, no details are given about the content.

This is a major obstacle in attempting to get inside the 'black-box' of parental decision-making. Parents report that they rely on the midwife as an information source and as a result these discussions are potentially pivotal to decision-making. Without insight into how parents use these, or what occurs in these encounters, the process of parental decision-making remains unexplored territory.

2.1.2 Satisfaction with information provision

Perhaps the largest area of research relating to the disclosure of information is that of parental satisfaction with information, with a consistent finding that parents are satisfied with the information that they have received (Sorenson et al., 1984; Sveger et al., 1999; Sveger and Thelin, 2000; Davey et al., 2005). In particular, one study in Massachusetts noted that the satisfaction was positively associated with the perceived amount of information received (Sorenson et al., 1984). Others note that satisfaction with information was correlated with belief in the value of the test (Davey et al., 2005). Despite the consistency of these findings they need to be carefully interpreted, for studies also show that the default reaction of patients is to express a high level of satisfaction with medical care (Avis et al., 1997; Sitzia and Wood, 1997). So, whilst parents respond to questions indicating a satisfaction with the information provided, several studies also note that parents also offer suggestions for improvement, often requesting more information (Sveger and Thelin, 2000; Campbell and Ross, 2004; Davey et al., 2005)

One study which sought to assess why parents were dissatisfied with the information they received found that parents suggested three possible causes for the dissatisfaction (Parsons et al., 2007). These were:

- (a) a lack of information; that is there wasn't enough
- (b) literature overload, and
- (c) a lack of time from the midwife

The first of these relates to the lack of detail in the information they were provided. This further emphasises the need for more studies to investigate the content of the communication process. The second point is not contradictory to the first, but relates to the other literature that parents are receiving at this time, with parents reporting that they lack the necessary time to read the voluminous literature given to them (Davis et al., 2006). Finally the lack of midwife time is in part an institutional effect of midwives having to conduct a number of clinics or visits and consequently they only have a limited amount of time per case. In the study by Parsons et al. (2007) parents often experienced this as a rush and were hesitant to ask questions about the screening process. All of these have the potential to impact negatively on information provision to parents.

2.2 Knowledge, understanding and decision-making for NBS

By far and away the most expansive area of research around newborn bloodspot screening is that of knowledge assessment. This has largely focussed on parental knowledge through the accurate recall of information. The assessment of knowledge is principally in two areas: the assessment of condition specific knowledge and the evaluation of parental knowledge of the newborn screening process, with the former generally found to be better than the latter (Faden et al., 1982; Dankert-Roelse et al., 1990; Statham et al., 1993; Campbell and Ross, 2004; Davis et al., 2006).

These studies, taking place mainly in the United States, have generally assessed

knowledge through the recall of technical information and found that parents often do not know the names of conditions or their major cause (Faden et al., 1982). UK research has suggested that ignorance is widespread with regards to screening for PKU and CH (Statham et al., 1993). Two recent studies support this assertion (Hargreaves et al., 2005a; Parsons et al., 2007).

Others, most worryingly, have found that some parents have an awareness of neither the screening programme, the names of the conditions tested for, the problems associated with these, nor whether their own child has been screened or not (Smith et al., 1990; Tluczek et al., 1992; Campbell and Ross, 2003, 2004). In an Australian study with women who had received written and oral information, only 37% recognised the term 'Guthrie test', 'heel prick test' or 'newborn screen'. Most disconcerting, given that the particular State operates under a principle of 'informed dissent', was the finding that whilst 40.5% of the women stated that their child had not undergone the test, when records were checked it was found that only 2% had not undergone the test (Suriadi et al., 2004). Conversely, parental knowledge of condition specific information is generally found to be good (Al-Jader et al., 1990; Mischler et al., 1998; Lewis et al., 2006). Similar findings were reported by a group in the Netherlands that found that knowledge of recurrence risk was very good within a small cohort of patients identified as having CF, with 94% knowing their recurrence risk (Dankert-Roelse et al., 1990).

2.2.1 Reliance on recall

The majority of the studies evaluating parental knowledge have done so through the recall of information. By only assessing the recall of specific information, no assessment is made of the understanding of the information, and at best the results reflect the individuals' memory (Beauchamp and Childress, 2001). Consequently it tells us little if anything about the quality of the parental decision-making or consent.

Two examples from the literature demonstrate this distinction. A sample of Dutch parents recognised the term 'heel prick' and knew that the heel prick had been conducted, but most did so "without really knowing what the heel prick was for" (Detmar et al., 2007, p241). In a similar example, two-thirds of parents of children with haemoglobinopathy could recall the recurrence risk of having another child with a haemoglobinopathy but eight of the 15 parents interviewed could not explain the risk figure (Warren et al., 1982). In both cases the parental knowledge, through the recall of medical terminology or statistics, may be seen to be adequate yet beyond this there is a lack of any real understanding or comprehension about what these terms or figures mean for them in terms of their immediate or future health.

2.2.2 Sample issues

Many of the studies of parental knowledge have heterogeneous samples in terms of the ages of children. The heterogeneity of the samples is important as clinical practice does not remain consistent over time and so parents would experience differing clinical practice. In the study by Hargreaves et al. (2005a) the ages of the children of those who participated varied between 23 years and six weeks of age. The same issue is raised in a number of other studies although the range of ages does vary (Dankert-Roelse et al., 1990; Dudding et al., 2000; Firth et al., 1983; Muchamore et al., 2006b; Hildes et al., 1993; Merelle et al., 2003). Consequently, when asked to recall their experiences, parents would have been recalling different clinical standards, meaning experiences are not comparable. Despite this issue, the authors draw together themes without acknowledging the context of the parental experiences.

A further limitation brought about by the range of ages is the large lapses of time between the study and the parents' experiences of newborn screening. This is of

particular relevance given the prominence of knowledge assessment by recall of information. Parental recall of an event, particularly technical information, may be expected to be greater in the short term as opposed to a longer period. This not only makes it less appropriate to use the recall of information as a method of assessment - due to the differing information that may have been provided at different times and the long expanse of time between screening and research - but becomes even more pertinent if it is considered in conjunction with the salience of information. In the studies which show good levels of knowledge, such as those by Mischler et al. (1998); Lewis et al. (2006); Dankert-Roelse et al. (1990) and Al-Jader et al. (1990) the selected groups were parents of children either affected by cystic fibrosis (CF), or who were carriers of CF. A parent who has to take a child for regular physiotherapy sessions in order to help their lung function is more likely to have a good knowledge of the effects of the condition in comparison with an individual who has an unaffected child. Therefore, it is perhaps no surprise to find that when questioned about specific aspects of CF, parents of affected children were aware that the lungs were affected in people who have CF (Lewis et al., 2006). Consequently more attention needs to be paid to the sample composition, the potential sample effects, and the implications for results.

2.2.3 Parent or professional perspectives?

It has already been argued that at best recall serves to demonstrate memory and not understanding. At worst the failure to recall information may indicate a difference in acceptance of information, as Wynne (1991) argues:

> "People may appear to be unresponsive or incapable of digesting scientific knowledge (which experts consider to be important to them) when they are rejecting the scientists' agenda. Our research shows that public nonreceptivity to scientific information is often based on a judgement that it is not *useful* or does not match public or personal experience." (Wynne, 1991, p116)

Hence a failure to recall the information selected by 'experts' as important may reflect not on parents' ability to understand the information, but on their own interpretation of the relevance or usefulness of that particular piece of information.

The implication of this is that information may not be relevant to parents, and their failure to recall this may say nothing as to their knowledge or understanding but only of the salience to them. The perceived saliency of the information has been found to be influential in prenatal screening research by Lippman (1999) and Potter et al. (2008). These studies found that there was an interaction between technical knowledge, other 'experiential knowledge' and moral beliefs (Potter et al., 2008). Where mothers held a strong belief that termination was immoral, the technical information regarding risks became irrelevant as they would not consider a termination. To say that they were uninformed fails to acknowledge that the information given to them by professionals has no relevance. Consequently it would seem more appropriate to base evaluations on information that is relevant to parents and utilised in their decision-making, as opposed to professionally derived information priorities.

The existing literature suffers from a conspicuous lack of studies that have sufficiently explored parental understanding and the factors that affect this understanding. This needs to be addressed if one is to gather relevant information as to whether parents are providing informed consent to have their child screened.

2.3 Decision-making

2.3.1 Uptake and perception of choice

Uptake of newborn bloodspot screening is consistently found to be almost universal (Campbell and Ross, 2004; Therrell et al., 2006; Bradley et al., 1993; Clayton, 2005; Liebl et al., 2002; Dhondt, 2005; Simpson et al., 1997), with reported refusal

rates being as low as 0.05% (Faden et al., 1982). A published audit of data in Bath found that only 5 parents refused screening in 1996, with 7 the previous year (Simpson et al., 1997). More recent data from France found refusals for cystic fibrosis screening decreased to a low of 0.2% . Over the same period no refusals were recorded for standard screening which included tests for phenylketonuria, congenital adrenal hyperplasia, congenital hypothyroidism and sickle cell disease (Dhondt, 2005). Even with optional screening programmes for less treatable conditions such as DMD, and screening tests with less predictive value as is the case with diabetes type 1, uptake rates have been found to be high with with studies exceeding a 90% uptake rate (Bradley et al., 1993; Campbell and Ross, 2003).

One potential explanation may be that the outwardly promoted requirement of informed consent is experienced by parents as institutional rhetoric. This is supported by research which has noted that parents experience the heel prick as routine (Hargreaves et al., 2005a; Moran et al., 2007; Parsons et al., 2007). Such 'routinisation' is found internationally. Dutch research into parental experiences of screening for CH found that most parents saw the screening "as a routine business that no one ever hears about again, and that people do not give much thought to." (Tymstra, 1986, p93). The same routinised attitude was found by Detmar et al. (2007), the authors noting that screening was "more or less automatic and that consent is not explicitly requested" (Detmar et al., 2007, p241). In some cases parents have even reported that the screening process was conducted in their absence, obliterating any notion of informed consent (Tymstra, 1986; Tluczek et al., 2005).

In a Welsh study it was found that when a second screening card for an optional screen for DMD was introduced it led to a significant increase in parental awareness that the test was optional. The intervention also led to a decrease in the uptake from 95% to 91%. Despite this increase in awareness, 15% of those involved in the

intervention arm did not realise it was optional. Four percent of these felt that they did not have a choice, despite the explicit way in which consent for screening was taken. This led the authors to conclude that:

> "It would seem that, although the majority of mothers felt they were being given a choice about screening, for some it was a choice about a test they thought was always done. This, in reality, is no choice at all." (Parsons et al., 2005, p168)

Thus the ability to provide an informed consent is not merely about having the facts to make a rational choice. The findings that parents do not necessarily experience a choice may also indicate that more subtle pressures are being exerted, such as the way that the screening tests are offered, the information that is provided and the institutions or individuals that provide them. Indeed the prescription within the current UK guidelines that "screening has been discussed and *recommended*" (UK Newborn Screening Programme Centre, 2008a, p2, emphasis added) may be seen as an example of these subtle pressures, with a recommendation being seen as potentially coercive.

Countering this suggestion there is evidence that at least some parents welcome the recommendation from the midwife. Australian research has found that whilst some parents felt that they had not given a specific consent, most were not concerned by this (Muchamore et al., 2006a). UK research suggests that parents are happy to accept a routine screen that is recommended by their midwife, with some parents commenting that "If they had been offered a choice, many said they would have opted for their child to be screened." (Hargreaves et al., 2005a, p165). As a result the mere offer of a screening test may result in a sequence of events that, under the prevailing social norms, means that ones actual experience of 'choice' is that there is no choice at all.

Yet despite all the evidence regarding the routine nature of screening, there is a lack of research which explores whether parents feel they can refuse such an offer, how they experience these offers and whether they feel the recommendations by practitioners constitute an 'undue influence'.

2.3.2 What is known about decision-making?

As laid out in the preceding section, much of the existing research has considered the uptake of screening, the sources of information that parents use and the quality of this information. These studies tell us very little about parental decision-making. In this regard there is a much thinner evidence base.

What research there is suggests that the benefit to the child is paramount (Parsons et al., 2002; Campbell and Ross, 2003; Hargreaves et al., 2005a; Parsons et al., 2006; Detmar et al., 2007, 2008). This perceived benefit is indicated in the way parents respond to a scenario where there is no direct health benefit to the child; such as DMD screening, with some parents saying that they would refuse screening because of the lack of health benefits (Campbell and Ross, 2003; Parsons et al., 2006). The consistency of this finding both over time and between countries, indicates that this is normative statement and not a context specific description.

Perceptions of screening, and the potential for benefits, has been found to be mediated by a general attitude towards medicine. For those parents who offered reasons why they accepted newborn screening, a belief in research, support of screening in general, and the simplicity of the test were all given as reasons (Parsons et al., 2006). For those who refused, a wish to avoid medical intervention and a perception that the test would not aid research were also given as reasons. In another study auditing an existing neonatal screening programme, the reasons given by parents for refusal appeared to be specifically related to their opinions of screening and/or medical intervention in general. The authors noted that several of the parents who

refused had delivered at home with minimal medical input and one family refused because they thought that "the screening test was a barbaric procedure based on the experience of an acquaintance" (Simpson et al., 1997, pF231). Whilst views regarding the medical intervention were cited as central to the decision, this was heavily influenced by the experiences of friends. As a result experience is not only an important source of information but may also be important in decision-making.

The finding that the health of the child is regularly invoked as the main reason for accepting screening is consistent with the original principles on which newborn screening was founded. These criteria state that not only should treatment at an early stage be of more benefit than at a later stage but also that the risks, both physical and psychological, should be less than the benefits (Wilson and Jungner, 1968). In some instances this is seen to be of such paramount importance that parents felt that the benefit to the child outweighed parental autonomy and decision-making rights over their child. For example, in one US study it was reported that:

> "The majority of respondents supported mandatory testing because of concerns that some parents may refuse out of ignorance (six focus groups), with three groups targeting their concern at teenage mothers. As several respondents suggest, some parents may not make the right choice for the health of their child." (Campbell and Ross, 2003, p210).

Consequently, the parents felt that the benefit to the child was more important than parental rights to decision-making. There is also an important interaction to be noted. Firstly there is a supposition that there is a right choice in terms of the health of the child. The availability of this 'right' choice may explain why some parents experience the screening as routine as there is no choice to make, one only has to action the correct solution. The second is that testing is beneficial. It is at this level that the social norms may come into play as discussed earlier. The offer of a test may imply it is beneficial. Therefore, if testing is beneficial no decision

needs to be made if one has already established that conveying benefit to the child is the correct moral position.

Trust may also be an important mediating factor, with one study finding that trust in the medical community is central to the attitudes of the mothers (Parsons et al., 2007). Thus a trust in the medical profession may be a causal element in determining that screening is beneficial due to the fact that screening is offered by the medical community.

Empowerment, through gathering knowledge, may also be an important factor. For those who accepted screening in a DMD study conducted by Parsons et al. (2006) the majority of reasons for accepting screening related to themes of gathering knowledge. As with the attitudes expressed by parents towards newborn screening, much of this knowledge-seeking was related to benefiting the health of the baby. The authors concluded that:

> "Most of the reasons given [...] indicated that women were aware screening for DMD might detect an abnormality. For those who requested the test, earlier identification was seen to offer the benefits of knowledge, time to prepare and get early help and choice in future pregnancies." (Parsons et al., 2006, p711).

This is not to say that all parents desire the screening results. In the same study, a cited factor for declining screening was the desire not to know. Thus knowledge may be central to decision-making, but not deterministic of the direction of the decision. In an additional study the same authors note that "Women who refused the test were far more likely to give a reason which indicated they were aware the test might detect abnormality" (Parsons et al., 2006). Thus it may be that increased knowledge may be linked, through such an awareness of abnormality, to an increased refusal of newborn screening.

These studies suggest that there are some areas of overlap between the sources of information that parents use and the factors that influence their decision-making. Personal interaction appears to be important for parents when making these decisions and this is exemplified through the evidence that the midwife and other parents are principal sources of information. Whilst it is unclear from the existing studies what parents take from these discussions into their own decision-making, a number of factors do appear to be key. Principally the concern for the health of the child is of utmost importance, with support for screening decreasing in line with a perceived decrease in benefit to the child. This perception of benefit appears to be mediated by a general attitude toward medicine and medical intervention; which in turn appears to be influenced by experience. As such it may be that parents enter discussion with other parents and/or the midwife seeking information as to the potential health benefits or detriments for their child and it is this that they take from discussions and on into their decision making.

2.4 Limitations of existing research

From the evidence, is it possible to answer the question of whether parents are giving informed consent to newborn bloodspot screening? The answer is no because the data thus far is unclear. The analysis of parental decision making, and in particular parental consent to newborn bloodspot screening, is something that has received little attention within the literature. The small quantity of research together with methodological issues preclude any great inferences being drawn about *why* parents accept or decline newborn bloodspot screening or *how* they come to their decision.

A limitation is that many of the studies make use of hypothetical scenarios, something which may offer few insights into actual decision-making. Examples from clinical genetics, particularly relating to uptake of predictive testing, have shown that there is a notable difference in predicted and actual uptake rates (Decruye-

naere et al., 1993; Binedell et al., 1998). This suggests that inferences drawn on the basis of hypothetical scenarios may be unsubstantiated when applied to actual situations and that there may be additional factors affecting actual decision-making.

Audit data may be suggested as a proxy for explicit research data and may include factors that are associated with the level of completeness of coverage of newborn bloodspot screening. These studies indicate a number of factors that affect the likelihood that a screening result is not recorded including reduced contact with hospitals, early discharge following a short post-natal period, birth outside of hospital and less than seven antenatal visits (Gray et al., 1997; Spady et al., 1998; Metz et al., 2003; Jaques et al., 2008). Two studies have also identified ethnicity related influences (Streetly et al., 1994; Metz et al., 2003). Whilst the estimation of coverage provides useful insights into factors that may be affecting uptake of newborn bloodspot screening, the analysis is purely descriptive and in no way provides information about actual decisions taken by parents. Instead they may be documenting administrative failings as much as actual refusals. A combination of data is required; the variables identified as significant may be explored further to examine potential causal mechanisms that lead to the lack of a screening result. It may be that parents who leave hospital earlier are less willing to have medical interventions than those who stay longer, and this accounts for their lack of test result. Alternatively, hospital pressures may mean that there is an increased impetus to have parents take children home earlier so that the facilities are free for the next parent who requires them. This may in turn lead to more administrative errors resulting in missed tests. Without further in-depth analyses either of these premises are plausible.

The existing literature also suffers from methodological weaknesses. Most of the studies which have assessed knowledge have done so using a questionnaire or a structured interview. There is, however, a pervasive lack of justification for the

questions asked. There are different studies, each assessing knowledge of a genetic condition, or NBS more generally, but the findings are not directly comparable because of the differences in questions. If there is a question asking parents to recall prevalence, why is this included? If the questions used were intrinsically linked to the information provided to parents, then one could make the case for the question as a measure of recall. One would still need to justify why the recall of this specific information was important, but one could argue that parents are being provided with information and so this should be recalled. If this justification were in place one could also compare the information provided to that recalled and look for any specific patterns and these patterns could be compared across studies. Yet, this sort of information, let alone descriptions of the current information provided to parents, is not presented within the published studies.

The use of quantitative research is not in itself problematic, but it is the specific application of these research methods which is the cause of concern. For example, despite the use of quantitative research methods there is a lack of statistical analysis amongst the published literature. In many of the studies only simple descriptive statistics are used, such as percentages of responses. Whilst appearing to provide detailed quantitative data, the results are open to misinterpretation. For example, whilst Al-Jader et al. (1990) demonstrate a difference in parental knowledge of CF between higher and lower educational groups, there is no analysis to see whether this is statistically significant. Consequently the percentage differences may be perceived to demonstrate a difference, but without further statistical analysis it is not clear if this difference is likely to have occurred by chance or whether it is a statistically significant difference. Further compounding this are the small numbers achieved by some studies meaning that the use of quantitative methods is likely to be inappropriate for testing statistical significance. Again using the study by Al-Jader et al. (1990) as an example, only 29 sets of parents were recruited. In the questionnaire by Dankert-Roelse and colleagues only 38 individuals completed the

questionnaire (Dankert-Roelse et al., 1990). Other studies such as Dillard et al. (2007) and Hildes et al. (1993) had samples of 40 or fewer.

In those studies where additional statistical analyses have taken place there are further issues in terms of their substantive usefulness. In the study by Dillard et al. (2007) their conclusions are somewhat diluted by the revelation that the observed increase in parental knowledge cannot be explicitly said to be due to the counselling intervention being assessed. In the study by Holtzman et al. (1983), which found several variables to be statistically significant in the explanation of parental knowledge scores, only a minimal amount of the variance was accounted for; in some instances leaving up to 80% of the variation unexplained. This points to a lack of prior research in terms of determining the appropriate variables that are implicated in parental knowledge about newborn screening.

It is this lack of prior research that makes much of the quantitative research inappropriate. This is symptomatic of a wider issue; that of a lack of studies addressing 'why?' questions. The exceptions here are the studies of Parsons and colleagues (Bradley et al., 1993; Parsons et al., 2005, 2006), which have sought to identify factors affecting parental decisions whether or not to accept an optional screen for DMD.

In conclusion, despite the published findings on knowledge, coverage of screening results and parental attitudes to newborn screening there is a paucity of research regarding parental decision-making, the information that parents use in making decisions, and the factors that affect their decisions. Furthermore, none of the studies relate to current UK practice and how parents experience this, perhaps not surprising given the delays between research and publication and the rapid rate at which clinical practice is altered. Nearly all of the research within the UK context has been conducted prior to the current guidelines. As such, and again this ties in

with the opaqueness regarding details of information provision within published studies on knowledge, it is difficult to gauge whether the existing literature would reflect the current status within the UK regarding knowledge, understanding and decision-making of parents. There is a need to conduct research within the current UK context, and to make this context explicit, so that one can assess the findings relative to the processes and mechanisms that are in place. Only then will it be possible to begin to identify, or at least suggest with empirical support, causal elements or mechanisms that are implicated in regarding parental decision-making and consent to newborn screening.

Chapter 3

Devising the structural model: Methods

The first stage in devising a model of parental decision-making is the determination of the appropriate variables to be included. Once a set of variables has been agreed upon one needs to generate a theoretical framework in which these variables interact. As evidenced from the literature review there is a paucity of data relating to either the variables to be included or the way in which these variables impact upon parental decisions. Consequently it was deemed necessary to undertake preliminary work to generate both the variables and the proposed model of parental decision-making that could then be assessed through statistical analysis.

3.1 Methodological overview

The study undertaken here follows an exploratory sequential mixed methods approach comprising of qualitative and quantitative phases (Creswell and Plano Clark, 2007). In doing so, I approach the study from a critical realist perspective, attempting to identify and explain causal mechanisms (Lawson, 1998; Sayer, 2000; Danermark et al., 2002). The use of both quantitative and qualitative data has been argued to be important in empirical research that seeks answers to causal relationships, such as decision-making for newborn screening, with Sayer arguing

that:

> "in any concrete study it is important not only to know what causal mechanisms are present and how they work but also to have a quantitative estimate of their number and their effects (if they are separable)" (Sayer, 1992, p181).

Recent years have seen an increase in mixed methods approaches (Creswell and Plano Clark, 2007; Creswell et al., 2004) or 'multi-method research' (Bryman, 2004). This development has been partly driven by increased funding for interdisciplinary research but also increasing support for methodological triangulation.

To the extent that the aim here is to identify causal mechanisms, it could perhaps seem that an appropriate orientation for a mixed methods design would be to begin with a quantitative phase. This would allow one to gather data regarding the commonality of a phenomenon, and then to use a qualitative stage to elucidate further and develop theories regarding the underlying mechanisms, this being what Creswell describes as an 'explanatory' mixed methods approach (Creswell and Plano Clark, 2007). To a certain extent this would be ideal; perhaps examining the details behind an observed pattern. As already stated, there is a paucity of research relating to parental experiences of decision-making for newborn bloodspot screening. The effect of attempting to model a causal relationship on limited data would lead to the problem of *specification error*, with the omission of theoretically key variables severely affecting any statistical inferences that could be drawn (Hair et al., 1998). Consequently, the exploratory sequential mixed methods approach was deemed the most appropriate as it not only provides the opportunity to develop causal explanations through the use of the qualitative phase, but also elucidates appropriate variables that will enable the development of statistical analyses regarding parental decision-making. This, of course, must be tempered by the practical restraints placed on empirical research and in particular the researcher in attempting to interpret the results. Thus, whilst one seeks

an exhaustive set of variables, one needs to remain conscious of the benefits of parsimony and concision, as well as the pragmatic limitations to attempting to identify causal relationships.

Consequently, it was decided that an initial phase of interviews would be conducted and that this would be followed by a quantitative stage comprising of a questionnaire that would be analysed using Structural Equation Modelling (SEM). In the remainder of this chapter I outline the approach taken to this first qualitative phase.

3.2 Why interviews?

Interviews are a preferred method when "the object of study is some form of social process or meaning or experience which needs to be understood and explained in a rounded way" (Mason, 1996, p96-97). This reasoning tends to be built around a constructivist way of thinking, in that our knowledge and experiences are built up through a series of social interactions that are not necessarily amenable to statistical analysis or other rigid methodologies (Arksey and Knight, 1999). Yet from a critical realist perspective it allows for the expression of motivations and for other influences to be derived so that they reveal or allow us to postulate causal mechanisms that can be further analysed through quantitative methods.

Semi-structured interviews were chosen for this study as they allow the respondents themselves to create their own definitions of their experiences rather than having definitions created and imposed by the researcher (Murphy et al., 1998). The flexibility over structured survey interviews is that they allow the researcher to follow up on topics discussed or introduced by the participant, as well as to probe responses for more clarity or detail (Bell, 2005). By allowing questions to be posed through different wording the interview can fit with the participants' understandings or beliefs. The individual interview is also preferable over alter-

native qualitative methods, such as focus groups, due to the ability to maintain confidentiality which is not possible in a group situation (Bowling, 2004).

It may be argued that what people say and do are different and that other ethnographic methods such as participant observation may be seen as more appropriate. Indeed a difficulty with interviewing rests on how one is to interpret the content of the interview. Some authors argue that the information garnered from an interviewee may be problematic, particularly within the context of recalling interactions with health services. As such, treating this data as a primary source can be misleading. This is not to say that individuals are consciously misleading the interviewer, rather the process of the interview itself may be seen as imposing, at least initially, a social structure which the interviewee may feel that they are required to conform to. As a consequence it may be more appropriate that the content derived from the interview "be treated as an account [...] rather than as literal descriptions of the respondent's reality." (Murphy et al., 1998, p121). With this in mind it was felt that the crux of the study was not the actual interaction between the professional and the parent but rather how this was experienced. This experiential nature of parental decision-making could not be accessed through observation and consequently interviews were chosen as the methods to be implemented.

3.2.1 Single vs. joint interviewing

Parents were interviewed individually. This was done for pragmatic and theoretical reasons. Pragmatically it may be more difficult to arrange to interview couples; theoretically, interviewing separately may allow the submissive partner more freedom to express their own individual views. Equally, a joint interview may stir up any antagonism that exists between the couple regarding the topic which would not be conducive to a good interview. By conducting one-to-one interviews with both partners, differences in knowledge or experience which would have remained hidden in a joint interview, may be revealed. Indeed, in the context

of this research, it was anticipated that differing accounts may be indicative of the process of taking consent. Prior research has found that questions of 'family' have traditionally been directed towards women (Valentine, 1999).

3.3 Sampling

3.3.1 Sample size

Qualitative interviewing necessarily requires small samples due to the complex nature of the data generated and the costs incurred in terms of collection and analysis of the data (Mason, 1996; Bowling, 2004). A total of twenty interviewees were sought. Appropriate sample sizes for in-depth qualitative interviews have been suggested as being as small as eight up to a suggested practical maximum of twenty for a single handed researcher (McCracken, 1988; Mason, 1996). The chosen sample size is also consistent with previously published research and recommendations (Senior et al., 1999; Bowling, 2004; Bryman, 2004).

3.3.2 Populations

Parents were purposively sampled in order to gauge a range of opinions and were selected from differing geographic areas on the basis of data available from the Audit Commission[1] to reflect differing socioeconomic status. Table 3.1 shows a selection of profile data from the areas where parents were recruited.

3.3.3 Sampling procedure

The study used a retrospective sample. Whilst the use of a prospective sample might be seen to provide a population who have more immediate experiences of the newborn screening process there is, in these circumstances, a greater possibil-

[1]Data taken from the audit commission website: http://www.areaprofiles.audit-commission.gov.uk. Accessed 9 October 2009.

Data profile item	Sefton	Macclesfield	Wirral	Liverpool
Rank of the average Indices of Multiple Deprivation Super Output Area (Scale 1 (most deprived) to 354 (least deprived))(2004)	78	276	48	1
Annual average House Price: Overall (2005)	£156855	£238122	£140153	£115244
% of children that live in families that are income deprived	23.7%	9.9%	29.9%	44.9%

Table 3.1: *Selected data profile items comparing recruitment areas*

ity that this could introduce a 'Hawthorne effect'. As Jones notes, this has been defined in a number of ways but "the central idea is that behavior during the course of an experiment can be altered by a subject's awareness of participating in the experiment." (Jones, 1992, p451). If a prospective study were conducted to examine parent's experiences of consent for newborn screening one may induce a change in professional practice when taking consent if the professionals are aware of a research project. A prospective sample would also have been more difficult to appropriate, taking longer to achieve. A retrospective sample was also the only way to identify accurately the small number of parents who had refused screening.

The use of a retrospective sample does, however, introduce the potential of a recall bias. This was minimised by having a relatively short time frame as part of the inclusion criteria. However, a time frame that was too short would be problematic on a practical level given the very small number of decliners and may preclude the opportunity to recruit those who have declined newborn bloodspot screening. Therefore a balance was sought between a time frame that was short enough to minimise recall bias and one long enough to identify at least some parents who declined screening.

The initial research design was to sample parents using a purposive, or what has also been termed a theoretical, sample, with sampling continuing until theoretical saturation had been achieved. Purposive sampling is a deliberate non-random method which specifically selects individuals on the basis of them holding a particular characteristic (Bowling, 2004). Purposive sampling is beneficial as one can target specific individuals with certain characteristics. The theoretical argument here was that parents who refuse consent to newborn bloodspot screening may have different experiences to those who accept screening. Random sampling and convenience sampling were discounted as they could have missed the small number of parents who had declined screening. This sampling process may also be considered a process of maximum variation sampling due to the deliberate seeking of parents who decline screening (Miles and Huberman, 1994). This strategy for sampling throws into light unexpected outcomes which may in turn identify underlying mechanisms or structures that are not apparent in other circumstances (Danermark et al., 2002).

3.3.4 Identification and recruitment

Identification and recruitment of parents was led by Dr Kevin Southern, Consultant in Paediatric Respiratory Medicine at Royal Liverpool Children's Hospital. Parents were selected on the basis of their child's sample being analysed by the screening laboratory and having been born within the twelve months prior to the start of the study in December 2008. This timeframe was selected in order to address the sampling issues highlighted in Chapter 2 but allowed the identification of a reasonable number of parents who had declined screening. With a small time frame between the screening and interview it was hoped that not only would recall bias be limited but parents would also be drawing on experiences relating to current practice that would, at least theoretically according to professional guidance, be standardised.

Parents were excluded if their child was severely ill, had subsequently died or if their parents were unable to converse freely in English. At Royal Liverpool Children's Hospital, Dr Southern and Elaine Hanmer, Laboratory Information Systems Manager, extracted the names of potential participants. Due to the low number of parents who decline newborn bloodspot screening and, in line with the purposive sampling approach, all babies whose parents had declined screening were selected for approach.

For those infants within the Liverpool area, the family health visitor was contacted by Dr Southern to identify any potential problem in approaching the family. In Macclesfield this contact was made by Liz Eccleston, Clinical Governance Lead Midwife at Macclesfield District General Hospital.

Despite best efforts, recruitment proved problematic using the screening laboratory list alone. Ultimately only two parents identified from the newborn screening laboratory list agreed to be interviewed, therefore additional recruitment methods were undertaken. To this end Sure Start schemes were contacted regarding the possibility of recruiting parents from play schemes and other organised events. It was hypothesised that the parents attending Sure Start schemes, in line with their aims and objectives, would be parents from lower socioeconomic groups, irrespective of the geographic region. In light of this supposition, regional branches of the National Childbirth Trust (NCT) were also approached. The NCT is a charity which aims to provide information and support to parents throughout pregnancy and early parenthood and also has a mandate to campaign on maternity and child health issues. In contrast to the Sure Start schemes, it was hypothesised that the composition of these groups would be largely middle class, white mothers.

Whilst initial interviewees were identified through these contacts, additional par-

ents were identified and recruited through a process of snowballing. This process uses respondents to identify further potential interviewees who are then contacted to see if they are willing to take part (Bowling, 2004). Hence, the final sample is more accurately described as a combination of theoretical sampling followed by snowballing which identified respondents in a situation where existing networks or lists did not exist.

In all cases, parents who met the inclusion criteria and who failed to meet the exclusion criteria were approached by letter in the first instance. This invitation letter (Appendix A) introduced parents to the study, as well as providing contact details. Included with the invite letter was a patient information sheet (Appendix B) which provided more details regarding the project. If parents wished to take part they indicated this on a reply slip which was returned in an included envelope. This reply slip asked for contact details so that an interview could be arranged. If no response was received within two weeks of sending the letter then a reminder letter was sent. If no response was received following this letter then no further action was taken.

3.4 Data collection

All interviews bar one were audio-recorded using a digital recorder. Audio recording is preferable to the taking of field notes during the interview as this can interfere with the process of interviewing (Britten, 1995).

All audio-recordings were transcribed verbatim. It is beyond the scope of this chapter to assess the advantages and disadvantages of the transcription of audio recordings; instead it is noted that this process is in itself selective and represents a translation or interpretation from what was said to what is to be written (Bourdieu, 1999; Miles and Huberman, 1994; Mishler, 2003). Transcripts were made available to interviewees for comment and were sent to those who had indicated that they

wished to receive them. Such feedback from participants has been argued for with regard to confirming the validity of the data (Miles and Huberman, 1994). In this study, however, no further comments were received.

3.5 Data analysis

3.5.1 Overview

Following the transcription of the audio data, the transcripts were loaded into Atlas.ti v5.2 (Scientific Sofware Development, 2007) which is a workbench for the qualitative analysis of large bodies of textual, graphical, audio, and video data. The major benefit of using qualitative data analysis (QDA) software such as Atlas.ti comes with the ability to store large amounts of data that can be retrieved in a short space of time. This does not reduce or eradicate the human element of coding and does not offer a simplification. Analysis still requires formal interpretation and analysis with the software assisting this by allowing the user to apply multiple codes quickly, negating the need for extensive paper copies of transcripts (Richards and Richards, 1998). This systematic retrieval of codes or quotations can allow for complex searches to be done with ease and one may easily look for co-occurrences of codings that may be nested, overlapping or which may occur with a specific proximity (Arksey and Knight, 1999).

The examination of the transcripts followed a thematic analysis approach (Boyatzis, 1998). Thematic analysis shares many features with other methods of qualitative analysis such as Grounded Theory in so far as textual data is coded and labelled and, in an inductive approach, are grounded in the data. The paucity of prior research on newborn screening precluded the use of any pre-defined coding scheme. The focus of the analysis was not only on the development of themes that would be taken into the second stage of the study but also the development of causal relationships between these. As such it diverges from traditional Grounded

Theory approaches where the development of a general or substantive theory is a key goal. The thematic analysis conducted here may differ from some qualitative research as it was conducted both within and across cases (Braun and Clarke, 2006).

3.5.2 Coding

Analysis began with a process of 'open coding' (Strauss, 1996). Open coding is the process by which codes are developed and attached to segments of the transcript. These codes are:

> "...tags or labels for assigning units of meaning to the descriptive or inferential information compiled during a study. Codes usually are attached to 'chunks' of varying size – words, phrases, sentences or whole paragraphs, connected or unconnected to a specific setting. They can take the form of a straightforward category label or a more complex one (e.g. a metaphor) [...] so the researcher can quickly find, pull out, and cluster the segments relating to a particular research question, hypothesis, construct or theme." (Miles and Huberman, 1994, p56-57).

This process of coding was iterative with the codes being developed using the constant comparison method allowing for the constant revision, combination or separation of codes in light of new data (Strauss, 1996). Each newly coded incident was compared both within and across parents to previous incidents in order to refine or revise the code (Fielding and Lee, 1998). Codes were either 'in vivo'; coded using a phrase used by the data source, or generated by the coder; what have been described as constructed codes (Patton, 1980; Strauss, 1996).

Codes were developed using a five step process:

1. A label (i.e., a name)

2. A definition of what the theme concerns (i.e., the characteristic or issue constituting the theme)

3. A description of how to know when the theme occurs (i.e., indicators on how to 'flag' the theme)

4. A description of any qualifications or exclusions to the identification of the theme

5. Examples, both positive and negative, to eliminate possible confusion when looking for a theme (Boyatzis, 1998, p31).

In addition to coding, a process of memoing was also introduced in order to maintain ideas and conceptual frameworks that became apparent during the process of coding. As such, the memo acts as a device to bridge the transition between the empirical data and the emerging conceptual structures (Miles and Huberman, 1994).

3.5.3 Data reduction

Following the initial open coding, themes were identified which subsumed multiple codes. In this respect it is similar to what grounded theorists describe as selective coding and involves the incorporation of the conceptual and theoretical developments captured in the memos. As such, the theme operates at a higher level than the immediate codes although it may still operate at the manifest or latent levels (Boyatzis, 1998).

This data reduction began through a process of pattern coding, that is "grouping those summaries into a smaller number of sets, themes or constructs." (Miles and Huberman, 1994, p69). As with the initial coding, themes were reviewed and revised in an iterative process. Cross case analysis was used to develop the notions of understanding and explanation highlighting differences in decision-making.

3.5.4 Reliability and validity

Reliability may be considered as "the consistency of a measure of a concept." (Bryman, 2004, p71) whilst validity "refers to the issue of whether an indicator (or set of indicators) that is devised to gauge a concept really measures that concept."(Bryman, 2004, p72). Reliability may be difficult to achieve in qualitative research; the same interview may yield different results due to different experiences. The process of the interview may encourage an individual to reflect upon their own experiences which in turn may modify their views. This poses the problem of how one can apply notions of reliability to qualitative research. In order to overcome this I follow the work of Mays and Pope (2000) in accepting a notion of reliability which requires modification for qualitative research. I accept a criterion of internal reliability whereby there is a consistency within the study. In this case it is the consistency of the application of codes, avoiding a definitional slide. This will be accompanied through procedural elements such as the constant-comparison technique and a reflexive approach to the research.

As previously mentioned, the notion of validity relates to the accuracy of the research results. To this extent the feedback of participants as a process of respondent validation is often advocated. It is argued that this not only provides a level of corroboration should the respondent agree with the analysis but also that:

> "... they should be able to pick up any nonsensical interpretations - the kinds of things they could not have possibly said. However, sometimes respondents will disagree with the transcript, even though it is clear from the recording what they really said." (Gibbs, 2007, p95).

In this study copies of transcripts were sent to those who indicated a wish to receive them. Feedback from participants has been argued for with regard to confirming the validity of the data (Miles and Huberman, 1994). Such post-interview interaction may also serve as part of the debriefing for both the researcher and

interviewee and as a way to obtain feedback about the research in general (Sieber, 1992). The finding that no parents provided additional comments or feedback on transcripts may indicate that they were satisfied that the transcripts represented their perspectives, although this assumption was not verified.

Chapter 4

Devising the structural model: Results

4.1 Parent participation

A total of 18 interviews were conducted. Two were with fathers, twelve with primaparous and six with multiparous parents. Unfortunately none of the parents who had been identified as refusing the heel prick took part in this study; three parents who declined screening actively declined to be interviewed with the remainder not responding. Consequently the data relates only to the experiences of parents who had accepted the heel prick. Only one parent had a child affected by one of the screened for conditions, although this was not their most recent child. Another mother had a child affected by an inherited metabolic condition with similar implications to Phenylketonuria. The interviews ranged in length between twenty and forty five minutes and all interviews except for one were audio-recorded and transcribed.

As hypothesised by the purposive sampling frame, participants varied in terms of their personal circumstances and experiences as well as age and number of children. However, specific data was not collected on these demographic charac-

teristics and only spontaneously offered data is available. As the data comes from a small sample it would be inappropriate to apply statistical notation, such as percentages, when discussing the thematic analysis and the number of respondents who discussed particular themes. Whilst the intention was to interview parents individually, on occasion partners were present during part of the interview. In all instances additional consents were taken to include their input.

Seven key themes emerged from the interviews as being either explicitly or implicitly causally related to parental experiences regarding consent for newborn bloodspot screening. These themes are classified as *Experience*, *Attitudes to medicine*, *Information-seeking behaviour*, *Perceived knowledge*, *Attitudes to screening*, and *Perceived choice*, all of which ultimately impact on *Perceived decisional quality*.

In the remainder of this chapter I discuss the themes that emerged from the interviews and how these relate to parental attitudes towards their decision-making. These are then drawn together in the proposed models that are to be taken forward into the statistical analyses.

4.2 Information-seeking behaviour

Whilst the focus of this study is on parental decision-making to accept newborn bloodspot screening and the factors that affect this decision-making process, an important prerequisite is the information on which the decisions are based. This information is important both in the theory and definitions of informed choice, but was also talked about at length by parents when they discussed their experiences of the heel prick.

Parents were found to gather or receive information at various times during the pre-natal and post-natal period through a range of sources including friends, family, books and the internet, each of which supplemented the information provided

by the midwife. Those parents who made reference to hearing about, or discussing, the heel prick prior to birth were in the minority.

The interviews identified differing information-seeking practices which were broadly classed as *active seeking* or *passive receiving*. Active seekers were those parents who sought out information, often from sources other than the National Health Service (NHS), and engaged with differing perspectives. Conversely, passive receivers engaged in information that was presented to them, primarily by the midwife, but did not necessarily forge their own avenues of enquiry. An active seeking behaviour was clearly shown when one mother detailed her experiences:

> "I: Did she give you anything to read, or was it just verbal?
>
> JG: Verbal
>
> I: And were you happy when she, to go with that?
>
> JG: Well, yeah, 'cos I looked it up anyway.
>
> I: So you looked it up any way.
>
> JG: After, afterwards - so erm, my friend's a midwife as well
>
> I: Ah right, OK I see
>
> JG: So I asked her about it as well.
>
> I: So where did you go and look it up?
>
> JG: Internet" *JG*

Here the briefing by the midwife generated a number of issues on which the mother then gathered information. The internet proved to be a starting point, but this was then corroborated and supplemented with information from a friend who was also a healthcare professional in the field. Whilst not taken as generalisable data, those parents who had an active information seeking personality tended to be those who were recruited on the hypothesis that they would be of a higher socioeconomic status. However, this may not be causal as these parents were also pro-active members of groups, such as the NCT and so it may be that membership is indicative of having an active information seeking personality. Furthermore the

distinction was not clear between the hypothesised social groups and consequently no definite conclusions can be drawn.

4.2.1 From whom and where do parents find out about the heel prick?

As reported in previous studies, a range of information sources were used by parents including official NHS leaflets, books, the internet, friends and family. The most consistent reference was made to the midwife who played a central role in information provision for parents. Parents cited difficulty in finding the time to read written information during the post-natal period and the advantage of the midwife, particularly for passive receivers, was that the visits provided an opportunity to gather information quickly during time that was already set aside. For others a benefit was the ability to clarify points through discussion or gather additional information that could serve as either an end in itself or a starting point for further investigation:

> "She was *a* source of information, but erm, certainly an important source of information, erm, but I'm not sure I'd say she was a primary one or not. Most of our decisions was informed by what [wife] had already, already read, and then by discussions with [name] the midwife. Erm, yeah I think there was a good level of discussion about it." *SPW*

Despite the time constraints, for some parents written information, in the form of books and magazines, was useful. Whilst the majority of parents recalled being given an official NHS leaflet it was used by few parents to inform their decisions. This lack of use was sometimes due to the way it was provided with other commercial literature or left for parents to find. Thus, for some, written information was provided not only at a time that made it difficult to use but also in a way that failed to draw attention to it. For those parents who read the content proved useful and in one case was their primary source of information.

The internet was not a great primary source of information for parents researching the heel prick. The decision to accept the heel prick was straightforward and the internet served a need only when researching a contentious issue, such as vaccinations or pre-natal testing. In the context of the heel prick, the internet was largely used as a supplementary tool for gathering information. One acknowledged limitation to the use of the internet was a lack of quality control. One mother, who sought information after not receiving any from the health service, recalls:

> "I did look on the internet [...] but you don't know, especially with the internet, you don't know what you should [...] take and not take, because it could be very scary if you looked at that, you know what things, the statistics and everything. You could get quite scared if you looked on like one internet site, you know if you googled it and put it in." *NW*

Consequently there is a huge volume of information that could be gathered without any form of guidance as to the relevance of that information to their own situation. For others as with the written information, time, or lack of, is a factor that precludes its use.

4.3 Experience

Personal experience and the experiences of others were implicated in various aspects of the decision-making process, ranging from the collating of information all the way through to the actual decision itself.

4.3.1 Experience and information-seeking

A third of the parents interviewed were multiparous. For these parents personal experience played a significant role in their information-seeking behaviour. For some this effect was to reduce the amount of information they sought, instead

relying on their own experiences. Yet for others prior experience of 'the system' and processes of childbirth meant that they were able to overcome their previous naivety and focus on the specifics of the information:

> "I: So what's changed this time?
>
> LW: But this time, I think you change as a parent anyway and you gain in confidence, so.
>
> I: Why do you think that is, why do you think you've sort of, where do you think you've got your confidence from?
>
> LW: [laughs] Well it's your child and you want to protect them at the end of the day and make sure that everything's right for them, you know, and it's learning that your views do count and learning that you are the expert on your child, because at first I thought 'oh my God I don't know what I'm doing', you know, it was like shock horror and then its, it's learning." *LW*

This experience meant that parents were able to not only question medical authority, but also to have pre-emptively thought about situations that they would face and prepare for these. Experience, however, was not limited to the process of the heel prick. Other experiences of illness or the healthcare system were drawn on by parents. One mother's own health issues motivated her to try and find out more information about congenital hypothyroidism:

> "I was quite happy for them to do it, one of them, cos one of them, one of them's a thyroid wasn't isn't it? [...] And I did ask cos I've got thyroid problems, and I did ask them about that one and they didn't give me any sort of [...] information on it. If it's hereditary, or anything, if she's high risk because I've got an over active thyroid or anything." *NW*

Although this mother did not have a technical understanding of congenital hypothyrodism, her own experiences of her thyroid problems led her to acknowledge

that this was something she wanted information about. She then sought technical information from the midwife, although it appears that this desire for information was not fulfilled satisfactorily.

Experience as a source of information

An important source of information for some parents is *experiential knowledge*. This encompasses both *embodied knowledge*, that is subjective knowledge that comes from one's own personal experience, but also *empathic knowledge* which is subjective knowledge gained through the interaction and association with others, such as friends or family (d'Agincourt Canning, 2005; Etchegary et al., 2008). In contrast to the midwife and the written resources, friends and family were rarely used to gather technical information. When parents did recall discussing the heel prick this had generally been limited to recollections of the process; how the baby cried when blood was drawn and the emotions that they felt, rather than any detail on the tests. As one mother recalled:

> "Well, erm, I kind of knew about it through friends and my sister in law, whose got two boys erm, five and two, so the littlest one was only, [boy] was only sort of less than a year old when we found out that [daughter] was on her way erm, and I remember there just being this sort of dreaded day three, midwife comes to the house and, and you just want to punch them. Was the general feeling I got from these other mums because they're making your baby cry kind of thing." *JM*

This contextual information appears to serve as a process of preparation, allowing parents to ready themselves for unfamiliar procedures.

4.3.2 Experience as a method of validation

Whilst experiential knowledge was sought, and as such one could argue that it was their experience that made the information valid, the use of experience as

a way to validate information was not restricted to friends or family. In some instances parents appeared to be using experience, assessed through a proxy of age or number of years in a particular field, as a way of validating the information that they had received and so determine its legitimacy.

> "I think they're probably as important, because I'm quite close to my parents and, and obviously being that close to somebody who has had children, mum was, mum was very useful, and also my husband's mum er was very useful as well, cos she's a nursery nurse and has loads of experience so yeah." *SMB*

So, for this parent, the experience of her mother-in-law within her role as a nursery nurse is a method of validation for the information she provides. Trust was placed in the individual, and this individual trust is generated not only by regulatory identifiers, such as qualification or association to an institution that implies both competence and worthiness, but also through experience and interaction. This was explicitly stated by one mother who had cited the midwife as an information source:

> "I: Erm, is there anything about the fact that it was sort of the midwife that's good, other than the fact that she works at the hospital, is there any other benefits that you can think of?
> NW: Well I think because she's know, with a baby, she's more, she's a bit more experienced isn't she." *NW*

Thus the experience of the midwife was linked to competency and trust and as such an additional tool of validation alongside her qualifications.

4.3.3 Assumed knowledge with experience

Whilst parents saw positive aspects to their experience in assisting them to make their decisions, it was also indicated that when experience, and particularly knowledge gathered through experience, was assumed, this could lead to them being less

informed. In part this was due to professionals assuming that the parent would know about the heel prick:

> "[...] I actually don't remember being told the details as much that time [with 2nd child], now whether that's because they said do you know what the heel prick test is, to which I've gone yes, because obviously they've done it before so they've just thought OK, take the blood and send it off. That's possible, that once you've had one child a lot of details are then skimmed over, that they just assume. I, and I probably assume, 'oh yes I know' when actually, really, if you'd have dug a bit deeper I actually didn't really know what it really was." *LM*

This is reflected in the way that parents often had greater recollections of their first child having their heel prick than their second. The assumption within recruitment was that the heel prick would be fresh and clear in their mind. Here the suggestion may be that even when parents have a second or third child, it is the experiences of their first that may be more lucid.

More concerning though is the way that this parent (LM) reported the assumption of knowledge by professionals. Consent was experienced as less thorough for their second child where it was felt that the professional assumed that they knew about the screening, resulting in potentially less thorough information provision. If this is the case more widely then it raises serious questions regarding the informed nature of any consent provided by multiparous parents.

4.3.4 Experience and decision-making

For some parents the decision to accept screening was largely based on their previous experiences with the heel prick. For those multiparous parents (N=6) the decision appeared straightforward:

> "She went through it very briefly with me and explained about the

> different reasons for doing the test and I'd had the test done on [son] and we'd already made that decision with him so to make the same decision with her was much quicker and easier like yeah, yeah, whatever come on, come round and do it [...]" *AG*

And later:

> "But it was much easier for me to decide for her to have it done than with [son] and we went through quite a lot of erm, [...] grief and research and soul searching with [son] the first time about immunisations and testing and amnio, all those sorts of things but when it's second time round you've already been through that decision making process, so it's like are we gonna do what we did the first time or have we changed our mind here so in a lot of ways it was an easier." *AG*

This perspective, of reviewing the experiences of their first child and basing their decision on that experience, was replicated in other parents. Thus whilst experiential knowledge from friends, family or professionals was an important information source, when this experience was personal there appeared to be far less deliberation or conflict. As suggested, this may be reflected in the ability to recall more clearly the heel prick from their first child.

The caveat here is that these parents had not previously had an adverse outcome to their decision, or a decision which they had regretted. Had this been the case it may have been that personal experience would have had a different effect on decision-making.

4.4 Attitudes towards medicine

For some mothers the decision to accept screening was informed through more general attitudes towards medicine. These attitudes were often affected by their own

experiences of the healthcare system and their perceptions of how the healthcare system in the UK works. In particular this was linked to a perceived knowledge of the processes through which testing becomes available:

> "[...] they don't do these tests just for the sake of it. You know, and the ethics, and all of the things they have to go through to be able to do these things they, you know, it's obviously enough historical evidence to prove that by doing this, a bit like cervical screening and breast screening, you know they've obviously got evidence to prove that 'if we do this at this point in their lives, we've got a better chance of finding it, diagnosing it, and treating it without it becoming too much of an issue further down the line' " *JM*

Consequently, a positive attitude towards medicine and testing was reflected in positive attitudes towards screening. This was occasionally framed by the financial constraints within which the NHS operates:

> "SB: Yeah, yeah. They obviously know what they're doing, they're trained professionals and they wouldn't just rou... you know cost wise and things like that, do these things if it wasn't necessary. You know, knowing the way the NHS is and things like that, I couldn't see them doing it just for something to do.
>
> I: Yeah. Sorry, what do you mean, you said knowing the way the NHS is?
>
> SB: Doesn't, with funds [...] You know, they all seem to be short of money, you know when they, you know, you want these cancer drugs and that but they say they can't afford it and things like that. So I shouldn't imagine something like that would be carried out unless it was absolutely necessary." *SB*

Without explicitly mentioning Herceptin or similar medicines, this parent has neatly summed up the perception of the approval process. This perhaps clarifies

the behaviour of parents who accepted screening without seeking much information. By virtue of screening being offered by the NHS, it is perceived to have been reviewed and assessed to a level whereby the NHS is happy to provide it, which indicates to parents that this is a good thing to have.

Decisions were not solely made on perceived knowledge of the wider processes of the NHS, but also more intimate aspects of personal contact. Whilst the midwife was an important source of information she also played a significant role in parents accepting newborn bloodspot screening. For some parents, as was the case with information provision, the offer by the midwife suggests a level of quality and it was trust in the midwife who was offering the test that led them to accept. As one mother explains:

> "I: And did you think about it between them forewarning you and actually doing it?
> SP: No
> I: No?
> SP: I think that because of who it is telling you, you just assume it's OK.
> I: Yeah
> SP: And that it has to be done and that, you know, it's just a routine thing and there's no issue with it.
> I: So because it's the midwife you kind of just...
> SP: Trust, yeah." *SP*

Thus, the offer by the midwife suggests a level of quality that in turn provides the parent with a trusting relationship. Trust in the midwife was built through their presumed qualifications – a presumption based on their affiliation to hospitals and the NHS more generally – together with parents' expectations of training. Ability was demonstrated through knowledge, itself assessed by answering questions or providing information. This was combined with personal qualities to generate

trust. These personal qualities were demonstrated through the interaction with parents:

> "I: And do you think, maybe, if she couldn't have answered your questions there and then it would have been a bit...
> JS: Yeah, yeah, well, it depends. I mean obviously if I would have asked her a question that's not, is something that she'd need to research, if she would have said I need to research that, then that would have been fine." *JS*

So a level of personal reflexivity and honesty was deemed to be important, even if that was to the detriment of the immediate level of information she could provide.

Whilst trust in the midwife and the NHS was relatively high, parents were less trusting of administrative aspects of hospitals. In particular, concerns were raised over the provision of results. Parents recalled being told that 'no news is good news' and that they would only receive results if there was anything of concern. This raised concerns in parents who would have liked to have received confirmation either way:

> "Because I don't feel you can trust, erm, I always think, what if, something goes missing, or doesn't get done, then you wouldn't know. Because when, when I was waiting to find out, I had the down syndrome blood test, the triple test, and erm, obviously they said 'if you, if you haven't heard within two weeks or so then, then consider it to be fine'. So obviously nothing came, nothing came, nothing came, and then when I went back for the next antenatal visit I said err, can I just check on my down syndrome results, they couldn't find them. They weren't in my file, so they had to ring the lab, and then they had to write them down on a piece of paper, and I really wasn't happy about that. I just thought, well what if it happ...I mean, what if it had been a problem?" *HP*

The concerns were not unwarranted and again drew on individual experiences in developing their attitudes. The examples given also show how the attitudes towards midwives, and the NHS more generally, can interact with specific attitudes towards screening and information-seeking behaviour; both of which are important in parental decision-making.

4.5 Perceived knowledge and understanding

Parental knowledge was not assessed through direct questioning. Given the shortcomings of knowledge assessed by recall, parents were asked to relate their own perceptions of their knowledge. When talking about their experiences of the heel prick, parents commented on their own perceived knowledge. Most parents were self-deprecating, with a number openly stating that they could not remember anything:

> "I have got no idea, er, and I didn't even remember it erm, until I spoke to my partner and said erm, I'm going to do this interview and what it's for and he said oh is that the one that was done in the house, so I think he remembers slightly better than me, and that's obviously because you haven't given birth to anyone." *SP*

In particular parents sometimes felt they had a poor technical knowledge, unable to recall details such as the exact names of the conditions, the specific prevalence or similar. To this end several mothers suggested that severe tiredness was detrimental to their ability to remember details. As one mother explained:

> "But when you've just got a new baby you, you're knackered aren't ya, and you're looking after the baby and just cos I'm, I couldn't, I couldn't have told you the next day and, you just forget." *LT*

Others talked about how physiological or psychological changes, such as hormones and emotions, affected their memory:

> "Because when you have a baby your hormones are completely gone for the first two weeks aren't they, so, which is probably why people can't remember what the heel prick test was actually for." *LW*

It should be remembered, however, that only one parent had a child with a screened for condition. It may well be that parents of unaffected children have a decreased knowledge, possibly due a perceived lack of need to remember details. Parents of children who are affected by the screened for conditions may have a greater knowledge, particularly for the condition which affects their child.

Despite this self-professed lack of technical knowledge, when parents began to talk about the heel prick, many exhibited some knowledge about the screening. This was often knowledge about the implications of the screened for conditions. One mother, who proclaimed to have little knowledge, revealed a clear understanding of the effects which she had considered in her own context:

> "Yeah, I just recognised it when erm, and I think also PKU, I'm sure I'd seen something on television about it, I can't remember what, what it was, but I do remember somebody on TV talking about the diet their child had to have with these special protein supplements and things so, that just stuck in my head as a condition that's qui.., could be quite difficult to manage in day to day life." *HP*

This knowledge was sometimes presented in their understanding of why the heel prick was taken. This occasionally included what may be viewed as technical information:

> "Yeah, because I think knowing erm, the conditions that were being tested for and knowing that they were erm, metabolic disorders, and knowing that they could easily, well I'm saying easily, but knowing that they erm, could be treated with either diet or medication, then it

> would be much, much more beneficial to have the test than to decline the test." *LH*

> "I can't remember if it's this one, is it to do with finding out certain, if, if ya child can only have certain food, foods and stuff like that, yeah. That's the only thing I can really remember really. And obviously if they, if they find out at this early stage then it can, then it can erm be very beneficial and it can basically, you know, eradicate any sort of future problems, can't it, and things like that." *KS*

The indication here is that whilst memory may be compromised, the parents understanding of implications is not. A corollary of this is that traditional methods of memory recall would have failed to capture this level of understanding. So whilst it may be argued that parents have a poor knowledge due to their inability to recall technical information such as risk figures, this obfuscates a more complex process of information integration and understanding.

4.6 Attitudes towards screening

As already detailed, general attitudes towards the NHS and the midwife appeared to dominate attitudes towards newborn bloodspot screening. This, however, was not unilateral and for many parents, particularly those *active seekers*, a more considered approach to risks and benefits was taken in developing their attitudes towards screening.

4.6.1 Risks

Whilst potential concerns were raised, these were all perceived to be low risk. The main concern voiced by parents was the potential pain that would be inflicted upon their child. This was often revealed when parents were asked if there would be anything that would stop them having their child screened:

> "Erm, maybe if there was quite bad side effects or something for the child or it could, I don't know if, if it was erm, probably just that really, or it, you know, cause harm in any way and stuff, which, I know it's not nice for them to suddenly get their foot pricked with a, when you've only been out of the erm your belly for two minutes and they're like getting poked and prodded aren't they and things like that but yeah, I suppose if it caused any damage really to the child, yeah." *KS*

The focus on the potential for distress and the effect of the disease may reveal why parental knowledge of this and procedural aspects were greater than other, technical, aspects. Despite these concerns, the actual risk of distress was seen to be minimal:

> "Erm, yeah, but it's something that you know's not gonna...and it's short term it's, you know, and they're not going to be psychologically marred by it for the rest of their lives, erm, and you can comfort them." *LH*

This perception was strengthened when they were provided with information as to how they could take steps to minimise the distress to their child:

> "It was things like this sort of what's, I was reluctant to, to, stab a newborn baby and cause it distress for a relatively small benefit, but she was able to reassure me, I had heard of the erm tenderfoot and she said that's what she used and that a lot of babies didn't feel it, especially if they're feeding at the time she did it." *PW*

One particular factor that affected the perceived level of distress was parental opinions on the cognitive abilities of the child. The benefit was that at such a young age the child would not remember it and so not be adversely affected.

Perceived risk: better out than in

Some parents spontaneously compared the heel prick with the measles, mumps and rubella (MMR) vaccinations which are offered. This distinction was based on the experience of the child but also on issues of risk and safety:

> "[...] also I think because there's a chemical element to the MMR and all the vaccines you go for, whereas the heel prick's very straightforward, erm, you know, it's literally just taking the blood isn't it and testing that blood [...] externally, so, erm, it's not, you know, if you're not filling your child with something that you don't necessarily think that would, that they would have anyway, that's an unnatural element, then I think that's more questionable than, than just kind of like a small scratch [...]" *SMB*

This safety related to the spatial location of the testing, with the screening done outside of the body and so posing little or no risk to the child. MMR was seen as more risky by the parents who raised it, with pain caused by boosters and uncertainty about the possible side effects and adverse consequences.

Accordingly, parents were found to view the heel prick as a minimal risk intervention. In line with their own expressions of knowledge and understanding, the procedural aspects appear to weigh most on their minds in terms of any potential harm or benefit that may result. This was not only to be influenced by their information-seeking behaviour and the sources from which they received the information about the heel prick, but also their own ethical beliefs regarding minimisation of harm and maximisation of benefit.

4.6.2 Benefits

Whilst risk was weighed in terms of harm from the process, benefits were weighed on the basis of the diseases being treatable. This was conveyed when parents

were asked about the possibility of screening for non-treatable conditions and for which some parents saw little benefit. Whilst treatability has been reported as an important concept (Natowicz and Zuckerman, 2009), this was found to be a subcategory of what I shall call the *ability to act*; that is parents could do something with the results from the screening:

> "SPW: I would have said that was a low, a low priority if you can't really do anything." *SPW*

The important aspects were practical steps that could be taken.

> "[...] in a way you see with a result from a test like that you're knowing something that you're going to know anyway about baby eventually so you want to know earlier and it's better to know very early....the process [...] You would have found out anyway eventually erm [...]" *JG*

Testing earlier rather than later was supported in terms of knowledge being gathered earlier, allowing treatment to progress. This knowledge was also beneficial as it was felt to help parents cope. Knowledge of disease as a method of coping emotionally has been invoked by some commentators as a reason for expanding newborn screening (Bailey et al., 2006). In the current context it may also be seen in terms of adopting treatment strategies that would allow them and their child to deal with the disease.

4.7 Choice

When discussing the heel prick with parents it soon became apparent that the initial conceptualisation of parents conscientiously considering information, making a decision based on this information and then enacting this decision, was not only overly simplistic but in some cases erroneous.

4.7.1 Routinisation

For most parents the offer of the heel prick was experienced as routine. That is, parents experienced the offer not as a distinct event that existed as something that one could opt for but something that was done to everyone. As one parent stated:

> "They said they need to do some, something routine, I think I signed something, erm, and then, they said they needed to do a couple of routine tests..." *SP*

This was perpetuated by its inclusion with other post-natal checks. The fact that the other checks were readily accepted led some to the conclusion that the process was an automatic one. This assimilation of optional tests with more 'routine' ones:

> "may contribute to an interactional context in which screening itself is also perceived as routine, and where any decision to be made is taken less seriously as a result. (Pilnick, 2008, p523)

This normalisation can be seen in the way parents talk of the processional nature of screening:

> "It was just, as I said, it was just one of those things that was all part of that, all of this, this big machine that happens as soon as you, as soon as you have a baby. You know, things like the health, triggering all these visits from people...its just, not like a tread mill but you realise that you are part of this as I said, system." *HP*

In some instances this led to a view that the goal was that parents comply with policies and practice. The potential effect of routinisation is that consent to screening may not be informed. In particular this could be detrimental to those passive information seekers where the midwife may be the only source of information.

4.7.2 Dualistic representations: importance and insignificance

The way in which screening was presented or talked about was seen as important by parents. Parents talked about the heel prick being presented in a way that sought to maximise uptake and minimise concern. As already suggested, the presentation of screening as routine served to suggest it was insignificant. This was compounded by the way that midwives talked and were dismissive about susceptibility and likelihood:

> "Erm, [long pause] I think...the way it's offered can throw people a little bit. If it's offered in a way that they're almost expecting the results to be negative, you know that, in as much as, as if you, if ya, if you hear the, the, the ratio of it being picked up, the ratio of it being in the population as being very, very low, erm, so as a result there's a very low risk of your child having this disorder, then you're going to go along with it." *LH*

Couple this with the way that the information is often provided, amongst other leaflets or talked about amongst other tests, and the test appears insignificant. Yet this was counterbalanced with the importance placed on the heel prick by the midwife, with parents not only interpreting it as important, but also explicitly recalling being told this.

> "If anything because of the way she brought, you know, approached certain, the way she explained it, and what it was for and stuff, and it made ya think oh yeah, and obviously it must be important that it needs, you know having, needs to be done." *KS*

This perception was enforced by the fact that the midwife actively recommended the screening:

> "[...] So I do remember them saying erm, well I don't remember them to be honest even saying it was real, well they must have said it was voluntary although I think they do say that it is very, very recommended that you do have it, and I do; I think, although I could be totally wrong, that I have a vague idea of why they do it." *LM*

As a consequence one can see how the perceived routine nature of screening; that it is offered to everyone in a way that does not draw attention to it, together with a firm recommendation that one should have the test, contrives to suggest to parents that the test should be taken.

4.7.3 Perceived choice

Parents recalled being told that "we're going to come and do this..." and so, as reported in other studies such as those by Parsons et al. (2007), screening was seen as a *fait accompli*. Indeed several parents prefaced their interviews by saying that they hadn't considered the screening a choice. The implication of this is that rather than providing an informed consent parents may simply be providing an assent to screening.

4.7.4 Constrained choice, competence and coherence

For some, the timing of the heel prick meant that actually making a considered and informed choice was difficult, if not impossible. This difficulty was clearly articulated by one mother who had only received her information post-natally:

> [...] I don't think you're given any time cos your just told that they're going to do it and they need to do it, [...] They don't say we'll leave it with you to think about and read, the literally say, bllllr the test and right here's the needle and they're about to take the blood. So it's a very very quick process and you're not given any option to think about it." *LM*

For others the difficulty lay in other aspects of the time, with some questioning their ability to focus and make a decision:

> "After you've given birth it's such,...well for me it was quite traumatic really [...] I think just my emotions kicked in, hormones or something like that. So I was, I felt more of a sound mind when I was going to antenatal" *SB*

This has potentially significant ramifications for any purported consent that would be given. As one parent explained:

> "[...] I didn't question it. Probably when, when, my natural way would have been to question it and say why is he, why is it, what and get a bit more information about it, but you just go 'oh OK'. You're a bit bewildered aren't you when you've just had a baby. And your husband isn't normally there because they come in the day." *LT*

For this mother the decision to accept the screening, without questioning or looking into it, was out of character and one may question whether this was an informed consent. This issue was raised by one mother who questioned whether she would have been in the same frame of mind then as she is now, some time after the birth.

> "Mmm. No, OK. But if, if they were gonna do a heel prick test tomorrow [...] On [daughter] I think I would erm, yeah my er, my, my thoughts about it might be different I suppose than to one day after giving birth to her [...] Erm. You know, I'd probably like to think. If you asked me at the time I'd probably think oh yeah, well don't be so ridiculous, of course I'm coherent of course I can, you know, you think you're pretty amazing once you err, the day after you've given birth actually [laughs], yeah, yeah, you're pretty infallible really so err, dunno." *EA*

The suggestion from these parents is that the timing of newborn screening information, which for most parents in this study took place after the birth of the child, was potentially detrimental to them giving of an informed consent. This alone may bring into question whether an informed choice was given, but coupled with the perceived lack of choice that parents also reported, this potentially raises serious questions about whether at least some parents are providing informed consent to newborn bloodspot screening.

4.8 Decision-making and decisional quality

For some the decision to have the screening had been made prior to the formal provision of information. Despite having made their decision these parents wanted to know about the implications of the decision. In explaining this, one mother suggested that there is a perceived need for testing that doesn't necessarily have to be informed prior to making the decision:

> "I: Yeah. Was that, did you speak to her at the same time or was that before or after.
>
> JG: Same time, around the same time so a mixture of all of them - it wasn't gonna change my decision though.
>
> I: No, you'd made your decision.
>
> JG: Yeah, but I wanted to know what I was letting myself in for.
>
> I: OK, yeah, it was more about preparation rather than making your decision.
>
> JG: Yeah, yeah, yes definitely.
>
> I: Right, so with the [JG talks to friend about coffee] so with the erm, decision you said it was sort of, you wanted to make sure everything was all right, was there anything that stood out that 'oh this is an important thing because x' that made you say yes - was there anything in particular?

> JG: No 'cos I'd made my mind up before that before I knew about the information." *JG*

The decision to accept the screening appears to have already been made by this mother and on the basis of a predetermined principle. In exploring this the mother states that:

> "I think it's just because of the person I am I think I hadn't wanted to refuse any tests for [daughter]. I do for me because I, I'm making the decision for myself, but for [daughter] and I think I had to consider my partner as well [...] I knew what he would say anyway as he would want all the tests erm, but erm, I wanted to find out what the thinking was current thinking about it." *JG*

And later

> "I:[...] you say you wanted to accept all the testing, was that because you had a, it was sort of testing's good an it's there for a reason or was it something else?
> JG: Yeah, I think I'd been brought up like that [...] And with my training as well erm, it's there for a reason, when I read the article about pain I erm I sort of, I did hesitate. [...] 'Cos I like to be unconventional as well, as much as possible, but I like to be safe." *JG*

Thus the process of active information gathering for this parent is not to inform the decision, the decision has already been made to accept the screening, but to make that decision an informed one. The driver behind the decision is a predetermined principle, beneficence. Beneficence has been described as "a group of norms for providing benefits and balancing benefits against risks and costs" (Beauchamp and Childress, 2001, p12) and was seen to be invoked by parents as a reason for screening on the basis that treatment was available. In this instance it appears that attitudes towards medicine in general are determinants of the attitudes towards screening, and as such no specific information about screening is required.

A positive attitude towards medicine, together with a general attitude to act to maximise benefit, saw this parent accept the screening on principle, and then inform themselves by gathering information.

This support of beneficence appears in contrast to the principles invoked earlier in the pregnancy:

> "Cos I wouldn't be getting rid of it anyway, so I wouldn't bother. But once you've had the baby, you've got it, you want to cope with it [...] I suppose then it wouldn't matter [...] Cos you need to know, wouldn't you, if there was something wrong but you didn't know what" *NW*

> "I know it's really weird, but the sixteen week test I don't, wouldn't wanna know because I'm having the baby anyway [...] But when she's born, I'd rather know if there was anything wrong with her to try and treat her." *LW*

So again, post-natally there is a focus on acting on information and actively-seeking testing. This is a change from pregnancy when the principle of nonmaleficence; that is the norm of avoiding the causation of harm (Beauchamp and Childress, 2001), appears to be invoked so that they do not cause harm to the child through the application of potentially risky testing. Once the child is born, a level of risk, or even harm, is to be tolerated so long as the perceived benefits outweigh these risks or harms. Yet beneficence was not all conquering, and the specific context may mean that detailed information about the process can still be important. In describing her encounters with clinicians regarding the provision of vitamin K one mother noted that:

> "[...] when we were in hospital the first time we said we didn't want him to have the K, vitamin K injection [...] And erm, there was this big gasp of you know, well nobody says they don't want it [...] It was really really strange so I thought well its a bit odd really an they just

> thought well why don't you want it, I said well I don't want him to have injections and there is an alternative and they can have it orally [...]" *AG*

Hence, for this parent, the availability of alternative modes of delivery meant that the principle of nonmaleficence was again invoked, suggesting an interaction in parental reasoning. Whilst there is a low perception of distress and the lack of alternatives it may be that the principle of beneficence is of greater consequence. With a greater distress/benefit ratio and alternative forms of administration then the principle of nonmaleficence becomes more prominent until a tipping point is reached whereupon parents are no longer willing to proceed. All of which suggests that there is a complex process of checks and balances covering not only the calculation of risk estimates in terms of likelihood, but also size of effect and potential alternatives as well as underlying principles that parents assess when considering the heel prick.

4.9 Summary

Parental decisions were found to have a range of influences, with these influences weighing differently for different parents. Taking these thematic analyses as the starting point, and assessing how different narratives were linked, a composite was developed in order to produce the proposed causal maps of influences on parental decision quality for newborn bloodspot screening.

4.9.1 Model 1

Figure 4.1 proposes that experience (EXP), informed by prior personal experience as well as those of friends and family, affects parental decisional quality (DCQ) indirectly through its effect on attitudes to medicine (ATTMED) and information seeking behaviour (INFSK). This proposed relationship can be seen in the way that positive attitudes towards the midwife and the NHS were reflected not only

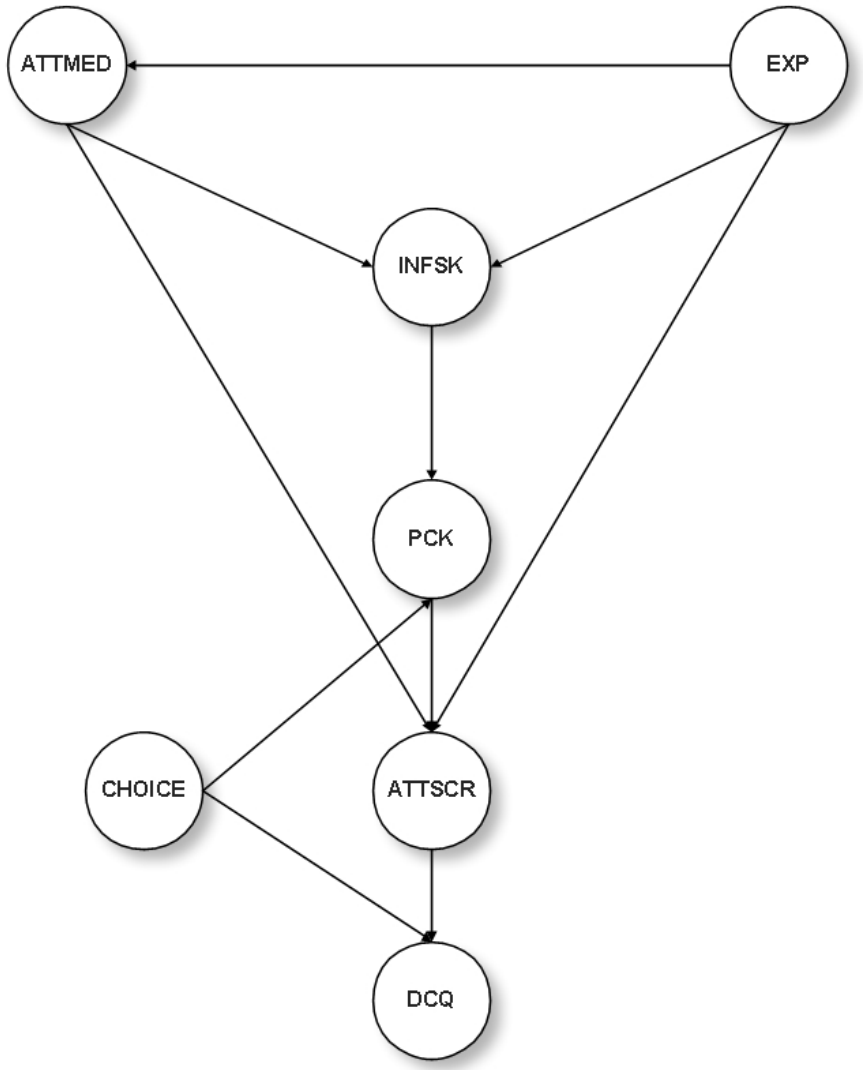

Figure 4.1: *Model 1. An initial proposed causal map for the factors affecting parental decisions to accept the heel prick*

in the use of the midwife as a primary information source but also with a theoretical reduction in information-seeking; those parents with more positive attitudes towards the midwife or hospitals discussed fewer information sources suggesting a decrease in information-seeking behaviour with increasing positive attitudes towards medicine. This corroborates with the classification of those parents as *passive receivers*. The analysis here also suggests that parents with multiple children may rely on their own experience more than other information sources, again potentially reducing information seeking behaviour. This finding gains some support from research into decision-making for prenatal testing in which relevance of personal experience was a significant factor in decisions relating to prenatal testing. (Etchegary et al., 2008; Potter et al., 2008). For some parents experience was reported as allowing a greater focus on the information provided. Both of these scenarios implicate experience, personal or otherwise, in the decision-making process.

Perceived knowledge (PCK), is postulated to be affected by experience indirectly through its relationship to information-seeking behaviour and attitudes towards medicine. As argued in Chapter 2, assessment of knowledge through recall of specific information is problematic. From the discussions with parents perceived knowledge is proposed to be more appropriate to perceptions of decisional quality. Perceived knowledge is argued to have a causal relationship with attitudes towards screening. This proposed relationship is demonstrated by the way parents demonstrated knowledge about implications of the diseases when discussing attitudes towards screening at birth.

Attitudes towards screening (ATTSCR), derived through perceived risks and benefits, are deemed to have a direct influence on decisional quality. This was exemplified by those parents who made a conscientious risk/benefit analysis. However, this may have been informed by attitudes towards medicine with positive

attitudes towards medicine more generally being reflected in positive attitudes towards screening. Consequently, attitudes towards screening are posited to be directly dependent on general attitudes towards medicine but also mediated through information-seeking behaviour and perceived knowledge.

Attitudes towards screening are hypothesised to have a direct impact on perceived decisional quality, with those with less certain attitudes anticipated to identify with a reduced perception of decisional quality. This perception of the final decision, however, is also suggested to be impacted on by the perception of choice (CHOICE), with those parents viewing their consent as being poor quality hypothesised to show a reduced perceived quality of the overall decision. This quality of consent may be demonstrated through parental perceptions of choice or perceived capacity to make a decision, both of which were issues relayed during the interviews.

4.9.2 Model 2

Model 2 is similar to model one except that information-seeking behaviour and experience are no longer proposed to have a causal influence on perceived decision quality, neither directly nor indirectly, and attitude to medicine is proposed to have a direct causal relationship with perceived knowledge. This is congruent with the supposition that decisions are largely principle-based rather than based on specific information about the screening. The model also proposes a direct relationship between perceived choice (CHOICE) and attitudes towards screening (ATTSCR). Choice is also proposed to have a direct influence on perceived knowledge, with the proposed relationship being that those parents who perceive that they have a choice being more likely to have a better knowledge. Parents who don't perceive a great choice are proposed to be less likely to have a good perceived knowledge, possibly due to a perceived lack of need to be informed. Furthermore, the relationship with the midwife is proposed to be influential on the basis of the

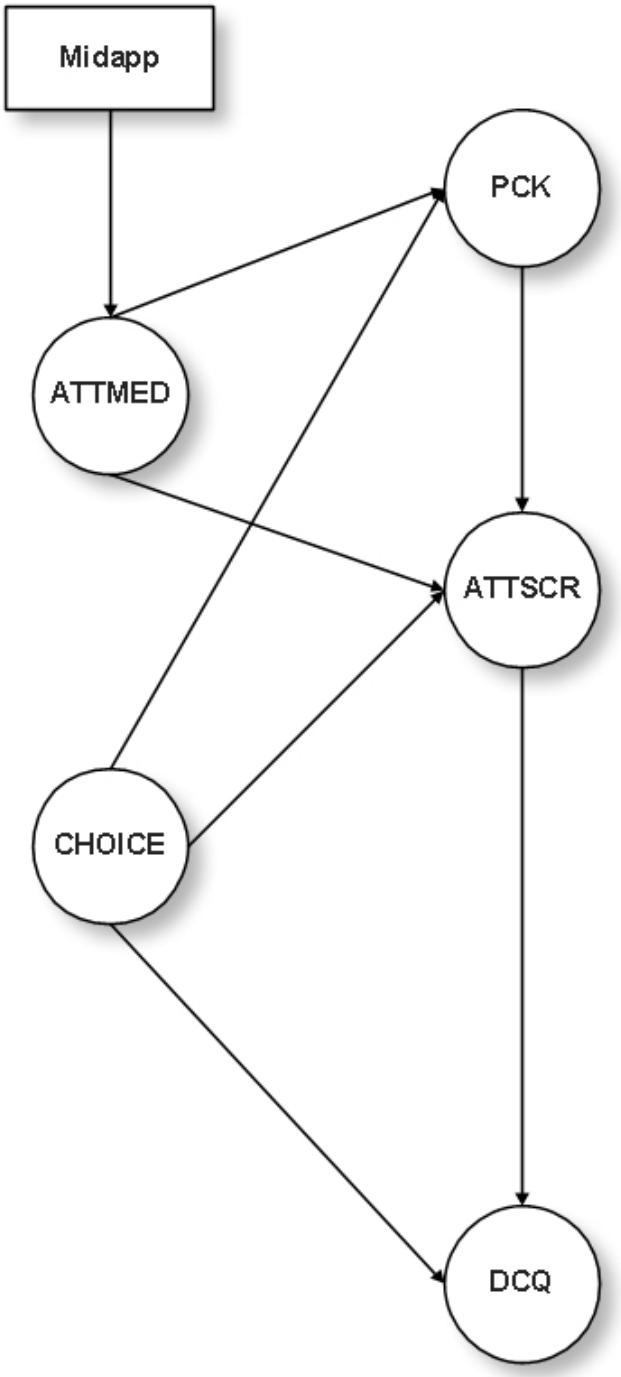

Figure 4.2: *Model 2. An alternative model proposing causal relationships between the factors affecting parental decisions to accept the heel prick*

comments made by some parents about why they didn't seek further information. Consequently the number of times they saw the midwife is suggested to be integral to the relationship; where seeing the midwife more often increases the strength of feeling about the relationship, and so is included within the model and proposed to have an effect on their attitudes towards medicine.

4.9.3 Model 3

The final model, shown in Figure 8.6, proposes that attitudes to medicine (ATTMED) and attitudes to screening (ATTSCR) are not exclusive concepts. Instead it is proposed that a single latent constructs of attitudes to medicine (ATTMED) is a determinant of responses relating to trust and specific attitudes towards screening. Again the midwife relationship is proposed to be integral to the determination of these attitudes.

These models lay out the proposed mechanisms through which the latent constructs interact and culminate in the perceived quality of the parents' decision to accept newborn bloodspot screening. These proposed relationships, together with the compositional elements derived from the interviews serve as the starting point for the development of the questionnaire and subsequent analyses detailed in the upcoming chapters.

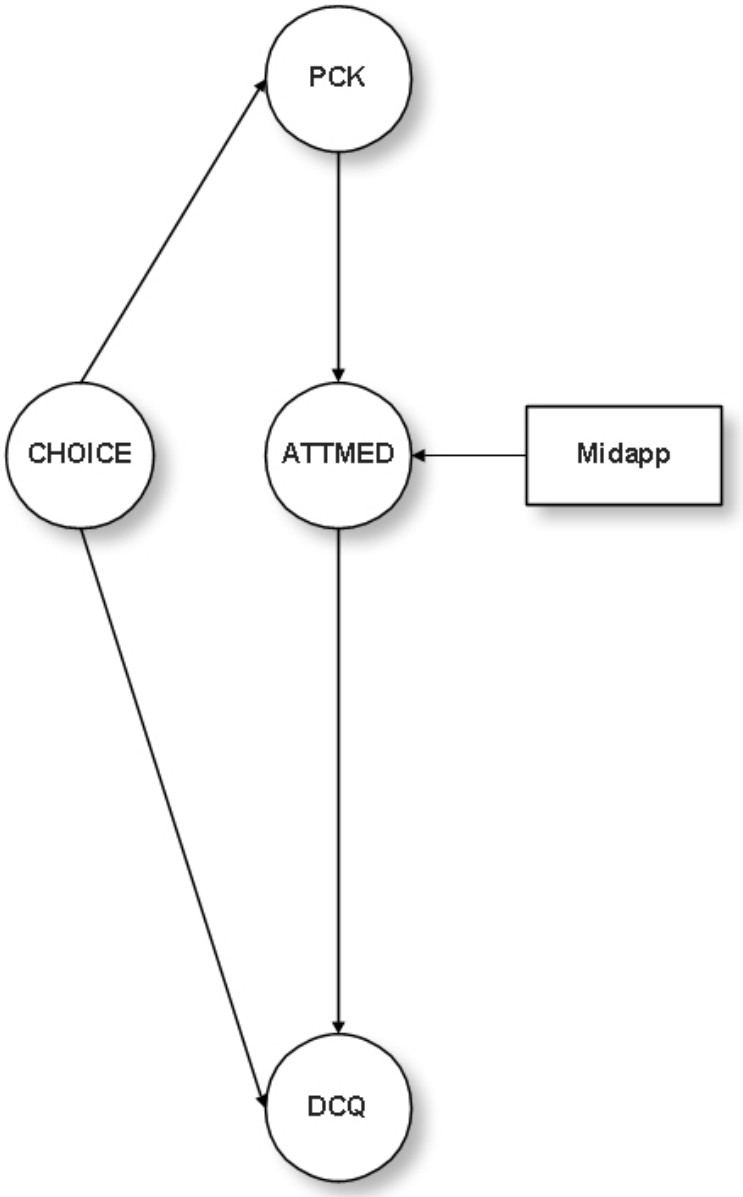

Figure 4.3: *Model 3. A simplified model proposing causal relationships between the factors affecting parental decisions to accept the heel prick*

Chapter 5

Defining the measurement model: Methods

5.1 Introduction

The aim of the first phase of the study was to gather an understanding of the parental experiences of newborn bloodspot screening and how decisions were made regarding whether or not to consent to having the screen. The outcome of this first phase was an understanding of the factors that parents identified as affecting their decisions and the way in which these interacted. This culminated in the proposed causal models presented in Chapter 4.

Despite the strengths of these findings, and the explanations they provide about the influences on parental decision-making about newborn screening, they provide little insight into the strength of these influences across populations. In order to gain insight into this one needs to conduct statistical analyses to provide explanations of the generalised patterns, or demi-regs (Lawson, 1997), that appear within the larger populations. To collect this data parents completed a postal questionnaire the results of which were analysed using structural equation modelling.

5.2 Why a questionnaire?

Questionnaires are highly structured tools. This structure comes not only in the successionist approach to the question sequence but also the finite and fixed set of responses which are available to the respondent. A limitation of this is that one has no way of probing or clarifying interpretation of the questions. Furthermore, with self-administered surveys one cannot guarantee that the individual completing the questionnaire is indeed the person to whom the questionnaire was sent. Postal questionnaires also suffer from a much lower response rate than interviews (Weisberg et al., 1996; Bryman, 2004; McColl et al., 2001).

So, given these limitations, why conduct a postal questionnaire? Perhaps the overwhelming argument for the use of this methodology, in appropriate circumstances, is the fact that a very large number of people can be contacted over a large geographic area in a relatively short space of time, at least in comparison to the amount of time that would be required to conduct an equivalent number of in-depth interviews (Bowling, 2004; Bryman, 2004; Czaja and Blair, 2005). Further benefits include the removal of any potential interviewer bias and that some people may be willing to complete a questionnaire on the basis that the data is usually anonymous and aggregated (Bowling, 2004). Another potential advantage to using a postal questionnaire is that respondents can complete the questionnaire in their own time; even doing so in stages. In the current context this could be a potential benefit as the questionnaire could be completed in evenings once the child is asleep.

5.3 Questionnaire development

The development of the questionnaire included formal peer review by the Research Review Committee at Royal Liverpool Children's Hospital NHS Foundation Trust in addition to pilot testing and revision prior to fielding. Following conventional

guidance it was decided that the questionnaire would begin with more simplistic questions, building up a background context and progressing with increasing complexity (Moser and Kalton, 1971).

5.3.1 The latent variable

The influential factors identified in the interview phase were largely latent constructs and consequently have no direct metric. If the latent constructs cannot be measured directly one must generate *indicators* of these or what may be referred to as *manifest variables* (Bartholomew, 1987).

5.3.2 Development of indicators

The present study used the concept of decisional quality (DCQ) as the outcome measure. To this end the Uncertainty and Effective Decision subscales of the Ottawa Decisional Conflict scale (O'Conner, 1995) were used as indicators of perceived decisional quality. The Uncertainty subscale includes items assessing perceived ease, certainty and clarity of choice whilst the Effective Decision subscale includes items relating to satisfaction with the decision and value reflection but also explicitly includes an item evaluating whether the respondent feels they have made an informed choice. Both subscales have been shown to have adequate test-retest correlations and good internal-consistency with Cronbachs alpha coefficients exceeding 0.78.

The use of the full Ottawa Decisional Conflict scale was resisted as the other subscales overlapped with aspects of perceived choice, perceived importance of risks and benefits as well as perceived knowledge. It was felt that these items, which arose within the interviews as key factors affecting parental decisions, needed to be explored in greater depth and in context specific ways and as such indicators were derived for these components.

Multiple indicators were developed for each latent construct identified within the interviews. The use of multiple indicators to represent the latent construct may act to reduce the measurement error inherent in all indicator measures and reduce the reliance on a single indicator (Hair et al., 1998). Using multiple items may also help to improve the validity of the measure by ironing-out idiosyncrasies in response (Fowler Jr, 2001).

Each indicator consisted of an item pool of scores summed to produce a total score. For this reason the total scores are sometimes referred to as *summated scales* (Moser and Kalton, 1971; Hair et al., 1998). Each item consisted of a question or statement which was rated using a five point Likert scale or binary response. Likert scales ranged from Strongly Disagree to Strongly Agree with corresponding scores ranging from 0 to 4. Items were a mixture of forward and reverse coding. Item scores within the scale were averaged and then multiplied by 25 to produce a response range of 0 to 100. In addition data was collected on a range of sociodemographic variables such as age group, number of children, and income.

The specific items within the questionnaire were derived from a range of sources: existing measures in other contexts, the content of the qualitative interviews and prior newborn bloodspot screening literature. A copy of the questionnaire including all questions and coding scheme is included as Appendix H.

The latent construct of *experience* was identified within the interviews when parents talked about how experience affected their decisions and that this was informed by prior personal experience and also those experiences of friends and family. Consequently indicators of experience were derived and indirectly measured through number of children, age of the parent, and whether a friend or family has a condition for which screening is offered.

In a similar way indicator variables were created for the other latent constructs identified from the interviews. *Attitude to medicine* (ATTMED) is indicated by proxy observed variables of attitudes towards the midwife, attitudes towards the hospital and attitudes towards the NHS. The scales have been developed for this study in order to place the concept within the current context. Development of the scale has drawn on existing measures such as The General Trust in Physicians Scale (Hall et al., 2002; Dugan et al., 2005).

The latent construct of *Information-seeking behaviour* (INFSK) is measured by responses to questions regarding prior knowledge, actual behaviour in terms of the number of information sources sought, and the information seeking preferences. The measure of prior knowledge has been developed for this study due to the lack of pre-existing measures in the newborn screening context. This draws on measures from other settings such as those used by Bunn et al. (2002) in their study of factors influencing intention to obtain a genetic test for colon cancer risk. Information seeking preferences are scored using the Informed subscale of Health Opinion Survey (Krantz et al., 1980). This subscale has been found to be reliable, scoring 0.76 on the Kuder-Richardson 20 reliability formula. Number of sources was indicated by parents marking all sources of information used to find out about newborn screening.

Perceived knowledge is indicated by responses to questions regarding perceived knowledge of the procedures, conditions and motivation for screening. These three groupings were indicated by parents as the important informational elements and as such the questions derive directly from the interviews. Questions were presented to allow parents to make a subjective assessment of their knowledge of each of these aspects as opposed to selecting specific pieces of information to be recalled. In each case scales have been developed for this study and measure responses on a five point Likert-type scale ranging from Strongly Agree to Strongly Disagree.

The measurement of *attitude towards screening* (ATTSCR) is indicated by responses to questions regarding the parents' attitudes towards the perceived risks and benefits of screening, together with their child's perceived susceptibility to the conditions being tested for. These scales have been devised for the current study as no scales exist for newborn bloodspot screening. Development of the scale has, however, drawn on existing measures in other screening contexts such as the Revised Susceptibility, Benefits, and Barriers Scale for Mammography Screening (Champion, 1999).

The final component of the model is the *parental perception of choice* (CHOICE). This perceived choice is indicated through two summated scales of parental perceptions of the availability of choice and perceived ability to make a choice. As with previous scales, these have been devised for the current study as no scales exist for newborn bloodspot screening.

5.4 Sampling

5.4.1 Populations

The population selected was that served by the Clinical Biochemistry Department at Royal Liverpool Children's Hospital in their role as a UK Newborn Screening Laboratory.

5.4.2 Sampling frame

The Clinical Biochemistry Department at Royal Liverpool Children's Hospital is part of the UK Newborn Screening Laboratories Network (UKSLN), an association of NHS laboratories who provide screening services to newborn babies. The regional newborn screening laboratory holds records of all infants born in the Merseyside and Cheshire regions. Specifically, the population for the study was

parents who have given birth within the year 2008. Whilst coverage may be incomplete (Coppinger and Cavanagh, 2008); the sampling frame is well defined. One further limitation, imposed following National Health Service (NHS) ethical review, was that all parents whose child had subsequently died were removed from the sample frame. As such the sample frame constituted all children born in 2008 for whom a screening result is recorded by the Merseyside and Cheshire screening laboratory and who had not subsequently died. This provided a total 28348 children from which to draw the sample.

5.4.3 Sample size

The sample has been calculated from both a statistical perspective in order to gain the required confidence levels, but has also been informed by practical constraints. Statistically, asymptotic theory implies that confident conclusions can be drawn from large samples. There is, however, a distinct lack of guidance as to when these samples are large enough, with sample size calculations for structural equation modelling analyses affected by the estimation procedure, model complexity, and degree of multivariate normality within the data (Jaccard and Wan, 1996; Jackson, 2003).

Using a pre-specified power, significance level, and degrees of freedom MacCallum et al. (1996) and Kim (2005) have derived a process through which a sample size can be calculated. A formal routine[1] is provided for the statistical software package R[2]. The R code is provided in Appendix D.

Table 5.1, generated from model 1, indicates that there are 109 degrees of freedom. Using the R code for a power of 0.8 and significance of 0.05 one would need a sample size of N=125. Building in an achievable response rate of 30% (Asch et al., 1997;

[1] http://timo.gnambs.at/en/scripts/powerforsem

[2] R Development Core Team (2009). R: A language and environment for statistical computing. R Foundation for Statistical Computing, Vienna, Austria. ISBN 3-900051-07-0, URL http://www.R-project.org.

Variable counts	Model counts
Variables: 46	Distinct sample moments: 153
Observed variables: 17	Distinct parameters to be estimated: 44
Unobserved variables: 29	Degrees of freedom (153 - 4): 109
Exogenous variables: 24	
Endogenous variables: 22	

Table 5.1: *Model data*

Bowling, 2004), a total sample of 500 was selected with the aim of achieving a final sample in excess of 125.

5.4.4 Sampling procedure

The availability of a well defined sample frame meant that a retrospective, rather than a prospective, sample would be more efficient in terms of approaching a sufficient number of parents in a limited time frame. To achieve this, all parents of children who received a screening result for 2008, and who had not subsequently died, were exported from the laboratory database. A random number generator function was then used to order the names. The first 500 names were then selected as the random sample.

5.4.5 Identification and recruitment

Identification and recruitment of parents was led by Dr Kevin Southern, Consultant in Paediatric Respiratory Medicine, at Royal Liverpool Children's Hospital. At Royal Liverpool Children's Hospital Dr Southern liaised with Elaine Hanmer, Laboratory Information Systems Manager who extracted the names and addresses of parents to approach.

5.4.6 Dealing with non-response

Non-response is the complete failure of a respondent to return the questionnaire. This has the effect of not only reducing the effective sample size but in turn

reducing the quality of the analysis by increasing the margin of error and reducing the generalisability of results (Bowling, 2004; McColl et al., 2001). The calculation of the percentage of non-responders is used to give a response rate although there is little agreement on acceptable response rates (Asch et al., 1997; Sitzia and Wood, 1998; McColl et al., 2001; Bryman, 2004; Bowling, 2004). If the non-response occurs in a systematic way; so that those who do respond differ in some critical way to those who do not respond, then there will be a response bias that will further undermine any conclusions drawn. In order to minimise the level of non-response:

1. The population has been specifically targetted to ensure the saliency of the topic (McColl et al., 2001).

2. A covering letter has been included from Dr Kevin Southern, Senior Lecturer and Consultant in Paediatric Respiratory Medicine and member of the Cystic Fibrosis Board, National Newborn Screening Committee. The inclusion of a covering letter with sponsorship and an influential signatory has been shown to increase response rates (Oppenheim, 1992; McColl et al., 2001).

3. A reminder was sent as studies have shown that there is a positive relationship between the number of post-notification contacts (reminders) and the response rate (McColl et al., 2001). Published evidence also suggests that the inclusion of a questionnaire, at least together with a second reminder, can significantly improve response rates (McColl et al., 2001). However, NHS ethical review determined that it would be unethical to send multiple reminders to parents who had received, but not completed, a copy of the questionnaire. As such only one reminder was sent, but this included a second copy of the questionnaire.

4. A self-addressed freepost envelope was included.

5. Parents who returned a completed questionnaire were entered into a prize

draw to win £20 of high street vouchers. Evidence suggests that the offer of small financial incentives to respondents increases the response rate.

In addition to this care was taken over the design of the survey, with the questionnaire being designed to be as short and efficient as possible and between four to eight pages in length (Czaja and Blair, 2005). The questionnaire was piloted with a convenience and purposive sample of parents with young children to ensure clarity of wording and design.

5.5 Data preparation

The completed questionnaire served as the data collection tool, with responses entered into SPSS version 17 (SPSS Inc, 2008) using the predetermined coding scheme (see Appendix H).

To reduce the error rate a process of double data entry was used. Once the data was entered then the data was screened using 'wild code checking' (Weisberg et al., 1996). In this process the numbers entered as the responses for each question are listed and compared to the predetermined coding scheme. If any questions contain a code that is not from the original scheme this is verified on the original questionnaire response to determine if an error was made during data entry.

5.5.1 Missing data

The way one deals with missing data is dependent on both the amount and the pattern in which it is missing. If large amounts of data are missing then the generalisabilty of the results may be compromised as a result of the sample size being reduced. If missing data is randomly distributed throughout the responses i.e., there is no systematic relationship within the data in terms of how it is missing, then the probability of data being missing does not depend on the value of the data in which there is missingness, nor does it depend on other variables. In this case

the data may be considered Missing Completely At Random (MCAR) (McKnight et al., 2007). This is the least problematic situation as one may remove the cases without introducing a bias or, if the amount is large, use some form of imputation to fill in missing values. The alternative may be that missing data is in some way influenced by another variable within the data set. This form of missingness may be described as Missing at Random (MAR) if the pattern of missingness does not depend on the values of the missing data itself. If data is missing on the basis of the values of the missing data then data may be said to be Missing Not at Random (MNAR). MNAR is problematic as it implies that there is a systematic relation between the causes for data being missing and missing data (McKnight et al., 2007). Any imputation or removal of cases would be seen to introduce bias on the basis that the unknown values are directly related to the fact that the data is missing. Should data be MCAR or MAR and be of an amount that renders complete case analysis inappropriate (i.e., large amounts of missing data) then multiple imputation will be used to impute the missing values.

To evaluate the amount and pattern of missing data the Missing Values Analysis (MVA) option within SPSS is used. If missing data is large $> 5\%$ and can be argued to be MAR then multiple imputation will be conducted using the inbuilt imputation processes of SPSS which allows for the imputation of categorical variables. Should this be required then five imputed data sets will be created, with subsequent data analysis using pooled results.

5.5.2 Scale reliability

Cronbach's alpha

Reliability is assessed through a measure of internal consistency within the summated scales (Birkett, 1986). Cronbach's alpha is a measure of internal consistency and is a common approach taken for scale development within the social sciences (Aldridge and Levine, 2001). Measures of internal consistency are appropriate

here as it may be expected that attitudes may change over time depending on experience and as such test-retest reliability would not be suitable. Cronbach's alpha is not in itself a statistical test, but rather a measure of correlation amongst the items within a summated scale and is computed using the following:

$$\alpha = \frac{k}{k-1}\left[1 - \frac{\sum var(i)}{var(sum)}\right] \tag{5.1}$$

where k is the number of items, $var(i)$ is the variance of an item, and $var(sum)$ is the variance of the totals for each participant.

The result of the Cronbach's alpha is a value that ranges between 0 and 1 (Carmines and Zeller, 1979; Bowling, 2004). As it is not a statistical test of internal consistency, there is a lack of consensus regarding appropriate levels, with some suggesting that a value above 0.6 is sufficient for exploratory research with others even suggesting that a figure above 0.5 is adequate (Hair et al., 1998; Bowling, 2004). As such if the Cronbach's alpha is less than 0.6 then items will be removed if it is indicated that doing so would improve the scales' internal consistency.

5.5.3 Outliers

Following this a process of data screening for multivariate outliers was undertaken. Failing to account for outliers may distort the assessments of normality. Multivariate outliers are those with 'extreme' scores on two or more variables, or with an atypical pattern of responses (Kline, 2005). Analysis for multivariate outliers was conducted through the use of Mahalanobis distance, D^2 (Tabachnick and Fidell, 2001). The Mahalanobis distance is defined as:

$$D^2 = (x_i - \bar{x})'\hat{S}^{-1}(x_i - \bar{x}) \tag{5.2}$$

The Mahalanobis distance is evaluated for each case using the χ^2 distribution. Cases that have $p < 0.001$ are indicative of multivariate outliers (Tabachnick and

Fidell, 2001). Cases identified will have responses individually reviewed. If these appear to be extreme they will be removed.

Chapter 6

Defining the measurement model: Results

6.1 Parent participation

Data collection took place between January and March 2010. Of the 500 parents selected for approach twelve were excluded on the basis that the named individual no longer resided at that address. Of the remainder eligible for inclusion a total of 154 questionnaires were returned giving a response rate of 32%.

6.2 Descriptive statistics

Table 6.1 presents details of the sample characteristics. Parents varied in terms of age, income, number of children and educational level. Data was not available on non-respondents. Demographic characteristics of respondents were compared with data collected as part of the 2001 census, aggregated at the Cheshire & Merseyside Strategic Health Authority (SHA) level. Comparisons were made for the variables of ethnicity and education. Age was not compared due to the distorted sample, attributable to the biological and legal limitations of child-bearing age; that is one expects few parents to be aged over 40 or under 16 years. Due to different grouping between the census and sample data, results are not directly comparable and

as such a qualitative comparison is undertaken. Income is not compared as the questionnaire requested household income data and not individual data. Existing surveys of income, such as the Annual Survey of Hours and Earnings (ASHE)[1] are based on individual level data. As the survey did not request information on the number of contributors to the household income the data is not comparable as it would be unclear if household income was based the income of one or more individuals.

The population who classified themselves as white was similar for both the sample and general population (95.45% and 95.75% respectively). All ethnic groups were within 1% of the population prevalence, although it should be borne in mind that these are based on small sizes within sub-groups in the present study.

Respondents to the survey appeared to be more educated than the general population of Cheshire and Merseyside. Whilst the percentage of parents with GCSE/school level qualifications was similar for both the sample and the general population, both around 20%, there were greater numbers of parents with College/A-level (20.8% within the sample compared to 7.87% within the population) and higher level qualifications (54.55% within the sample compared to 17.73% in the general population). Accurate comparison is difficult as the census data aggregates some school and college data. Within the census data, individuals with one A-level are classed as having a level 1 qualification, the same as someone who had 5 GCSEs but no college education. In the present sample college and school education were separated and this may account for at least some of the discrepancy. Despite this there is still a large difference with regards to the higher level qualifications (degree, higher degree, and professional qualifications). This may in part be due to increasing numbers of University places and degrees awarded since the census, with over 40,000 more degrees awarded in 2008/9 than 2003/4 according to the

[1] http://www.statistics.gov.uk/StatBase/Product.asp?vlnk=15313

Item	**Number**	**(%)**
Age group		
Under 21	2	(1.3)
21 - 30	48	(31.2)
31 - 40	94	(61)
41 - 50	10	(6.5)
Number of children		
1	55	(35.7)
2	68	(44.2)
3	16	(10.4)
4	11	(7.1)
5 or more	4	(2.6)
Ethnicity		
White	147	(95.5)
Black Caribbean	1	(0.6)
Indian	2	(1.3)
Chinese	1	(0.6)
Other	3	(1.9)
Highest educational level		
School/GCSE	31	(20.1)
College/A-level	32	(20.8)
Undergraduate degree	32	(20.8)
Postgraduate degree	20	(13.0)
Professional qualification	32	(20.8)
Other	4	(2.6)
Household income		
Less than £11500	16	(10.4)
£11501 - £18500	20	(13.0)
£18501 - £28000	18	(11.7)
£28001 - £41000	27	(17.5)
£41001 - £75000	44	(28.6)
Over £75000	21	(13.6)

Table 6.1: *Demographic characteristics of valid responses (excludes missing). Percentages may not sum to 100 due to rounding*

Higher Education Statistics Agency (HESA)[2].

The sample is relatively representative in terms of the ethnic mix, but generally older and more educated than the population from which it was drawn. This may indicate a response bias which must be considered when interpreting the results, particularly in relation to questions of knowledge which may be associated with education.

6.3 Data preparation

6.3.1 Missing data

The first stage of the data preparation was to assess both the level and pattern of missingness within the returned questionnaires (excluding those questionnaires that were returned but were wholly incomplete). A total of 111 questionnaires had complete data without any missingness. The patterns of missingness were assessed visually within SPSS.

Figure 6.1 shows the thirty two different patterns for the 43 questionnaires with missing data. Item missingness ranged from 0 to 7.2%. Two items, corresponding with patterns 26 and 28, had over 5% missing data. Pattern 26 relates to Q.16, income level, and pattern 28 to Q.3 concerning the most important source of information. Missing data in Q.3 was due to three mechanisms: in one instance the facing pages of the inside cover and next page were missed completely suggesting the parent had failed to notice these pages. In several cases multiple items had been marked contrary to the question instructions. No response was subsequently recorded on the basis that one could not determine *the* most important source. In the remainder no source had been marked but the preceding and subsequent questions had been completed.

[2]http://www.hesa.ac.uk/index.php?option=com_content&task=view&id=1578&Itemid=161

Figure 6.1: *Patterns of missing data*

A case level analysis showed that 10 cases had over 5% missing data. These are shown in Table 6.2. As can be seen, most are around or just over 5%. Three cases contain large amounts of missing data and account for patterns 29, 31 and 32 in Figure 6.1.

Cases 37, 99 and 102 were removed on the basis that there would be limited information on which to generate imputed values for the missing data.

As the data from the questionnaire was categorical, the appropriate exploration of patterns of missing data was through crosstabulations (not shown). These suggested that missing data in the variable *income* may be associated with educational level, with 12.9% missing in the School/GCSE category. This was compared to the next highest level of missingness in the postgraduate degree category which was

Case number	**Number missing (%)**
76	4 (5.1)
153	4 (5.1)
150	6 (7.6)
61	4 (5.1)
6	6 (7.6)
90	6 (7.6)
13	6 (7.6)
99	16 (20.3)
102	26 (32.9)
37	27 (34.2)

Table 6.2: *Cases displaying* $> 5\%$ *missing data*

missing at 5%. Analyses also suggested that the number of children may be associated with missing data in both declaring one's most important source and income. Increasing number of children was associated with increasing levels of missingness. However, the small numbers are likely to have inflated the percentages and as a result the relationships may not be valid. In order to evaluate further whether the missingness within the identified variables could be construed as Missing at Random (MAR) dummy variables were created for missingness in Q.16 (income) and Q.3 (important source). Using number of children and education level as explanatory variables a binary logistic regression was used to see if the missingness could be predicted by these variables. In both cases none of the variables proved to be significant predictors of the missing data.

Consequently the low general level of missing data, together with the apparent randomness once number of children and education level were accounted for, suggests that the missing data is indeed at random. Theoretically, the apparent lack of consistent mechanism for the missing data is also suggestive that the data is missing at random. As such, imputation would be appropriate in order to generate a complete data set.

Five imputed data sets were created using the in built SPSS multiple imputation

procedure for categorical variables. This created a dataset that in effect contained five datasets, each defined by a newly created variable "imputation".

6.3.2 Outliers

Computation of the Mahalanobis distance revealed four cases that were consistent outliers across the five imputed data sets. Individual reviews of these cases indicated that all were valid for inclusion and as such the final sample was n=151.

6.3.3 Scale reliability

During the process of data input several items appeared to be problematic. Within the questions relating to choice, the statement: "The heel prick is a routine test for all babies", appeared to be agreed with, irrespective of perceptions of whether the test was optional or whether parents felt they had a choice to decline the test. Upon reflection it was felt that the wording of the statement, and in particular the use of the term "routine" had been ambiguous and problematic in terms of scale consistency. This was confirmed through assessment of internal consistency using Cronbach's alpha which indicted that this item had an item-total correlation of 0.077, indicating that this was poorly correlated with the other items. As a result the item was removed improving the mean Cronbach's alpha to 0.730.

The individual subscales of hospital and NHS trust were inadequate. A revised composite scale, which combined the subscales of trust in hospitals and trust in the NHS was assessed. This suggested that the item "The National Health Service (NHS) only provides tests that are important" was inconsistent with the other items, having an item-total correlation of -0.048. Removing this item achieved an adequate alpha and so is incorporated and redesignated as a scale of trust in the hospital system. A full list of the questions, scales and alpha scores are presented in Table 6.3.

Scale	Item	Cronbach alpha mean (std.dev)
Inf	Q2 - Instead of waiting, I usually ask the doctor or nurse immediately after an examination about my health† Q2 - I usually ask the doctor or nurse lots of questions about the procedures during a medical examination† Q2 - I'd rather be given many choices about what's best for my health than to have the doctor make the decisions for me† Q2 - I usually don't ask the doctor or nurse many questions about what he or she is doing during a medical examination†‡ Q2 - I'd rather have doctors and nurses make make the decisions about what's best than for them to give me a whole lot of choices†‡ Q2 - It is better to trust doctor or nurse in charge of a medical procedure than to question what he or she is doing†‡ Q2 - I usually wait for the doctor or nurse to tell me about the results of a medical exam rather than asking him or her immediately†‡	0.710 (0.001)
PrK	Q7 - Before your first baby was born had you heard about the heel prick?† Q7 - Before your baby was born had you heard about testing for phenylketonuria (PKU)?† Q7 - Before your baby was born had you heard about testing for congenital Hypothyroidism (CH)?† Q7 - Before your baby was born had you heard about testing for cystic fibrosis (CF)?† Q7 - Before your baby was born had you heard about testing for sickle cell disease (SCD)?† Q7 - Before your baby was born had you heard about testing for medium chain co-acyl dehydrogenase deficiency (MCADD)?†	0.782 (0.001)
Mot	I feel I understand why the heel prick is done I feel I understand why the test is done at the time it is I feel I understand what the test results mean	0.854 (0.001)
Proc	Q8 - I feel I understand how the test is done Q8 - I feel I understand when the test is done Q8 - I feel I understand when the results will be available	0.816 (0.012)
Cond	Q8 - I feel I understand what the conditions are that the heel prick tests for Q8 - I feel I understand how the conditions could affect my child Q8 - I feel I understand how the condition would be dealt with if found	0.898 (0.005)
Risk	Q9 - The heel prick is likely to cause a lot of pain to my child‡ Q9 - The heel prick causes long lasting pain to my child‡ Q9 - The heel prick is likely to have a long term impact on my child‡ Q9 - The heel prick exposes my child to the risk of infection‡	0.775 (0.003)
Ben	Q9 - The heel prick is a quick test Q9 - The heel prick can provide useful information. Q9 - The heel prick will allow treatment of an identified health problem Q9 - The conditions tested for in the heel prick are serious Q9 - Testing earlier is better than testing later	0.871 (0.002)
Abch	Q10 - I felt I had enough time to make a decision about the heel prick Q10 - I was too tired to make a decision about the heel prick‡ Q10 - I was too emotional to make a decision about the heel prick‡	0.793 (0.007)

Continued on Next Page...

Table 6.3 – Continued

Scale	**Item**	**Cronbach alpha mean (std.dev)**
	Q10 - I did not feel able to make a decision about the heel prick‡	
Avch	Q10 - It was expected that my child had the heel prick‡ Q10 - The heel prick was presented as an optional test Q10 - I felt I had a choice to decline the test	0.730 (0.002)
Unc	Q10 - I am clear about the best choice for me‡ Q10 - I feel sure about what to choose‡ Q10 - This decision is easy for me to make‡	0.907 (0.014)
Eff	Q10 - I feel I have made an informed choice‡ Q10 - My decision shows what is important to me‡ Q10 - I expect to stick with my decision‡ Q10 - I am satisfied with my decision‡	0.898 (0.003)
Mid	Q11 - I want to have all the tests offered by the midwife or hospital Q11 - I completely trust my midwife's decisions about which medical tests are best for my child Q11 - The midwife is totally honest in telling you about all of the different options available Q11 - The midwife will answer my questions to the best of her abilities Q11 - I am confident in the abilities of the midwife Q11 - All in all I trust the midwife completely	0.831 (0.001)
Trustsys	Q11 - I am confident in the test results from the hospital Q11 - I always receive test results from the hospital Q11 - I feel like I have to double check everything the hospital does‡ Q11 - The National Health Service (NHS) provides unbiased information Q11 - The National Health Service (NHS) ensures tests are safe	0.629 (0.006)

Table 6.3: *Cronbach alpha scores for the finalised scales. Items marked † are binary responses agree/disagree or yes/no. Items marked ‡ were reverse coded.*

These scale items were used in conjunction with additional indicator measures relating to the number of sources used (Numsce), the individuals' age group (Age), their number of children (NumCh), and a binary measure of whether they had a friend or family with a screened for condition (Ff). To confirm that summated scale values did not differ between the imputed data sets, a one-way ANOVA was conducted using imputation number as the group variable. Mean values of the scales never differed by more than 1% and all p-values were non-significant indicating scale consistency across imputations.

This confirmed the final set of indicators that were to be taken forward and assessed as within the full structural equation model.

Chapter 7

Structural Equation Modelling: Methods

7.1 Introduction

In the present chapter I consider the analysis of the data collected by the questionnaire, using structural equation modelling (SEM). A major benefit of SEM over traditional statistical methods such as multiple regression is the ability to model complex interactions with multiple independent and dependent variables, and to do this in a single analysis as opposed to a series of analyses.

Structural equation modelling has been used extensively within the psychosocial literature, particularly in relation to decision-making, and has been used within the context of prenatal screening (van den Berg et al., 2008) and genetic testing for cancer (Bosompra et al., 2000; Bunn et al., 2002). The remainder of the chapter outlines how the data collected by the questionnaire will be prepared and analysed.

7.2 Structural equation modelling: an overview

Structural equation modelling, sometimes, mistakenly, referred to as causal modelling, is a general statistical methodology based on the analysis of covariances

and can be seen as a combination of factor analysis and multiple regression (Hox and Bechger, 1998). The method utilises covariances to analyse proposed causal relationships which are specified *a priori* on the basis of an underlying theory or previous empirical data. Whilst two variables may co-vary and may be *evidence* of a causal relationship (Baron, 2000) the development of the causal model needs to be theoretically derived and not driven purely by the statistical analysis. Consequently a purported limitation of SEM is that there needs to be prior theory, and it is because of this need that the first, qualitative, phase of the study was required in order to inform the theoretical aspect of creating a causal model.

A limitation of traditional methods of regression is that that they only model the relationships between observed variables. These methods are inappropriate when one wants to consider relationships between latent variables such as the attitudes or experiences expressed by parents during the interview phase of this study. SEM allows not only the estimation of multiple interrelated relationships in a single analysis (Hair et al., 1998), but also takes into account the relationships between latent variables. In addition, structural equation modelling accounts for measurement error in the latent variable's corresponding indicator variables. To do this SEM models include error terms that become parameters estimated within the model (Raykov and Marcoulides, 2000). In order to clarify the relationships between variables within the model one uses a *path diagram.* A simple example is shown in Figure 7.1

From Figure 7.1 we can see that there is the *structural model* which posits the relationship between the latent variables; in effect the proposed causal models derived from the interviews. Each latent variable also forms part of a *measurement model* that shows the hypothesised relationships between the indicator variables and the latent variable for which they act as proxies. This is, in principle the relationship between the questions within the questionnaire and the proposed latent

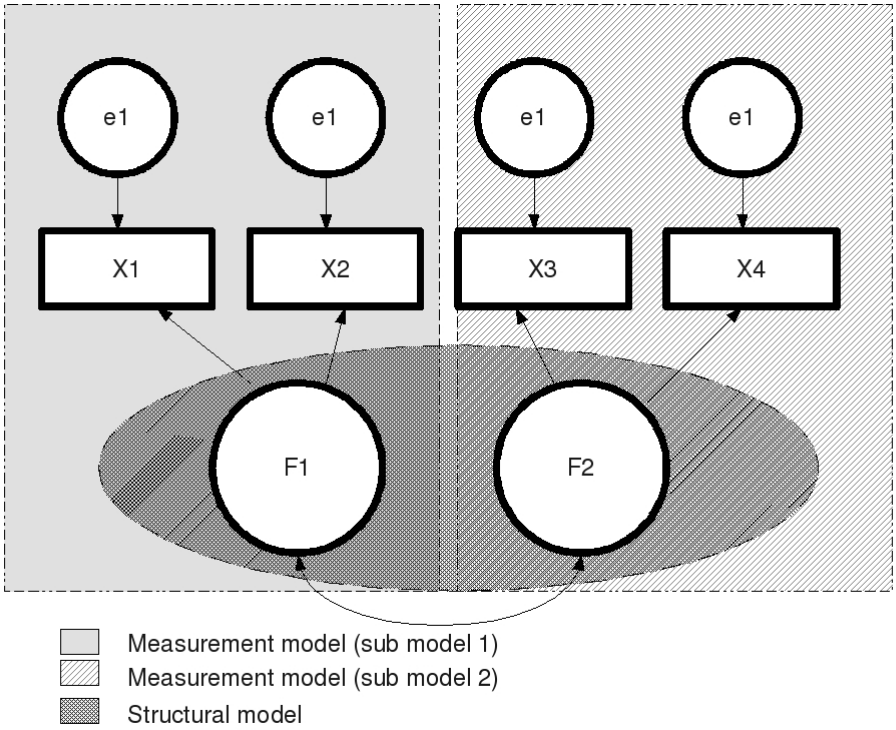

Figure 7.1: *Schematic of a structural equation model*

constructs. Measurement error is explicitly incorporated and modelled for both the observed and latent variables variables through an *error term*, and in the case of latent variables this is referred to as a *disturbance* (Byrne, 2001).

Variables that do not have a causal input within the diagram, that is they have no arrow pointing *towards* them, are described as exogenous variables (Loehlin, 1998). These may be likened to the traditional independent variable as they are causally independent of the other variables within the diagram (Loehlin, 1998; Byrne, 2001). Additionally, one may also have endogenous variables which may be likened to dependent variables. These variables have at least one causal source from within the diagram and is identified through having a single headed arrow directed towards it. One may also link two exogenous variables using a double-headed curved arrow. This indicates a covariance between the variables and is not interpreted as causal.

Using the path diagram as the starting point the parameters, such as path coefficients, are estimated to create an implied population covariance matrix; that is one generates a covariance matrix based on the model. As described in more detail later in the chapter, the parameter values are estimated using a discrepancy, or minimisation, function which aims to minimise the difference between the implied population covariance matrix and the sample covariance matrix generated from the sample data. In short, parameter estimates based on the model are used to generate a model implied population covariance matrix which is then compared with the sample covariance matrix. Whilst a number of discrepancy functions are available this is most commonly achieved through a process of maximum likelihood estimation. The level of discrepancy between the implied covariance matrix and the sample covariance matrix can then be evaluated, principally through the application of the χ^2 statistic, with the aim being to minimise the discrepancy. The χ^2 statistic tests the null hypothesis that the factor loadings, variances, covariances and residuals are valid. The probability associated with the χ^2 represents the probability of a value greater than this, given the null hypothesis (i.e. that there is no difference between the model and sample covariance matrix) is true (Byrne, 2001). The higher the probability, the closer the fit between the implied population covariance matrix and the sample covariance matrix. A range of fit indices have been created to supplement the traditional χ^2 and are discussed in Section 7.7. The availability of fit indices allow for the comparison of different, or competing, models in order to identify models which may be a better approximation to the data.

7.2.1 Modification

The availability of goodness-of-fit measures allows one to make modifications to a model in order to attempt to improve the value of a given index. Such an approach, whilst intuitively appealing, is potentially problematic if one is unable to

explain the modification in a substantive way, that is the modification must also be theoretically valid (Ho, 2006). To this end it has been proposed that a holistic approach to assessing model fit be taken in which the goodness-of-fit indices provide only a partial role, albeit a significant one. In assessing the whole model, individual parameter estimates are also assessed ensuring size and direction are reasonable and consistent with the theory from which the model was derived. The overall fit may be inferred to be good on the basis of a goodness-of-fit measure but if the parameter estimates are not statistically significant or have signs opposite to those predicted then the interpretation is problematic (Bollen, 1989). Consequently all models will be assessed not only through the use of fit indices, but also by evaluating the parameter estimates.

7.2.2 Limits of SEM

Perhaps the biggest issue for SEM is the interpretation of the analysis. No matter what the statistical results are, the attribution of causality must be made in developing and designing the path diagram. The statistical analysis provides evidence to support or contradict the model being evaluated. This limitation of what SEM can say appears to be the most significant, and often misinterpreted, limitation with the potential for authors to misappropriately state that their model proves a causal relationship between variables. Yet if one bears this caveat in mind then there is no reason why SEM cannot be used as a statistical tool to gather evidentiary data that may support or undermine a theory of causal relationships.

The remainder of the chapter provides more detail of the structural equation modelling approach. Starting with model specification I set out each of the competing models to be analysed before turning to the issue of inference in which I discuss issues of identification and parameter estimation. I then conclude the chapter by considering model fit and the evaluation of structural equation models.

7.3 Model specification

Proposing a single model based on the first phase results and obtaining a result that is acceptable in no way determines that this is the 'best' model (Hair et al., 1998). There may, for example, be a number of alternative models that are equally as good, or even better. The approach taken here is to take a *competing models strategy* or what Jöreskog has called an *alternative models* strategy (Byrne, 2001) whereby several competing, or alternative, models are evaluated against a selection of model-fit indices.

7.3.1 Model 1

From Figure 7.2 it can be seen that the model proposes that *Attitudes to Medicine* (ATTMED) is indicated through scores on the scales developed to measure trust in the midwife (Mid) and the healthcare system more generally (Trustsys). The latent construct of *experience* (EXP) is proposed to have a direct causal relationship on attitudes to medicine and is measured through responses to whether the individual has a friend or family member with a screened for condition (Ff), number of children (Numch), and grouped age (Age). Both individual experience and attitudes to medicine are suggested to have a direct influence on the individuals *information-seeking behaviour* (INFSK), indicated by responses to questions on prior knowledge (PrK), their information seeking responses (Inf) in accordance with the Informed subscale of Health Opinion Survey, and the total number of sources of information used (Numsce). This directly affects the latent construct of *perceived knowledge* (PCK) indicated by self-reported knowledge of the motivation behind screening (Mot), the procedures used (Proc) and the conditions tested for (Cond). *Perceived choice* (CHOICE), indicated through responses regarding ones perceived ability to make a choice (Abch) and the perceived availability of the choice (Avch), is proposed to directly affect both perceived knowledge and perceived decisional quality. *Attitudes towards screening* (ATTSCR) are measured through the perceived risks (Risk) and benefits (Ben), and has a direct causal

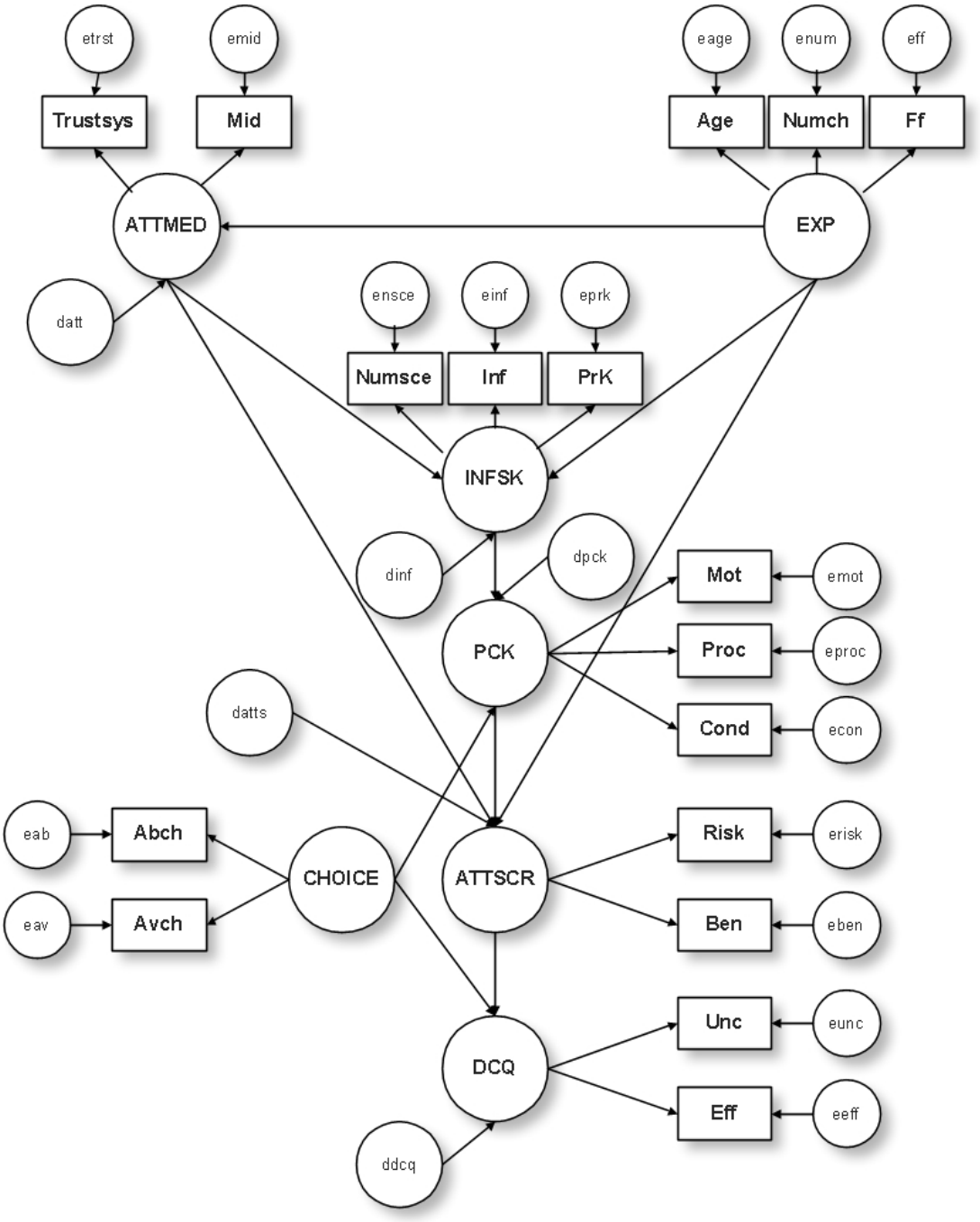

Figure 7.2: *Path diagram showing the full combined structural and measurement components of model 1*

relationship with overall perceived decisional quality (DCQ) which is indicated through responses on the Uncertainty (Unc) and Decision Effectiveness (Eff) subscales taken from the Ottawa Decisional Conflict Scale (O'Conner, 1995).

7.3.2 Model 2

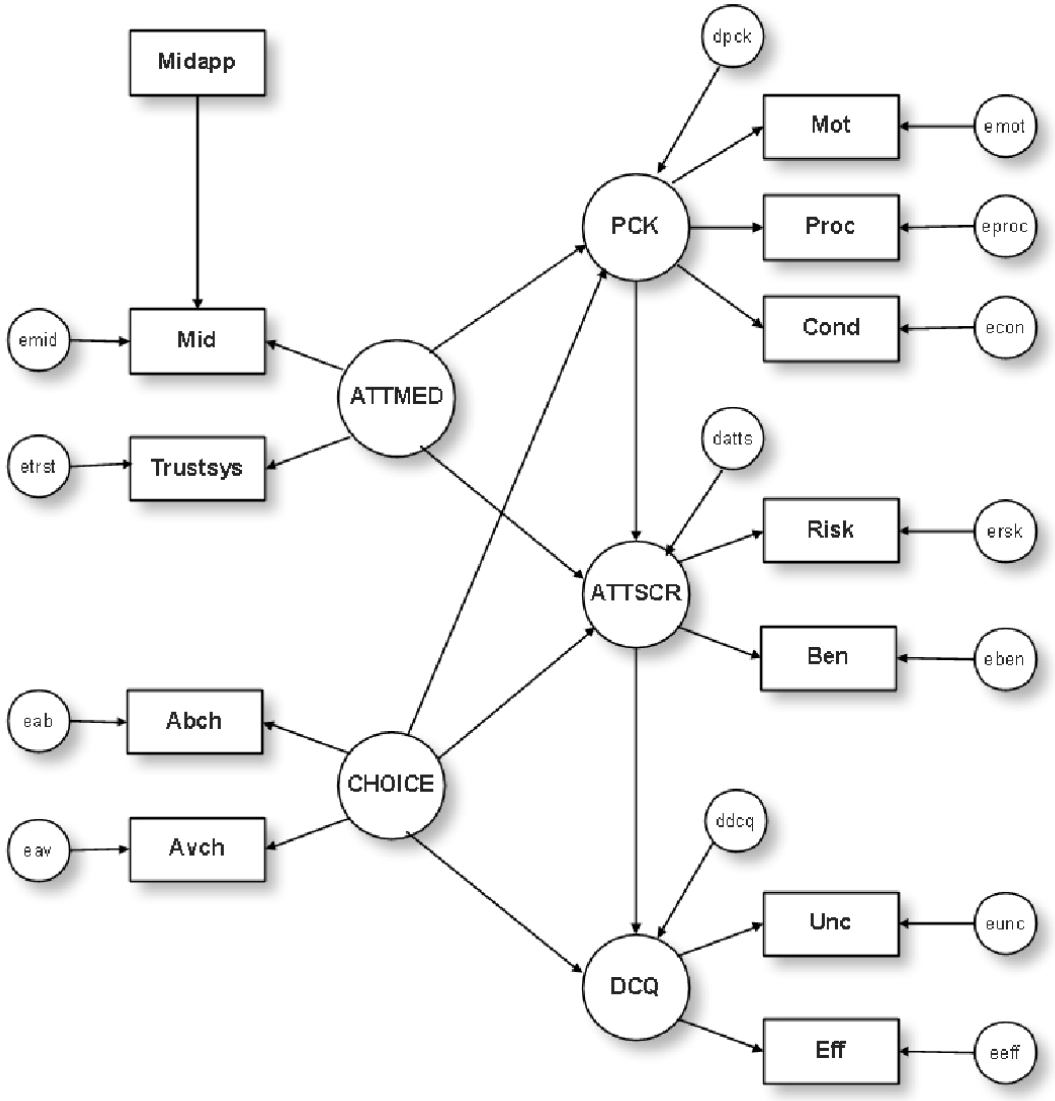

Figure 7.3: *Path diagram showing the full combined structural and measurement components of model 2*

Model 2 differs from model 1 in that information-seeking behaviour and experience are no longer proposed to have a causal influence on perceived decision quality, either directly or indirectly. Attitudes to medicine retains the proposed causal link to specific attitudes towards screening but is also proposed to have a direct

causal relationship with perceived knowledge. This change is in keeping with the description by some parents that decisions had already been made on perceptions of beneficence as opposed to specific information-seeking. Furthermore, choice is proposed to have a causal influence on perceived knowledge. This is implemented on a theoretical basis with the hypothesis that an increase in perceived choice will lead to greater perceived knowledge through the passive uptake of information. The number of times that the mother saw the same midwife (Midapp) is included as an additional covariate and is proposed to be a determinant of trust in the midwife. All other relationships remain the same as in model 1.

7.3.3 Model 3

Model 3 differs from model 2 and model 1 by combining the previously defined constructs of attitudes to medicine (ATTMED) and attitudes to screening (ATTSCR). It is proposed that a single latent constructs of attitudes to medicine (ATTMED) is determinant of responses to the indicators of trust in the system (Trustsys), trust in the midwife (Mid) together with the perceived risks (Risks) and benefits (Ben) of screening. This is in keeping with current models of parental decision-making that aggregate specific and general attitudes. This aggregation will allow for a comparison with model 2 and assess the potential for differing effects between specific and general attitudes. Choice is no longer proposed to act on this combined construct. All other indicator/latent variable relationships remain as with model 2.

7.4 Inference

As already alluded to, analysis starts with the specification of the model and the posited relationships between variables. The next stage is to statistically analyse these relationships. In the next section I review this process, beginning with the process of path analysis.

7.4.1 Path analysis

Whilst the components of the path diagram were outlined in Section 7.2, the process for analysing the diagram has yet to be discussed. In this next section I introduce this process through *path analysis* which forms a major component of structural equation modelling. In path analysis the hypothesised relationships between observed variables are quantified through the analysis of correlations. In order to clarify the relationships within the model these relationships are usually represented pictorially using a path diagram.

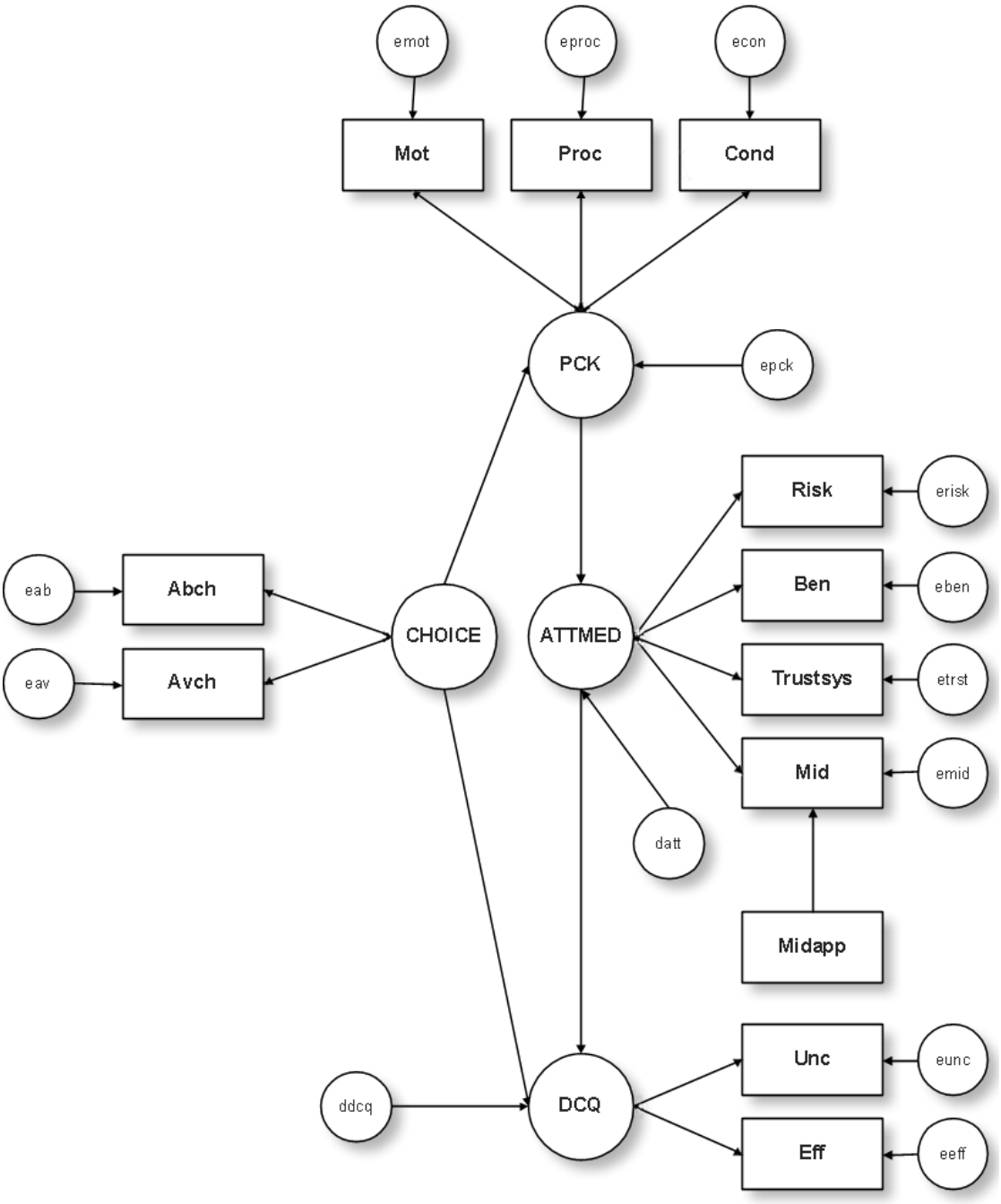

Figure 7.4: *Path diagram showing the full combined structural and measurement components of model 3*

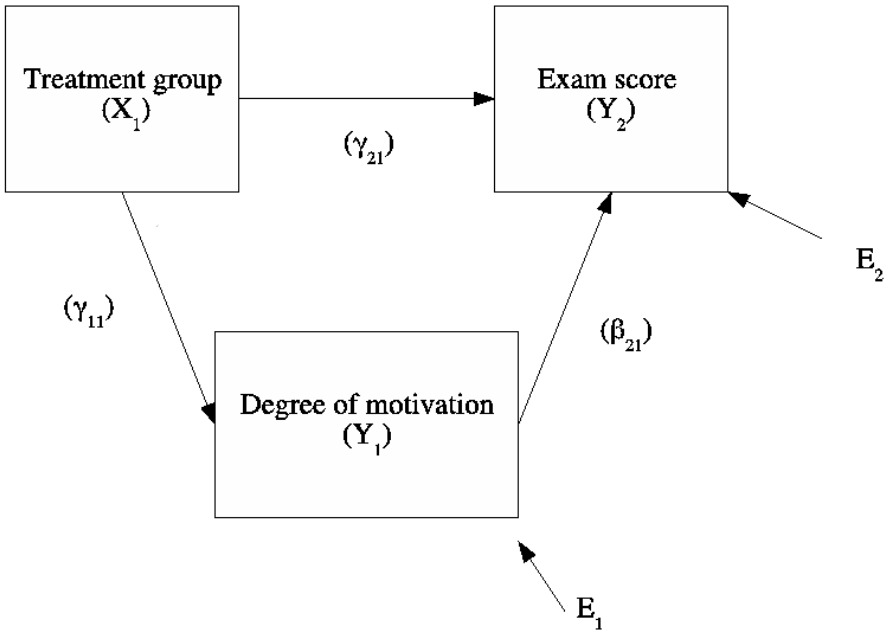

Figure 7.5: *A simple path diagram using observed variables only*

Figure 7.5 is taken from Tabachnick and Fidell (2001) and shows a simple example of a path diagram. In this example exam score (Y_2) is modelled to be an outcome of degree of motivation (Y_1) but also a study skills treatment group (X_1). Both exam score and degree of motivation are assumed to be measured with some degree of error and so explicitly include error terms E_1 and E_2 which are assumed to be independent from the variables (i.e. they do not covary). In analysing the model one wishes to take account of both the *direct* effects of the treatment group, but also the *indirect* effect via the variable *Degree of motivation*. In specifying the model each of the dependent variables, Y_1 and Y_2 can be written as a function of the independent variables thus:

$$Y_1 = \gamma_{11} X_1 + \varepsilon_1 \tag{7.1}$$

where ε_1 corresponds with E_1 in the model, and

$$Y_2 = \beta_{21} Y_1 + \gamma_{21} X_1 + \varepsilon_2 \tag{7.2}$$

.

Using the sample data and covariance algebra it is possible to generate parameter estimates for $\beta_{21}, \gamma_{21}, \text{and} \gamma_{11}$. The covariance of variables X and Y; $COV(X, Y)$, is defined as:

$$COV(X, Y) = \frac{\sum_{i=1}^{n} (x_i - \overline{x})(y_i - \overline{y})}{N - 1} \tag{7.3}$$

where $(x_i, y_i), i = 1, ..., n$ are the sample values on the two variables and $\overline{x}$ and $\overline{y}$ are their respective means.

In the determination of a covariance it is worth remembering:

$$COV(a, X) = 0 \tag{7.4}$$

That is, the covariance of a variable X and a constant a is 0. But also that:

$$COV(aX, bY) = abCOV(X, Y) \tag{7.5}$$

and

$$COV(X_1 + X_2, Y) = COV(X_1, Y) + COV(X_2, Y) \tag{7.6}$$

and noting finally that:

$$COV(X, X) = VAR(X) \tag{7.7}$$

Returning to the example, one can show that the parameters of interest from the regression equations can be re-written in terms of the variances and covariances:

$$COV(X_1, Y_1) = COV(X_1, \gamma_{11}X_1 + \varepsilon_1) \tag{7.8}$$

applying Equation 7.6 this can be re-written as:

$$COV(X_1, Y_1) = COV(X_1, \gamma_{11}X_1) + COV(X_1, \varepsilon_1) \tag{7.9}$$

where we assume that $COV(X_1, \epsilon_1) = 0$. Consequently this can be further revised on the basis of Equation 7.5 so that:

$$COV(X_1, Y_1) = \gamma_{11}COV(X_1X_1) \tag{7.10}$$

which by Equation 7.7 gives us the variance so that:

$$COV(X_1, Y_1) = \gamma_{11}\sigma_{x1x1} \tag{7.11}$$

so that the covariance of X_1 and Y_1 is equal to the path coefficient γ_{11} multiplied

by the variance of X_1. Ultimately this can be re-arranged in order to calculate γ_{11} thus;

$$\gamma_{11} = \frac{COV(X_1, Y_1)}{VAR(X_1)} \tag{7.12}$$

This can be extended to calculate the remaining path coefficients, although the algebra soon expands and for brevity is not shown here. Rather the example is used to demonstrate that one can estimate the path coefficients from the covariance and the variance. One can then use these parameters together with the proposed model to create the implied population covariance matrix.

7.5 The structural model

Path analysis, whilst useful, is limited to the analysis of the observed variables. This can, however, be extended to include latent variables. This is considered by decomposing the full structural equation model into its constituent structural and measurement components and considering how one includes the latent variables within these.

The structural component of the model posits the relationship(s) between the latent variables. This is formally defined as:

$$\eta = \alpha + \mathrm{B}\eta + \Gamma\xi + \zeta, \tag{7.13}$$

where η is an $m \times 1$ vector of latent endogenous variables, ξ is an $n \times 1$ vector of latent exogenous variables, α is an $m \times 1$ vector of intercept terms, B is an $m \times m$ matrix of coefficients that give the influence of ηs on each other and Γ as an $n \times m$ matrix of coefficients for the effect of the ξ on η. Finally, ζ is an $m \times 1$ vector of disturbances that contains the unexplained parts of the ηs (Bollen, 1998; Heck and Thomas, 2009; Ullman, 2001). This model also assumes that, as with the measurement model, the expectation of ζ is 0 and that errors and exogenous

latent variables are independent, i.e. $\text{cov}(\xi, \zeta')=0$. The $n \times n$ covariance of the exogenous (independent) latent variables ξ is labelled Φ and the $m \times m$ covariance matrix of the equation disturbances (ζ) is labelled Ψ.

7.6 The measurement model

The measurement model, that is the relationship between the latent construct and its indicator variables, is evaluated using a process of confirmatory factor analysis (CFA). CFA can be considered a limited application of the more general structural equation modelling framework (Everitt and Dunn, 2001). Unlike the structural component the equations are not proposed to have a causal direction, but rather allow the latent constructs to covary.

In CFA, a smaller number of latent factors (n_q) are hypothesized to be responsible for the specific pattern of variation and covariation among a set of observed indicator variables within the sample covariance matrix. Each of the observed variables is considered as a linear function of one or more factors (Heck and Thomas, 2009). The measurement model is usually expressed as two equations, one accounting for indicators of latent endogenous variables, and one for indicators of latent exogenous variables. Extending the previous notation:

$$y_i = \Lambda_y \eta_i + \varepsilon_i, \tag{7.14}$$

$$x_i = \Lambda_x \xi_i + \delta_i, \tag{7.15}$$

where y_i, is a vector of observed endogenous (dependent) variables, Λ_y is a matrix of factor loadings, η_i is a matrix of factor scores and ε is a vector of errors for observed variables that represents the combined effects of specific factors and random measurement error and is uncorrelated with the other variables. Equally, x_i, is a vector of observed exogenous (independent) variables, Λ_x is a matrix of

factor loadings, ξ_i is a matrix of factor scores and δ is a vector of errors for observed variables. Consequently the observed variables are linked to the underlying factors through the factor loading matrix (Λ). The covariance matrix of ε is denoted Θ_ε with Θ_δ denoting the equivalent for δ. Equation 7.14 can be extended with the inclusion of covariates:

$$y_i = \Lambda\eta_i + \mathrm{K}x_i + \varepsilon_i, \tag{7.16}$$

where K is a matrix of regression coefficients relating the covariates to latent variables and x_i is a vector of the covariates.

Thus if we wish to consider the covariance matrix of the observed variables we can employ covariance algebra. This will allow us to develop the relationship between the data matrix and the population (Reyment and Jöreskog, 1993). Here I extend only the notation for Y for the purpose of brevity.

$$\mathrm{Y} = \eta\Lambda' + \varepsilon \tag{7.17}$$

and

$$\mathrm{Y}' = \Lambda\eta' + \varepsilon' \tag{7.18}$$

In terms of variances and covariances:

$$\frac{1}{N}\mathrm{Y}'\mathrm{Y} = \Lambda\left[\frac{1}{N}\eta'\eta\right]\Lambda' + \Lambda\left[\frac{1}{N}\eta'\varepsilon\right] + \left[\frac{1}{N}\varepsilon'\eta\right]\Lambda' + \frac{1}{N}\varepsilon'\varepsilon \tag{7.19}$$

In line with asymptotic behaviour, increasing sample sizes would, in general, lead to variances and covariances more likely to be closer to the population values than those of a smaller sample. Thus if we let the size N increase indefinitely, each term in 7.19 will converge towards its population value. Hence

$$\frac{1}{N}\mathrm{Y}'\mathrm{Y} \rightarrow \sum, \qquad \frac{1}{N}\eta'\eta \rightarrow \Psi, \qquad \frac{1}{N}\eta'\varepsilon \rightarrow 0, \qquad \frac{1}{N}\varepsilon'\varepsilon \rightarrow \Theta \tag{7.20}$$

and we have

$$\Sigma = \Lambda\Psi\Lambda' + \Theta \tag{7.21}$$

where Λ and Λ' are the factor loading matrix and its inverse respectively, Ψ is a $q \times q$ covariance matrix of factors and Θ is a $p \times p$ covariance matrix of the error terms assumed to be normally distributed with a mean of 0 and some variance (Heck and Thomas, 2009).

The significance of these formulations is that if one has details of the factor loadings and factor scores one can use these to model an implied population covariance matrix. Comparing the model implied population covariance matrix and the sample covariance matrix we can summarise the degree of correspondence between the two matrices. As the level of correspondence increases the population covariance matrix more closely resembles the sample covariance matrix, or put another way, the model more closely resembles the data. This comparison between the population covariance matrix and the sample covariance matrix is the starting point for evaluation of the model and the development of goodness-of-fit indices.

7.6.1 Identification

A pre-requisite to the evaluation of the structural equation model is that it must be *identified*. Identification is the process of attempting to estimate each parameter for which a correlation or covariance cannot be directly calculated. For a model to be identified there must be an effective number of parameters equal to, or less than the number of sample variances/covariances available from the observed variables (MacCallum, 1995; Raykov and Marcoulides, 2000; Kline, 2005). This is some-

times referred to as the order condition (Hair et al., 1998).

If there is insufficient data to provide parameter estimates the model is said to be *under-identified*. One remedy is to constrain one of the parameter estimates to one. This can be achieved through the standardisation of the latent variable by fixing its variance to 1.0 (although this may take on another non-zero numerical value and not be standardised) (MacCallum, 1995). An alternative method is to fix the value of a path coefficient parameter (again usually to one) which constitutes a direct influence onto an observed variable by the proposed latent variable construct (Byrne, 2001; MacCallum, 1995). This can create a model which is *just-identified* and consequently a perfect fit to the data (Hair et al., 1998; Raykov and Marcoulides, 2000). Not only is this uninteresting, but it is somewhat problematic as one cannot test the plausibility of the model and consequently it cannot be rejected (Raykov and Marcoulides, 2000; Byrne, 2001).

In structural equation modelling one seeks to achieve a model that is *over-identified*, that is one wishes to have more information than parameters to estimate. To check whether the model is over-identified one calculates the *degrees of freedom*:

$$\frac{p(p+1)}{2} - (\text{Number of model parameters}), \tag{7.22}$$

Where p is the number of observed variables. So, for example, model 1 in Figure 7.2 can be shown to have:

$$\frac{17(17+1)}{2} - (44) = 109 \quad df$$

The degrees of freedom, denoted df, is a measure of the number of free model parameters (Kline, 2005). A just-identified model would have 0 degrees of freedom. An over-identified model would have 1 or more degrees of freedom. Thus in this case the proposed model can be seen to be over-identified and amenable to statistical analysis and model testing.

This appears to present a problem as an overidentified model will not have an exact solution due to differing parameter estimates that can be generated from the sample data. For example if we consider the following equations:

$$x_1 + x_2 = 5 \tag{7.23}$$

$$2x_1 + x_2 = 8 \tag{7.24}$$

$$x_1 + 2x_2 = 9 \tag{7.25}$$

Unique solutions can be found if the equations are considered in pairs but no unique solution exists for all three equations. In the same way, an over identified model will not have a single unique value for the parameter estimates. Whilst not providing a unique solution, it does provide the opportunity to test different models and compare them using specified criteria (Chou and Bentler, 1995). We can, for example, consider the equations to be a combination of parameters and error which provide us with our data. So that

$$5 = x_1 + x_2 + \epsilon_1 \tag{7.26}$$

$$8 = 2x_1 + x_2 + \epsilon_2 \tag{7.27}$$

$$9 = x_1 + 2x_2 + \epsilon_3 \tag{7.28}$$

If we implement the criterion to minimise the residual sum of squares (RSS), where

$RSS = \sum_{i=1}^{n}(y_i - f(x_i))^2$, we find

$$RSS = (5 - (x_1 + x_2))^2 + (8 - (2x_1 + x_2))^2 + (9 - (x_1 + 2x_2))^2 \tag{7.29}$$

Solving for both x_1 and x_2 gives the solution of $x_1 = 2.273$ and $x_2 = 3.273$ with an RSS of 0.36. Smaller values of RSS indicate a better fit and so different models can be evaluated and the RSS values compared (although one needs to control for the number of predictor variables). Hence overidentification brings with it advantages of being able to create and assess different models by comparing the fit of the parameter estimates. This, however, requires one to not only develop methods with which to estimate the parameters, but also ways in which to evaluate the overall fit of the model.

7.6.2 Estimation procedure

In order to find a unique solution in an over-identified model an estimation procedure is implemented, through which the parameter estimates are created. The aim is to identify parameter values which, when entered into the model, minimise the discrepancy between the sample covariance matrix and the implied population covariance matrix. By imposing some form of statistical criteria to control the estimation process, as done in the above example, we describe a *minimisation function.*

The parameter estimates are plugged into the structural equations derived from the model in order to produce the implied covariance matrix. Most commonly this is achieved through the Maximum likelihood fit function (Bentler and Dudgeon, 1996) which minimises the following:

$$F_{ML} = log|\Sigma| - log|S| + tr(S\Sigma^{-1}) - \rho \tag{7.30}$$

where Σ is the implied population covariance matrix, Σ^{-1} is the inverse of the

implied population covariance matrix, S is the sample covariance matrix and ρ is the number of measured variables.

The benefit of maximum likelihood estimation over other methods, such as ordinary least squares; where one attempts to minimise the sums of squared deviations, is that the estimates are computed simultaneously, as opposed to sequentially. Whilst alternative estimation techniques such as the *Asymptotically Distribution Free estimator (ADF)* have been shown to work well, they require very large sample sizes - in the order of 1000 to 5000 cases (West et al., 1995). Such large samples are beyond the practicalities of this study. Other methods, such as generalised least squares, have been found to produce negatively biased estimates in comparison to maximum likelihood estimation (Chou and Bentler, 1995).

A further advantage of maximum likelihood estimation is its flexibility for interpretation. This is because the procedure is 'scale free' but also 'scale invariant', meaning that the scale of the fitting function is consistent, irrespective of the observed variables' scale (Kline, 2005, p115).

7.6.3 Data assumptions for parameter estimation

In Section 7.2 it was noted that the evaluation of model fit is often based on the χ^2 statistic. This statistic, derived from the fit function, is distributed χ^2 if the data are multivariate normal (Hoyle, 1995). The requirement of multivariate normality means that maximum likelihood estimation is susceptible to errors if data is severely non-normal (West et al., 1995). The importance of this lies in the potential effects that non-normality has on both the parameter estimates and the model fit, potentially over-rejecting models (Hair et al., 1998).

Non-normality appears to have differing effects on different aspects of model estimation. Lei and Lomax (2005) found that whilst severe non-normality affected χ^2

values of model fit, moderate deviations from normality (defined as skewness of less than 0.45 and kurtosis less than 1.5) had negligible effects. Parameter estimates, on the other hand, have been shown to be robust to even severe non-normality (Schermelleh-Engel et al., 2003; Lei and Lomax, 2005). Other studies have suggested even greater robustness of the χ^2 noting only substantial overestimation when skew was 2.028 and kurtosis 2.898 (Muthén and Kaplan, 1985). Variability in the acceptable levels of skew and kurtosis exist with some recommending values of skew less than 2.0 and others up to 3.0 (Kline, 2005) are acceptable. For kurtosis there is less agreement with some suggesting that absolute values less than 7.0 may be appropriate for maximum likelihood estimation (West et al., 1995; Curran et al., 1996; Kline, 2005; Finney and DiStefano, 2006), although these recommendations range as high as 10.0 (Kline, 2005). More recent research has, however, suggested that for models with combined skew of 2.0 and kurtosis of 7.0, the maximum likelihood χ^2 may not be robust, even with large sample sizes (Nevitt and Hancock, 2001). Research has also found that given non-normal data, the maximum likelihood-based standard errors will be underestimated (Finney and DiStefano, 2006). Consequently, parameter estimates appear to be relatively robust to deviations from normality, but χ^2 and standard errors of parameter estimates may be more susceptible to bias from increasing normality.

Consequently the data will be tested for normality. Univariate normality will be assessed through the evaluation of skewness and kurtosis. Visual analysis will be through the production of histograms and will be supported by statistical evaluations in the form of the Kolmogorov-Smirnov and Shapiro-Wilks statistics. If univariate analyses reveal only mild to moderate non-normality (here using a conservative estimate of skew less than 2.0 and kurtosis less than 3.0) then maximum likelihood estimation will be employed. If data is shown to be be severely univariate, and hence multivariate nonnormal, then maximum likelihood parameter estimates with standard errors and a Satorra-Bentler χ^2 will be used. The Satorra-

Bentler scaled χ^2 is discussed in the next section.

7.7 Assessing model fit

Much of the debate around structural equation modelling has focussed on the assessment of model fit, with much disagreement regarding appropriateness. Model fit is usually evaluated using the χ^2 statistic:

$$\chi^2 = (N-1)F_{min} \tag{7.31}$$

where N is the sample size and F_{min} is the minimum fit function.

If, as already suggested, the data is not multivariate normal then the χ^2 statistic in its usual form is inappropriate. To account for non-normality one can employ the Satorra-Bentler χ^2 which corrects the normal theory chi-square by a constant c, that is determined in part by the observed multivariate kurtosis, and the model degrees of freedom. This is defined as

$$Satorra - Bentler \quad \chi^2 = \frac{df}{tr(\hat{U}S_y)}T_{ML} \tag{7.32}$$

where T_{ML} is the traditional maximum likelihood test statistic, df is the degrees of freedom in the model, $\hat{U}$ is the weight matrix and residual weight matrix under the model and S_y is the asymptotic covariance matrix (Hu et al., 1992; Ullman, 2001). This scaled test statistic has been shown to be reliable under various distributions (Curran et al., 1996; Byrne, 2001).

In contrast to the conventional application of the chi-squared statistic one seeks an insignificant difference between the sample and the fitted covariance matrices (Byrne, 2001; Ullman, 2001; Ho, 2006).

Reliance on the χ^2 statistic alone is problematic as the null hypothesis of a per-

fect fit is not credible and most models are useful approximations (Arbuckle and Wothke, 1995). Furthermore, as the value is linked to sample size it can be affected by changes in this. One consequence being that in very large samples the result becomes unreliable, increasing N generally leads to an increase in χ^2, even though the degrees of freedom may remain the same (Raykov and Marcoulides, 2000). Thus in large sample sizes there tends to be an increase in the rejection of models, so that even reasonable models may be rejected if the sample size is large enough. These identified shortcomings of the χ^2 statistic has led to a range of goodness-of-fit indexes being developed [1]. Research has shown that fit indices vary in performance (Hu and Bentler, 1999) with current best practice suggesting that several measures of goodness-of-fit be chosen to evaluate the proposed model (Hu and Bentler, 1999; Kline, 2005). In line with these recommendations the χ^2 statistic will be used and supplemented with an absolute, an incremental, and a parsimonious fit measure.

The *Root Mean Square Error of Approximation (RMSEA)* penalises for model complexity so that, given the same amount of explanatory power, a simpler model would be preferred. In determining the usefulness of the RMSEA value one has to acknowledge that any "cut-off" is arbitrary, and more likely to be based on intuition and experience, rather than a statistical basis (Marsh et al., 2004). Bearing this in mind several "rules-of-thumb" have been suggested when considering RMSEA values. Browne and Cudeck (1993) have suggested that an upper limit of 0.08 is a reasonable error of approximation, suggesting that models with an RMSEA greater than 0.1 should not be employed (Arbuckle and Wothke, 1995). Others authors suggest that RMSEA values of 0.05 or even to 0.06 may be seen as indicating a good level of fit (Hu and Bentler, 1999; Byrne, 2001).

[1] It is beyond the scope of this paper to go into detail regarding the differences between the individual fit measures. For a discussion of the differences and effects of the different measures of fit see Hu & Bentler (1999), Fan et al., (1999), Marsh et al., (2004) and Barrett (2007)

The *Comparative Fit Index (CFI)* is an incremental fit measure. The CFI has a range from 0 to 1.0, with a higher value indicating a better fit of the model. It is worth noting that a CFI of 1.0 is not equivalent to a perfect fit, but rather that $\chi^2_m < df_m$ (Kline, 2005). A general rule of thumb is that a CFI > 0.9 is indicative of a model that is a good fit (Byrne, 2001; Kline, 2005). The CFI has also been shown to be robust to non-normality (Lei and Lomax, 2005).

The *Akaike information criterion (AIC)* is a comparative measure (Hair et al., 1998) and so has no fixed metric. Lower values indicate better fit.

Table 7.1 describes the formulae for each fit index, together with information on interpretation.

Index	**Formula**	**Comment**
RMSEA	$\sqrt{\frac{\hat{\delta}_m}{df_m(N-1)}}$	$\hat{\delta}_m$ =max$(\chi^2_m - df_m, 0)$ with χ^2_m representing the chi-square statistic of the model, and df_m the degrees of freedom within the model. < 0.05 indicates good fit, > 0.08 indicates poor fit.
CFI	$1 - \frac{\hat{\delta}_m}{\hat{\delta}_b}$	$\hat{\delta}_m$ =max$(\chi^2_m - df_m, 0)$ where χ^2_m represents the chi-square statistic of the model, and df_m the degrees of freedom. $\hat{\delta}_b$ is the equivalent but calculated for the baseline model. Range from 0 to 1.0, a CFI > 0.9 indicates good fit.
AIC	$\chi^2 + 2q$	χ^2 is calculated as before and q is the number of free model parameters. Lower values indicate better fit.

Table 7.1: *Formulas and descriptions for the selected fit indexes*

Should the fit indices indicate a reasonable or good level of fit then the free parameter estimates are evaluated through comparisons between the estimate and it's standard error (Hoyle, 1995). These tests are based on the *unstandardised* parameter estimates as the use of standardised estimates may lead to incorrect

standard errors being derived (Kline, 2005). Unstandardised parameter estimates, such as regression coefficients, are interpreted as in multiple regression with estimates controlled for correlations among multiple presumed causes (Kline, 2005). Unstandardised estimates retain the metric of the variable and as a result estimates can be seen to indicate the number of units change in the dependent variable per unit change in the independent variable (Hoyle, 1995). For the purpose of comparing the effect of coefficients, *standardised* estimates are also produced (Ullman, 2001). Standardised estimates remove scaling and can be seen to correspond to effect-size estimates (Hoyle, 1995). Citing Cohen (1988) Kline (2005) suggests that

> "Standardized path coefficients with absolute values less than .10 may indicate a "small" effect; values around .30 a "typical" or "medium" effect; and "large" effects may be indicated by coefficients with absolute values $\geq .50$. These guidelines are intended for cases where the research has little theoretical or empirical basis to differentiate between smaller versus larger effects, which is most likely to happen in new research areas." (Kline, 2005, p122)

Accordingly parameter estimates will be reviewed not only for their significance but also the direction and size of effect in order to develop a substantive interpretation for the model.

Chapter 8

Structural equation modelling: Results

Following the previous chapters in which a structural model was proposed and the measurement model defined, the final stage is to bring the two aspects together to evaluate the proposed full latent model. In this chapter I first assess the indicator variables to check the assumption of normality. I then present some simple descriptive statistics before evaluating the three proposed models. Initial descriptive statistics were generated using SPSS (SPSS Inc, 2008). Confirmatory factor analysis and structural equation modelling were conducted using MPlus v6.0 (Múthen and Múthen, 2010).

8.1 Normality

Visual assessment of histograms for each indicator (not shown) suggested non-normality. This was then confirmed through descriptive statistics of skew and kurtosis as well as appropriate test statistics, which are shown in Table 8.1. In all cases the variables are found to be non-normal.

The extent to which the variables are non-normal does, however, differ and whilst skew is within the proposed limits within which maximum likelihood estimation is

Indicator	**Skew; (std.dev)**	**Kurtosis; (std.dev)**	**Shapiro-Wilk statistic**	**Kolmogorov-Smirnov**
Inf	-0.367 (0.019)	-0.877 (0.031)	0.149**	0.908**
PrK	0.478 (0.004)	-0.845 (0.012)	0.184**	0.905**
Mot	-0.800 (0.008)	0.728 (0.006)	0.193**	0.898**
Proc	-0.668 (0.014)	1.057 (0.054)	0.239**	0.873**
Cond	-0.183 (0.006)	-0.665 (0.004)	0.116**	0.959**
Risk	-1.076 (0.016)	2.973 (0.097)	0.164**	0.914**
Ben	-0.228 (0.001)	-0.842 (0.008)	0.187**	0.875**
Abch	-0.747 (0.012)	0.671 (0.010)	0.226**	0.920**
Avch	0.080 (0.011)	-0.225 (0.018)	0.110**	0.973**
Unc	0.749 (0.075)	0.918 (0.226)	0.194**	0.904**
Eff	0.617 (0.062)	1.173 (0.170)	0.196**	0.907**
Mid	-0.221 (0.004)	0.530 (0.015)	0.133**	0.967**
Trustsys	-0.315 (0.016)	0.970 (0.014)	0.116**	0.970**

Table 8.1: *Measures of skew and kurtosis for the indicator variables together with associated statistics of normality. Note that kurtosis is normalised from the absolute value (i.e. is -3). * indicates significance at* $p < 0.05$*, ** indicates significance at* $p < 0.01$

robust, kurtosis exceeds the upper boundary of 3.0 in eight of the thirteen indicator variables. This indicates that not only are the indicator variables non-normal, but they are to such a degree that standard maximum likelihood estimation is not appropriate for the analysis. Consequently the Satorra-Bentler χ^2 and robust maximum likelihood estimation are used.

8.2 Descriptives

Prior to the full structural equation modelling analysis, simple descriptive statistics were reviewed in order to gain a better appreciation of the data.

The majority of parents used a limited number of sources with a mean number of 2.0071 sources being used. The number of sources used by parents was not found to differ by age, number of children, education level, or income. The health service appeared to be the most common source of information with the midwife cited by 93.4% of parents who responded to the question and the NHS leaflet by 50%. As with the *most used* sources of information, the midwife was generally

regarded as the *most important* source of information. Of those parents answering this question 76.2% cited the midwife as the most important source. The sources cited as most important generally reflected the pattern seen in those most used, with the NHS leaflet being second only to the midwife. Friends and family were not cited by a single parent as the most important source of information.

There was, however, a significant difference in terms of the most important source depending on how many sources parents used (Fisher's exact test, p=0.004). As the number of sources used increased, so the percentage of parents citing the midwife as the most important source decreased.

Figure 8.1 provides a boxplot of the indicator variable scores. As can be seen, and suggested by the interview data, parents viewed the risks as being minimal and the benefits as being great, indicated by their high median scores. These views are relatively consistent and have a relatively narrow range of responses.

The responses relating to knowledge of newborn screening are also consistent with the interview results, with parents scoring their perceived knowledge to be greater in terms of the reasoning behind the screening tests and procedures as opposed to knowledge about the conditions themselves. Responses to questions relating to knowledge about conditions were not only on average lower but were also highly variable with scores ranging from 0 to 100 and consequently covering all possibilities. Scores on the indicators of perceived knowledge did not differ by education or income.

Differences were also seen in terms of responses to questions regarding choice. Whilst parents tended to feel that they were capable of making a choice, i.e. were competent to do so, perceptions regarding whether they had a choice were much more variable. A mean score of of 42.5 also suggests a distinct limitation of choice

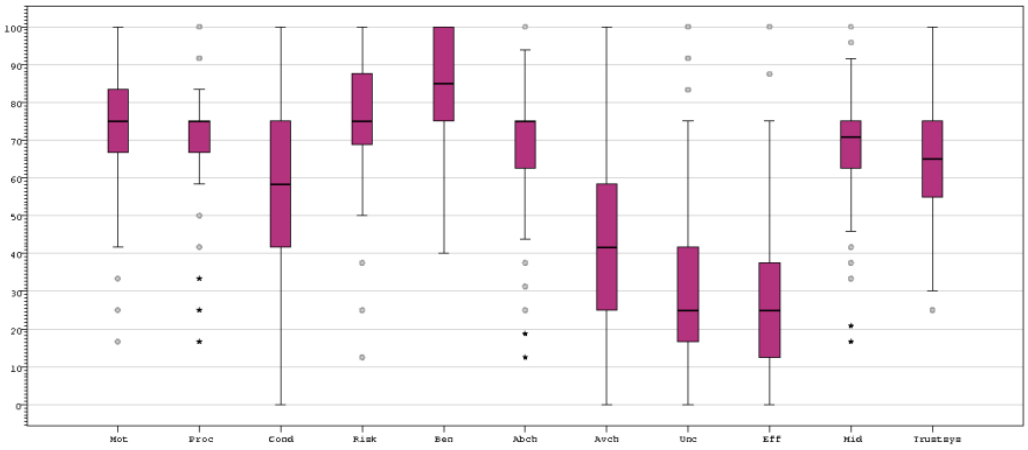

Figure 8.1: *Boxplot of indicator variables (Inf and PrK not included due to different scale metric)*

in some cases. This appears to be largely affected by the way in which screening is presented with 81% of parents agreeing to some extent with the statement that "it was expected that my child had the heel prick" and only 48% feeling that they had a choice to decline the test, with over 30% disagreeing. Any lack of perceived choice points, potentially, to a parent who has not given an informed consent as defined previously.

The average trust shown in the midwife is higher than that in the healthcare system, although there is more variation in relation to trust in the healthcare system, with some very high ratings. However, the scores were similar with significant overlap in the range of scores achieved.

The indicator variables of decision uncertainty and decision effectiveness are in the main scored low, indicating a low degree of uncertainty and perceived ineffectiveness. Both variables are well matched in terms of both average ratings and variance suggesting a consistency in terms of perceived decisional quality.

8.2.1 Correlation matrix

In order to gain a better appreciation of possible associations one can consider the correlations between the indicator variables. Table 8.2 shows the simple bivariate correlations between the variables included within the analysis. As expected, indicators which relate to the same latent variable show an acceptable level of correlation.

The bivariate correlations suggest that the relationship between information-seeking and decision making may not be a simple process. A negative correlation of -0.088 exists between the number of children (*Numch*) and the number of sources used by the parent (Numsce) suggesting that as parents have more children they access a more limited number of sources of information. This, however, is not significant ($p > 0.05$) indicating a lack of any real difference depending on number of children. There is also a positive correlation between the number of children and trust in the midwife (0.060), yet a negative association with more general trust in the health-care system (-0.091). However, both of these correlations are small and again are not significant ($p > 0.05$), suggesting that experience - indicated through number of children - has little association with trust nor manifest information-seeking.

Another expected relationship proposed in model 1 is that increased information-seeking will lead to increased knowledge. Correlations between number of sources of information (Numsce) and the knowledge items of *Mot, Proc* and *Cond* are non-significant at the 5% level, nor are correlations between information-seeking behaviour (Inf) and the three items assessing perceived knowledge. Perceived knowledge also did not appear to be affected significantly by the source of information deemed most important (Kruskall-Wallis non-parametric ANOVA $p > 0.1$ for each knowledge item). This may suggest that the relationship between information-seeking (and provision) and knowledge is either missing, or more complex than predicted.

	Ff	Age	Numch	Mot	Proc	Numsce	Inf	Prk	Cond	Risk	Ben	Abch	Avch	Unc	Eff	Mid	Trustsys
Ff	1.000																
Age	-0.015	1.000															
Numch	0.007	**0.215**	1.000														
Mot	0.032	0.137	0.043	1.000													
Proc	-0.014	0.031	0.068	**0.752**	1.000												
Numsce	-0.048	0.122	-0.088	0.134	0.050	1.000											
Inf	-0.047	0.100	0.067	-0.029	0.027	**0.163**	1.000										
Prk	0.059	-0.009	0.039	**0.303**	**0.247**	0.120	0.045	1.000									
Cond	0.143	0.018	0.065	**0.680**	**0.617**	0.028	-0.132	**0.347**	1.000								
Risk	**-0.198**	0.112	0.048	**0.183**	**0.160**	-0.131	-0.135	-0.035	0.098	1.000							
Ben	-0.083	0.125	-0.005	**0.474**	**0.440**	-0.033	-0.044	0.134	**0.293**	**0.441**	1.000						
Abch	-0.136	-0.035	-0.012	**0.446**	**0.385**	0.125	-0.068	**0.243**	**0.370**	**0.294**	**0.332**	1.000					
Avch	0.013	-0.015	0.031	**0.355**	**0.294**	0.077	-0.079	**0.197**	**0.407**	0.081	0.082	**0.370**	1.000				
Unc	-0.001	0.061	0.113	**-0.572**	**-0.492**	-0.138	0.003	**-0.287**	**-0.426**	**-0.196**	**-0.459**	**-0.455**	**-0.393**	1.000			
Eff	-0.021	0.019	0.024	**-0.596**	**-0.481**	-0.083	0.056	**-0.303**	**-0.471**	**-0.196**	**-0.523**	**-0.383**	**-0.382**	**0.850**	1.000		
Mid	0.058	**-0.210**	0.060	**0.190**	**0.219**	-0.044	**-0.254**	0.093	**0.275**	0.152	**0.294**	0.149	**0.182**	**-0.297**	**-0.393**	1.000	
Trustsys	0.014	-0.034	-0.091	**0.339**	**0.346**	0.086	**-0.244**	0.147	**0.351**	**0.216**	**0.359**	**0.238**	**0.213**	**-0.301**	**-0.359**	**0.517**	1.000

Table 8.2: *Bivariate correlations of the observed indicator variables. Significant correlations are indicated in* ***bold*** *($p < 0.05$)*

Furthermore, the number of sources used appears to show little association with trust in either the midwife or the healthcare system more generally. This may indicate a lack of relationship between ATTMED and INFSK as predicted in model 1. Yet, if one considers the most important source of information, then this is significantly associated with trust in the midwife (Kruskall-Wallis non-parametric ANOVA $p < 0.05$). Perhaps unsurprisingly those parents who cite the midwife as the most important source show the highest levels of trust in the midwife. Those parents citing the internet as the most important source of information showed the lowest levels of trust in the midwife. Such a finding is in keeping with the interview data where parents indicated that contentious topics or where uncertainty existed, may be candidates for internet use. The lower levels of trust in the midwife may, therefore, be conducive to increased uncertainty leading to increased internet use.

Number of children (*Numch*) shows small, but non-significant associations with perceived knowledge of the motivation for screening, procedures, and conditions. Equally *Age* shows small and non-significant correlations with perceived knowledge of the procedures and conditions for which screening is conducted. Whilst age appears to show a small correlation with knowledge of the motivation behind screening, this is found to be non significant also.

The predicted relationship between perceived choice and perceived knowledge is also suggested, with significant positive correlations between perceived choice and perceived knowledge of the motivation for screening (0.355, $p < 0.01$), procedures (0.294, $p < 0.01$), and conditions for which screening is conducted (0.407, $p < 0.01$). In addition perceived choice, perceived risks and perceived benefits all show a significant (all $p < 0.01$) negative association with decisional uncertainty and decisional effectiveness. These suggest that as perceived choice, the perceived benefits and perceived lack of risks increase, so decisional uncertainty and perceived

decisional ineffectiveness decrease, as proposed in the structural model. These bivariate correlations are, therefore, not only of the proposed directionality but are also in keeping with the proposed structural models regarding the factors influencing parental decisional quality in relation to their acceptance of newborn bloodspot screening.

8.3 Model development

In line with best practice the models were evaluated in a two-stage process. Firstly the measurement model was evaluated in terms of how well the data fit the latent variables. Secondly the full structural model was evaluated to include the proposed relationships between latent constructs.

In the first stage each of the models are evaluated for their measurement component only. Each indicator, the summated scales previously generated, loads onto a single theoretical construct, or latent variable. In light of the amendments made during scale construction the confirmatory factor analysis models are shown below with a description of the loadings. In order to better evaluate the measurement model fit, the latent variable variances were fixed at one, allowing the factor loadings to be freely estimated.

Following the confirmatory factor analyses models 2 and model 3 are evaluated on both the measurement and structural aspects. In contrast to the CFA the variances of the latent variables were freely estimated and, in order to ensure the model was identified, the loading on the first indicator for each latent construct was constrained to one.

8.3.1 Model 1: Confirmatory Factor Analysis

Analysis of the measurement component of model 1 (not shown) reveals a borderline good fit according to the CFI (mean 0.938, standard deviation = 0.002) and RMSEA (mean 0.057, standard deviation = 0.001) although a χ^2 of 146.153 on 98 degrees of freedom is highly significant suggesting poor model fit ($p = 0.001$). Despite the apparently reasonable model fit, a review of the parameter estimates suggests that both the latent variables of INFSK and EXP poorly predict their associated indicator variables (all $p > 0.1$). This suggests that INFSK and EXP should be removed from the model. Modification of the model, through the removal of the non-significant variables, generates the proposed measurement component for model 2 and as such the analysis proceeds with the evaluation of this second model.

8.3.2 Model 2: Confirmatory Factor Analysis

The initial analysis of the model shown in Figure 8.2 revealed a negative error variance for the variable *Ben*. This value of -33.984 is not, however, significant ($p = 0.658$). Given the lack of negative error variances in model 1 it is not assumed to be caused by misspecification of the model but, rather, may be due to a combination of a relatively small sample together with only having two-indicators for the factor (Kline, 2005). As this is the only variable that is erroneous it is constrained to only allow error variances of zero or greater (Kline, 2005). Refitting the model with this constraint provides a good fit by the CFI (mean 0.981, standard deviation = 0.003) and RMSEA (mean 0.049, standard deviation = 0.004). All factor loadings are significant at the 5% level and so are retained.

Modification

No modifications are suggested and so the initial model is retained.

8.3.3 Model 2: Structural Equation Model

The initial model fitted is that shown in Figure 8.3. As with the measurement model, the fit indices suggested that model 2 was a reasonable fit to the data with a CFI of 0.950 (standard deviation of 0.003) and an RMSEA of 0.067 (standard deviation 0.002). These were slightly contradicted with a χ^2 value of 78.609 on 47 degrees of freedom, indicating a significant value at the 5% level ($p = 0.002616$). Reviewing the parameter estimates indicated that the majority of estimates are highly significant and should be retained. However, the path coefficient CHOICE $\rightarrow$ ATTSCR is not significant at the 5% level ($p = 0.312$) suggesting that this should be removed from the model.

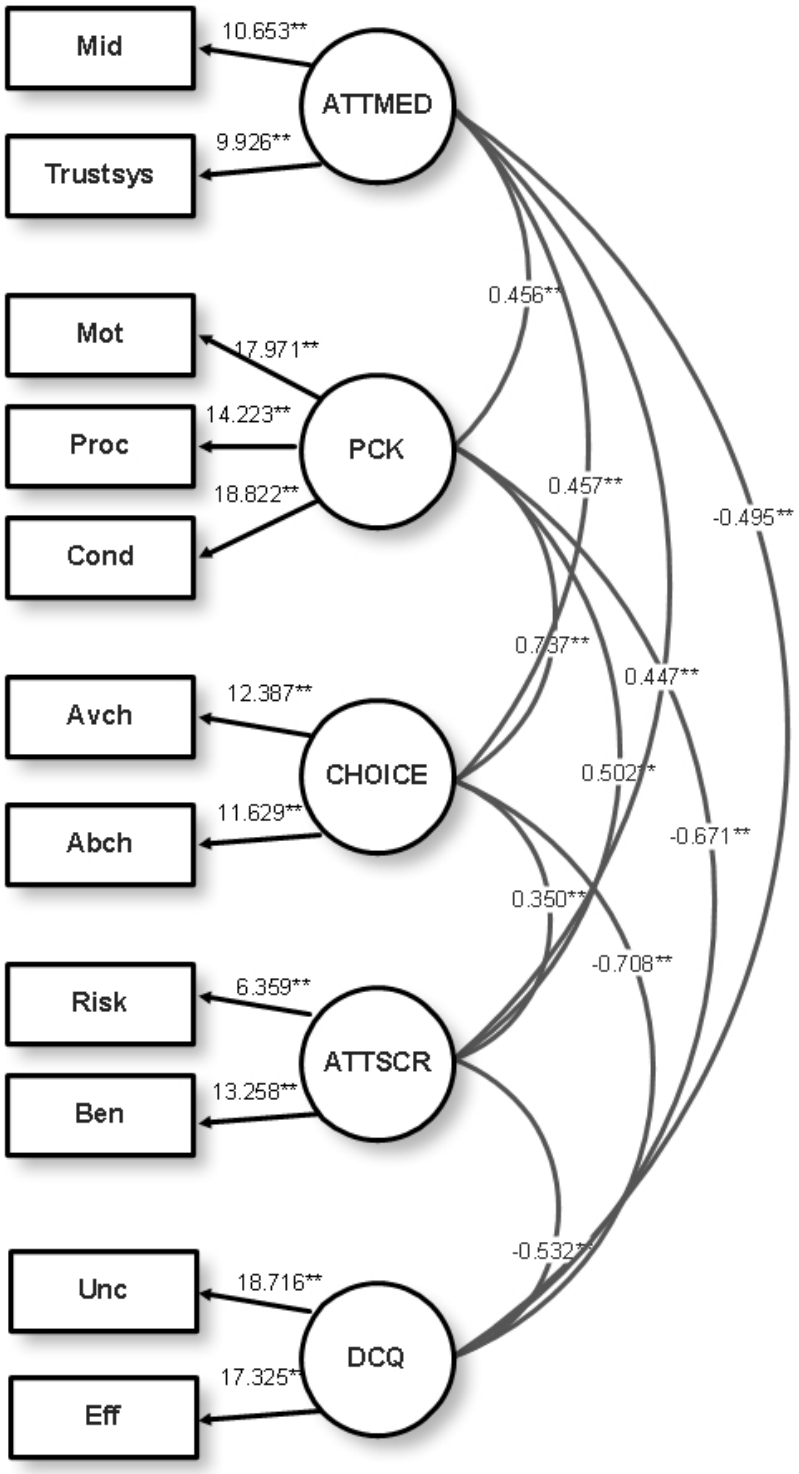

Figure 8.2: *CFA of model 2 showing unstandardised parameter estimates (errors not shown). NOTE: *$p < 0.05$, **$p < 0.01$, n=151*

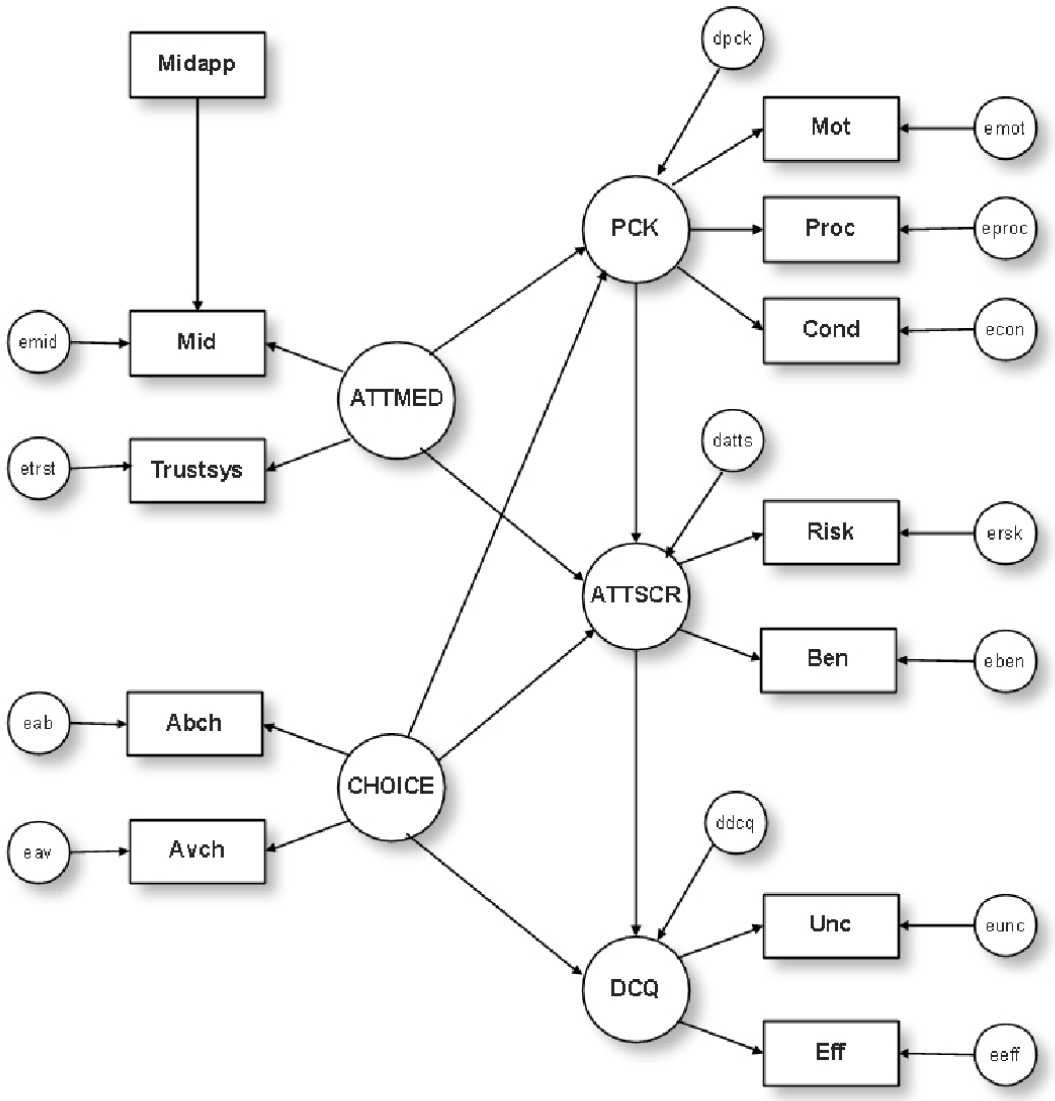

Figure 8.3: *Path diagram showing both the measurement and structural components of model 2*

Modification

In addition to the suggested removal of non-significant paths, the modification indices also suggest that the exogenous latent variables of CHOICE and ATTMED should be allowed to covary. Implementing this, together with the removal of the non-significant path improves the model. This once more generates a negative error variance for the variable *Ben* and so this is constrained to be zero or above and the model re-fitted. Fitting this iteration provides a further suggestion to remove the path ATTMED $\rightarrow$ PCK ($p = 0.488$).

The final model, together with parameter estimates, is shown in Figure 8.4. Variances, residual variances and R-squared values are also given in Table 8.3.

Variable	**Variances (S.E)**	**Residual variances (S.E)**	**R-squared (S.E)**
Midapp	3.585 (0.270)**		
ATTMED	103.162 (28.318)**		
CHOICE	120.769 (40.133)**		
Mot		60.666 (17.829)**	0.842 (0.047)**
Proc		109.983 (23.661)**	0.648 (0.076)**
Cond		285.118 (36.359)**	0.554 (0.063)**
Risk		165.463 (38.110)**	0.196 (0.073)**
Ben		0.002 (0.003)	1.000 (0.000)**
Abch		228.591 (36.301)**	0.346 (0.098)**
Avch		308.513 (43.076)**	0.293 (0.080)**
Unc		79.338 (24.670)**	0.817 (0.058)**
Eff		45.610 (16.142)**	0.868 (0.048)**
Mid		149.702 (31.809)**	0.416 (0.100)**
Trustsys		59.201 (19.938)**	0.632 (0.124)**
PCK		113.599 (27.323)**	0.649 (0.087)**
ATTSCR		27.968 (8.228)**	0.307 (0.071)**
DCQ		119.052 (33.271)**	0.664 (0.072)**

Table 8.3: *Unstandardised variances, residual variances and R-squared values for the final iteration of model 2 (*= $p < 0.05$, ** = $p < 0.01$; N = 151)*

The model clearly shows that an increase in perceived choice (CHOICE) has a negative effect on DCQ, thus lowering uncertainty and perceived lack of effectiveness. This is in line with the theoretical assumptions that improved choice leads to better decisions. The inverse being, of course, that those with a lower perceived

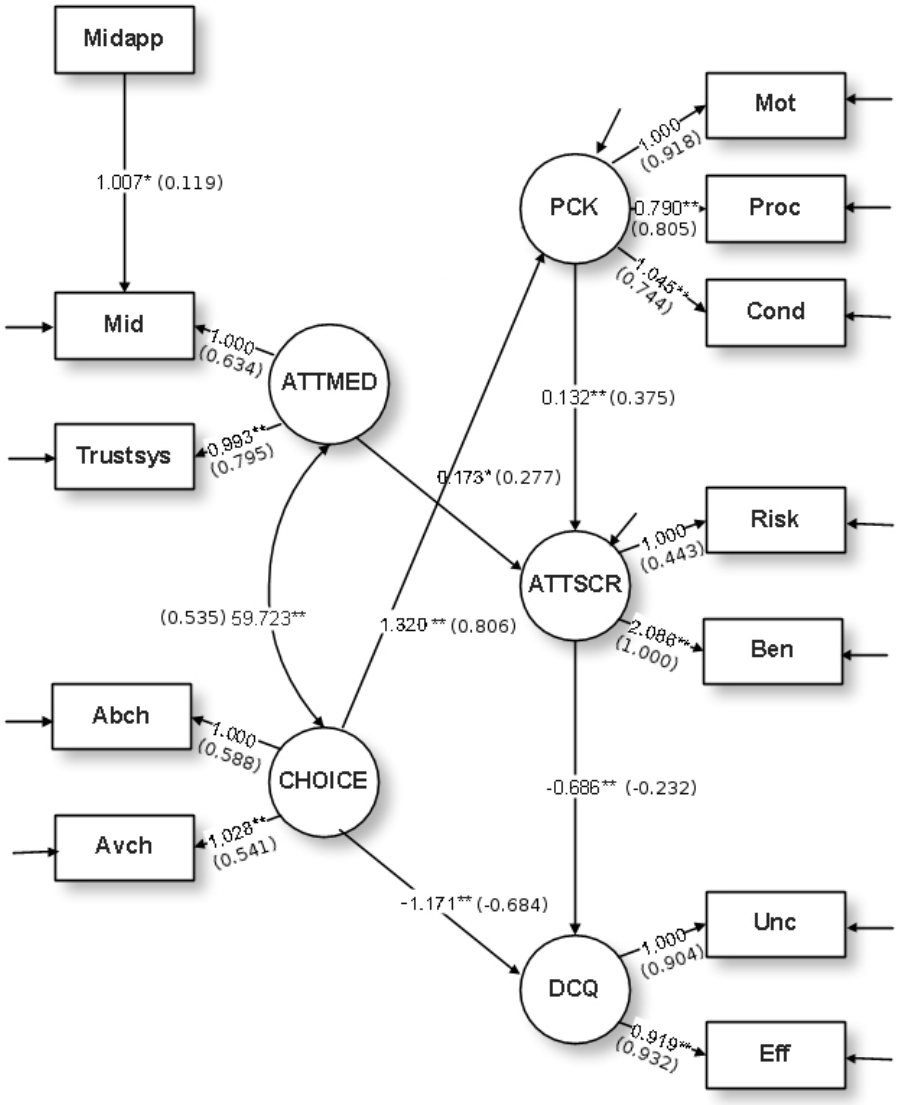

Figure 8.4: *Path diagram showing the final iteration of model 2 with unstandardised parameter estimates. Standardised parameter estimates are in parentheses. NOTE: *= $p < 0.05$, **= $p < 0.01$, n=151 (variable errors and disturbances not shown).*

choice will tend to have a lower degree of perceived decisional quality. As already suggested, this may mean that not only are these parents more likely to make more uncertain decisions, but any impediment to their ability to make a choice or the availability of a choice, may be indicative of a lack of informed consent.

In addition to the influence of perceived choice, increases in attitudes towards screening also lead to improved decisions on the basis of a negative unstandardised path coefficient of -0.686. Again this is as expected, with stronger positive attitudes being associated with increased decisional quality. The strong negative correlation (-0.459) between perceived benefits and decisional uncertainty suggesting potentially that the perceived benefits play an important role. Stronger positive attitudes may also indicate a stronger degree of certainty, particularly in relation to benefits. Again, this is in keeping with the interview data where parents talked about the obvious benefits of screening and the lack of harms which made the decision a relatively easy one.

The model indicates that the role of specific attitudes towards screening is secondary to perceived choice; choice has a greater effect on decisional quality. This is clearly shown with CHOICE having a standardised regression coefficient that is almost three times that of the path between ATTSCR and DCQ. The size of the coefficient points to this effect not only being of statistical significance but substantive significance as well. Accordingly any explanation of parental decisional quality needs to consider not only the rational evaluation of direct risks and benefits of the immediate test but also wider aspects of the context in which the test is offered. The substantive contribution to the explanation of parental decisional quality is confirmed with the squared multiple correlations which suggests that around 66% of the variance in DCQ is explained by the two independent latent constructs of CHOICE and ATTSCR.

Parental knowledge has been the subject of much research in newborn screening. Whilst the confirmatory factor analysis and correlations suggest the relationship between information-seeking, experience, and knowledge is a complex one requiring greater study, the structural model suggests that knowledge has a significant association with attitudes towards screening. The latent construct of perceived knowledge not only explains the variation within the indicator variables to a reasonable degree (over 84% of the variation in knowledge of the motivation for screening), but also shows a significant and positive relationship with attitudes towards screening, with increases in positive attitudes being causally attributable, in part, to increases in knowledge. This finding of a statistically significant relationship is also suggested to have a substantive role, with the standardised regression coefficient falling within the typical or medium effect size range. The concept of 'treatability' may be a significant factor in this association. Those parents who talked of treatability during the interviews not only showed a knowledge or understanding of the condition in order to relate to the fact that it was treatable, but they also understood that screening was made available and that testing at this stage allowed for the greater effectiveness of the treatment. It is in this way that increased knowledge may contribute to the increases in perceived benefits and positive attitudes towards screening.

Perceived knowledge is also found to be significantly related to perceived choice, with increases in perceived choice leading to increases in perceived knowledge. With a standardised regression coefficient of 0.806 this is of both statistical and practical significance. This again is in keeping with the model proposal that a perceived choice instigates, perhaps through discussion with the midwife, a greater awareness of the options and information uptake leading to a greater perceived knowledge. Equally, a lack of perceived choice or a perception of screening as mandatory, may be argued to suppress information-seeking or knowledge, potentially through a perceived lack of need.

Attitudes to screening were also significantly influenced by more general attitudes of medicine, although standardised regression coefficients suggested that the substantive effect is more limited than the effect of perceived knowledge. These indicators, which probed notions of trust, again show that the process of parental decision-making is not purely based on the rational evaluation of risks and benefits of the immediate test but also wider implications. The model indicates that in general those parents with more positive attitudes towards healthcare staff and the healthcare system show more positive attitudes towards screening. The R-squared value of 0.307 for ATTSCR suggests that around 70% of the variance remains unexplained suggesting that knowledge and more general attitudes only play a minor role in explaining attitudes towards screening. Indeed, whilst The R-square values suggest that for many of the variables much of the variation is explained, the perceived risks and benefits of screening are poorly explained by the underlying factor of attitudes to screening, again re-emphasising the need for further research in this area.

This final iteration of the model has an AIC of 14226.278. This is a decrease of 18.657 over the original conception, indicating an improvement in the model. With parameter estimates that are consistent with the model development theory and excellent fit on all indices (χ^2 of 61.396 on 48 degrees of freedom ($p = 0.093$), CFI of 0.979 (standard deviation 0.003) and an RMSEA of 0.043 (standard deviation 0.003)) the model appears to provide an adequate explanation of the factors influencing the perceived quality of parental decisions to accept newborn bloodspot screening.

8.3.4 Model 3: Confirmatory Factor Analysis

The initial iteration of model 3 is similar to that of model 2 except that the variables of *Risk, Ben, Mid* and *Trustsys* all load onto a single factor. This measurement model has a borderline acceptable fit by both the CFI (mean 0.941, standard deviation = 0.003) and RMSEA (mean 0.080, standard deviation = 0.002). The model also has a AIC of 13632.263 (standard deviation = 14.034) suggesting a slightly worse fit than model 2. All factor loadings are significant at the 5% level and so are retained.

Modification

The analysis for model 3 reveals suggestions for model improvement. The first suggested modification is to allow the errors for *Mid* and *Trustsys* to covary. Allowing the errors to covary indicates that some of the unexplained variance is not random measurement error, but is meaningful yet not explained by the latent construct defined within the model. In the same way it is also suggested that one should allow the error covariance between *Risk* and *Ben* to be freely estimated. Again the data suggests a substantive reason for this with highly skewed responses to questions relating to risks and the reasonable assumption that low perceptions of risk will in turn be indicative of high perceptions of benefit. Consequently we allow the errors of *Risk* and *Ben* and *Mid* and *Trustsys* to covary.

Implementing these changes and refitting the model provides the final model shown in Figure 8.5. A CFI of 0.981 (standard deviation 0.002), an RMSEA of 0.047 (standard deviation 0.003) together with an AIC of 13605.172 suggests not only an improvement in the model, but also a marginal improvement over model 2. All factor loadings are significant and thus retained.

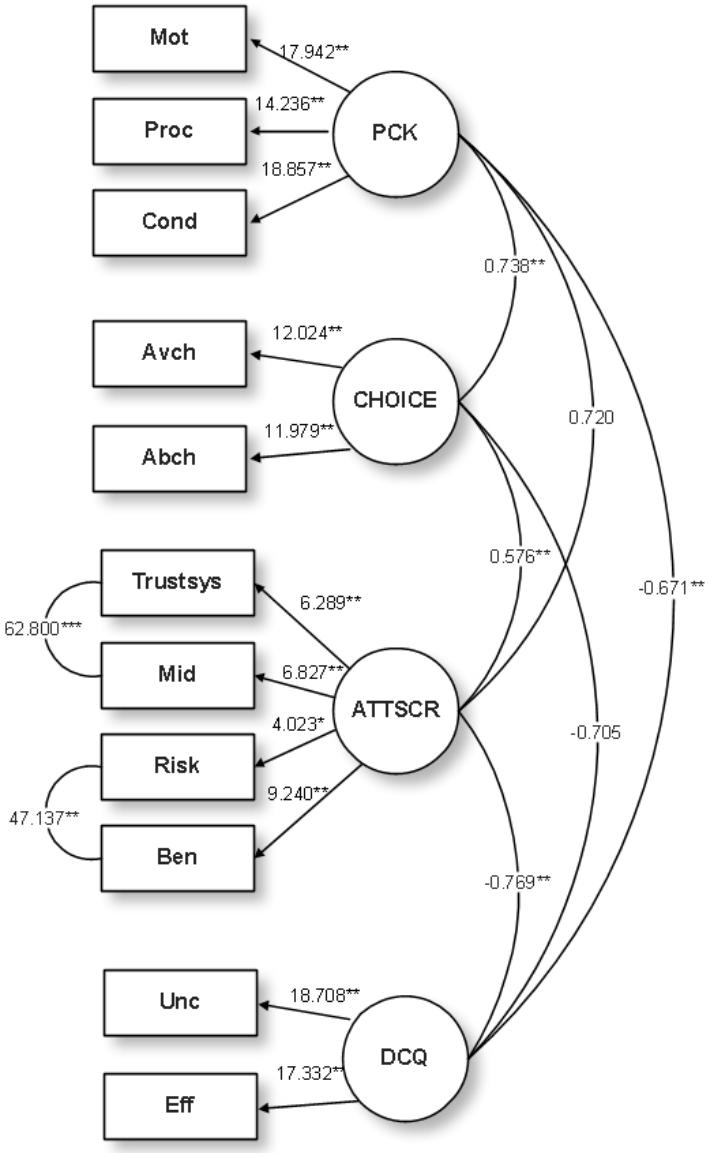

Figure 8.5: *CFA of model 3 showing unstandardised parameter estimates (errors not shown). NOTE:* $*p < 0.05$, $**p < 0.01$, *n=151*

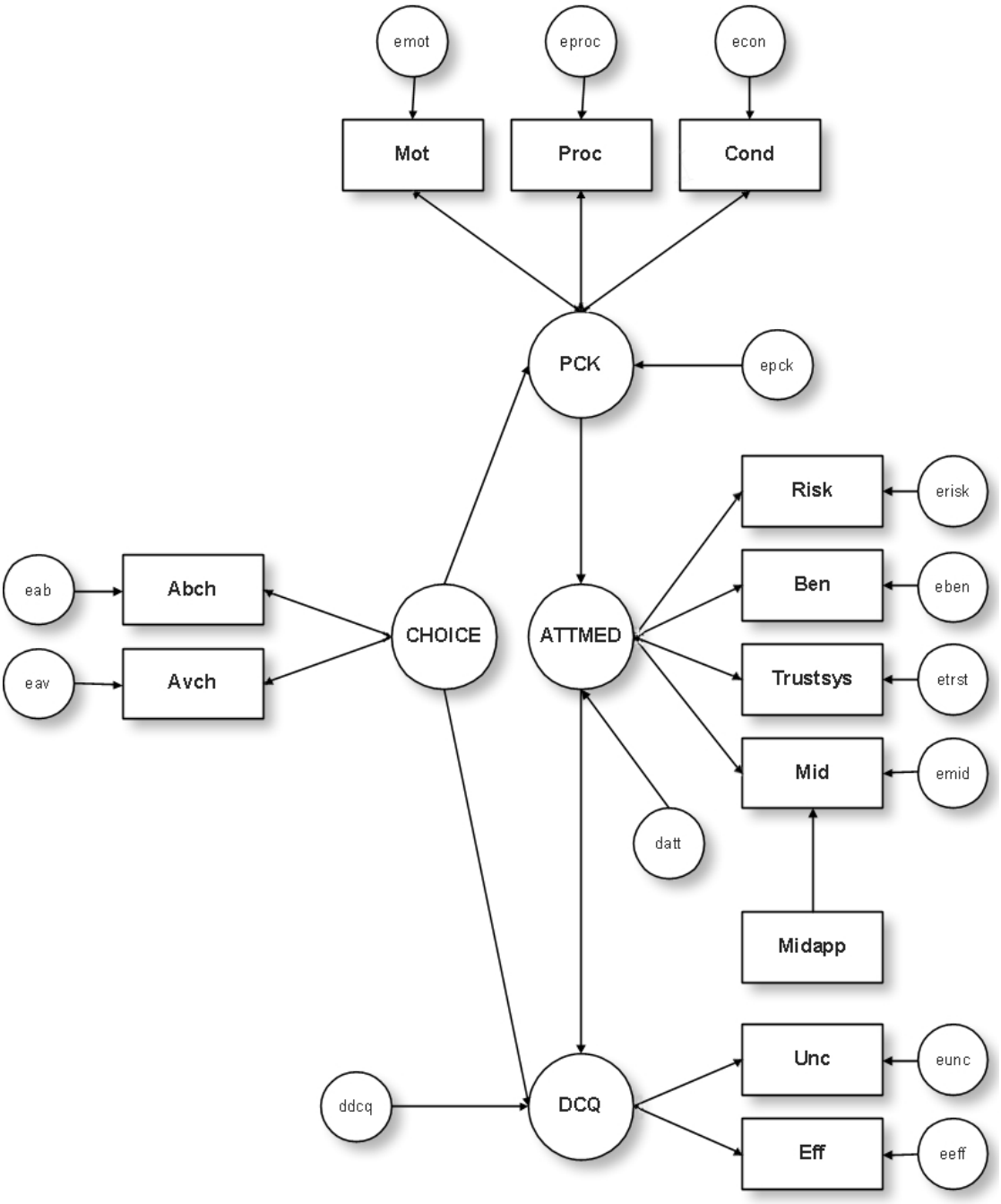

Figure 8.6: *Path diagram showing both the measurement and structural components of model 3*

8.3.5 Model 3: Structural Equation Model

Fitting the third model (Figure 8.6), and including the error covariances suggested from the confirmatory factor analysis, provides a good level of fit with a CFI of 0.977 (standard deviation 0.002), an RMSEA of 0.044 (standard deviation of 0.002) and a χ^2 of 62.372 on 48 degrees of freedom ($p = 0.080$). The model also has an AIC of 14225.317 (standard deviation 13.873) which is comparable to model 2.

Modification

Whilst the model is a good fit, review of the modification indices suggest that an improvement can be made by including the path Midapp $\rightarrow$ CHOICE. This is substantively and logically reasonable, given the official instruction that midwives should discuss newborn screening with parents. It, therefore, seems reasonable that those parents who have seen the midwife more often are more likely to have discussed newborn bloodspot screening and consequently the need for informed choice. These parents would, by this logic, be expected to have a greater perception of choice, particularly regarding the availability of choice if not necessarily the ability to make a choice. The final model which includes this additional causal link is shown in Figure 8.7. Variances, residual variances and R-squared values are also given in Table 8.4.

Direction of the relationships between the latent constructs are found to be the same in model 3 as they were in model 2. Both attitudes to medicine (ATTMED) and perceived choice (CHOICE) appear to have a significant effect on perceived decisional quality (DCQ). Perceived choice retains the positive causal effect on perceived knowledge (PCK). Whilst this remains a large and substantively significant effect, the size of effect is reduced in comparison with model 2 (0.699 compared with 0.806). In contrast to the previous model, attitudes to medicine appear to be having a greater effect on perceived decisional quality than perceived choice. Both coefficients do, however, remain significant at the 5% level. Perceived knowledge

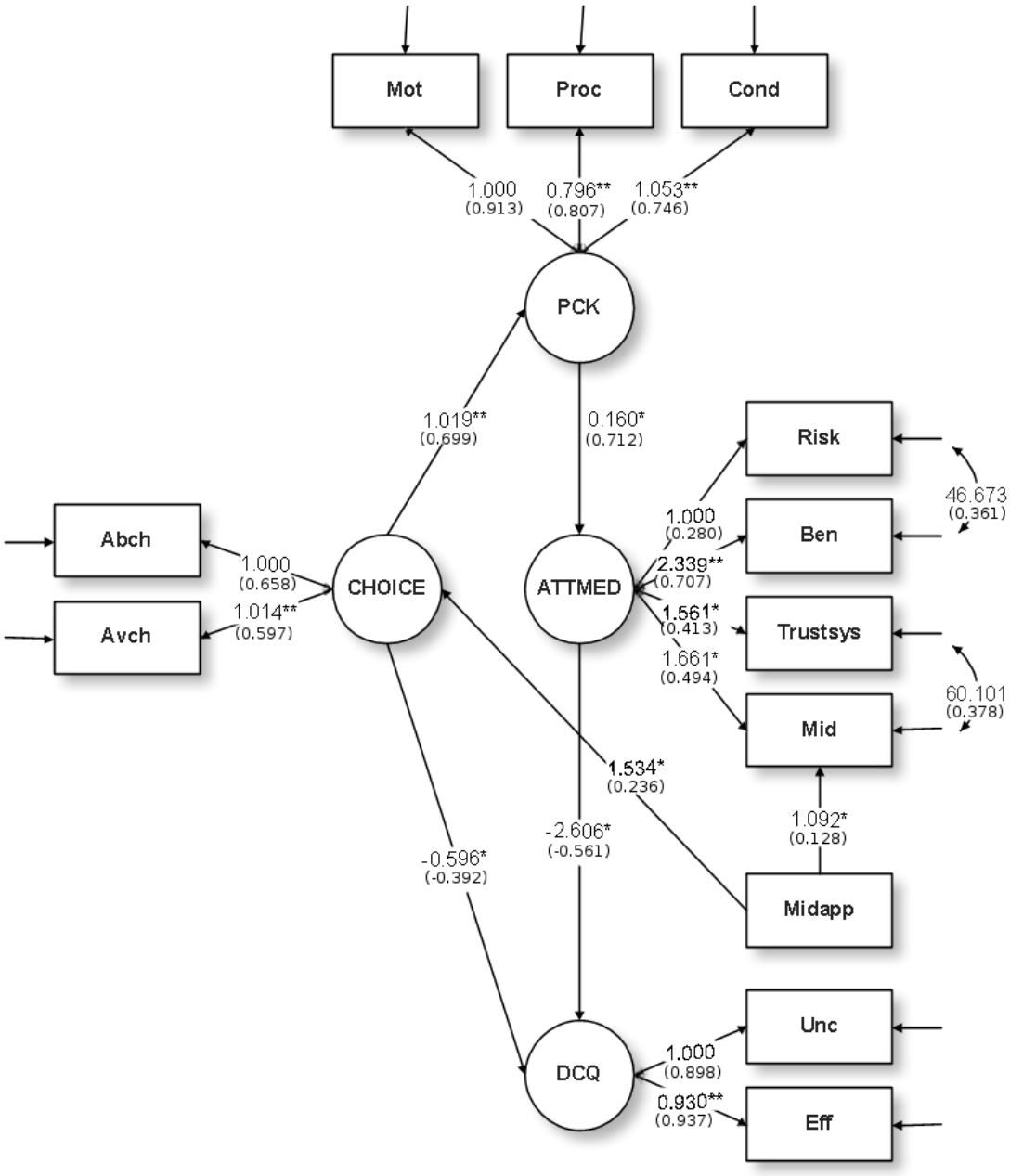

Figure 8.7: *Path diagram showing the final iteration of model 3 with unstandardised parameter estimates. Standardised parameter estimates are in parentheses. NOTE: *= $p < 0.05$, **= $p < 0.01$, n=151 (variable errors and disturbances not shown).*

Variable	**Variances (S.E)**	**Residual variances (S.E)**	**R-squared**
Midapp	3.585 (0.272)**		
Mot		63.724 (17.708)**	0.834 (0.047)**
Proc		108.917 (23.450)**	0.651 (0.073)**
Cond		283.235 (36.528)**	0.557 (0.062)**
Risk		189.792 (34.158)**	0.078 (0.061)
Ben		87.834 (15.598)**	0.500 (0.090)**
Abch		198.130 (39.420)**	0.433 (0.116)**
Avch		280.631 (44.996)**	0.357 (0.089)**
Unc		83.357 (25.618)**	0.807 (0.061)**
Eff		42.174 (16.612)*	0.877 (0.050)**
Mid		208.070 (33.162)**	0.200 (0.076)**
Trustsys		121.733 (15.730)**	0.244 (0.078)**
PCK		163.897 (33.609)**	0.489 (0.100)**
ATTMED		7.926 (6.592)	0.508 (0.103)**
DCQ		109.108 (33.264)**	0.687 (0.075)**
CHOICE		142.789 (47.534)*	0.056 (0.045)

Table 8.4: *Unstandardised variances, residual variances and R-squared values for the final iteration of model 3 (*= $p < 0.05$, ** = $p < 0.01$; N = 151))*

itself has a significant and positive effect on the newly combined construct of attitudes to medicine. This again retains its statistical significance but now presents a much greater substantive effect, indicated by an increased standardised regression coefficient when compared to model 2 (0.712 and 0.375 respectively).

The effect of perceived choice on perceived decisional quality is again consistent with the proposed directionality, with increases in perceived choice leading to a perceived improvement in decisional quality. This effect is less significant, both substantively and statistically, than in model 2 with a standardised regression coefficient of -0.392. Further to previous models, model 3 also provides evidence for a positive relationship between the frequency with which the parents saw the same midwife and perceived choice, although the effect size is toward the lower end. This once more points to the importance of the relationship between the midwife and the parent in decision-making for newborn bloodspot screening. The level of explanation in terms of variance in perceived choice is low, with only around 6% of the variance in perceived choice being explained by the frequency of midwife appointments. More work is, therefore, required in terms of identifying the factors

which influence perceived choice.

The final iteration of the model has an AIC of 14221.416, reduced from 14225.317. This is a decrease of 3.901 indicating only a marginal improvement over the original formulation. With parameter estimates that are consistent with the model development theory and excellent fit on all indices (χ^2 of 56.801 on 47 degrees of freedom ($p = 0.155$), CFI of 0.985 (standard deviation 0.002) and an RMSEA of 0.037 (standard deviation of 0.003)) the model appears to provide a good explanation, through the application of fit indices alone, of the factors influencing the perceived quality of parental decisions to accept newborn bloodspot screening. Despite the good model fit, this final iteration of the model raises questions regarding the substantive application of the model. In particular the low R-squared values for *Risk* and CHOICE suggests that the explanatory power, in terms of substantive explanation, is limited when compared to model 2.

As already noted, attitudes towards medicine, in contrast to the previous model, appear to be having a greater influence on perceived decisional quality than does perceived choice. This in part may be due to the combining of the previously separate latent constructs of attitudes to medicine (ATTMED) and attitudes to screening (ATTSCR). It will be remembered that in model 2 ATTMED had a significant and positive effect on ATTSCR and so the increase in effect seen here is not necessarily surprising. Despite the large effects and the good fit of the CFA model, the R-squared values suggest that the model may be substantively weaker than model 2, with the single latent construct of ATTMED explaining much less of the variance in the indicator variables than model 2. The variable *Risk* has R-square value of 0.078, indicating that only around 8% of the variance in the variable is explained by the newly combined latent construct of ATTMED. This is now non-significant at the 5% level ($p = 0.2$). Compare this to model 2 where almost 20% of the variance was explained by the underlying construct of ATTSCR.

The R-squared values of *Mid* and *Trustsys* are also reduced in model 3. When coupled to the required error covariances between *Risk* and *Ben* and *Mid* and *Trustsys* this finding may be seen to suggest that the application of the model with a single latent construct of ATTMED is substantively questionable.

8.3.6 Summary

From the three initial models two competing models were found to be adequate in terms of their measurement components. Applying these measurement aspects to the proposed structural relationships derived from the interviews provided the full structural equation models and allowed for a complete assessment of the proposed causal relationships. In both models 2 and 3 minor modification, through the removal of non-significant paths and the inclusion of additional paths, generated two models that demonstrated adequate explanation by a number of fit indices. However, a review of the parameter estimates and R-squared values indicated that model 2 was substantively preferable. The application of this model provides a number of novel insights.

The lack of correlations between indicators of experience or information-seeking with either knowledge or attitudes to medicine suggest that the role played by information-seeking and experience appears not to be one affecting parental satisfaction with decisions, or potentially parental decision-making at all. This is further compounded by the lack of significant association between the latent constructs within the confirmatory factor analysis of model 1. This is of particular interest given the central role of information provision and education within informed consent procedures generally, and may point to other processes through which parents make decisions relating to newborn bloodspot screening specifically. Two particular factors are indicated by the models assessed here.

Firstly, more general attitudes towards healthcare staff and the healthcare sys-

tem appear to play a strong role in determining attitudes towards screening and in turn parental perceptions of the quality of decision made. This is indicated through significant path coefficients between the latent constructs of attitudes to medicine (ATTMED) and specific attitudes to screening (ATTSCR) and the subsequent significant negative path coefficients between these specific attitudes and perceived decisional quality (DCQ). These suggest that positive attitudes towards the components of the healthcare system more generally lead to more positive attitudes towards screening in terms of perceived risks and benefits. More positive attitudes towards screening in turn lead to greater perceived quality of decision. This is perhaps to be expected given that all parents who completed the questionnaire also accepted screening. Further work would be of benefit to see if the relationship holds in the opposite case so that stronger *negative* attitudes are associated with a greater perceived quality of decision for those parents who *declined* screening.

The second significant factor identified is that of perceived choice. Perceived choice appears to affect the perceived quality of the decision directly as well as indirectly through a positive effect on knowledge which in turn has a positive effect on attitudes towards screening. This, as already described, was found to have a significant direct effect on perceived quality of decision. Whilst perceived choice has previously been related to informed consent or parental decisions, this study quantifies this role and suggests that the effect may be even greater than specific attitudes towards the screening test.

Whilst both models 2 and 3 demonstrated good fit, the further evaluation of both parameter estimates and R-square values suggests that model 2 is to be preferred. This is for several reasons. Firstly, the fit of model 3 was based on the inclusion of covariances between error terms for several observed variables. Whilst substantively reasonable, the requirement to allow error terms to covary is indicative of

some underlying model misspecification or at least lack of model fit. This is further suggested by the extremely low R-square values for *Risk*, and *CHOICE*, as well as decreases for both *Mid* and *Trustsys* in model 3. These low R-square values correspond to the error terms allowed to covary suggesting that the single factor of attitudes to medicine does not adequately capture the complexities of perceived risks and benefits when combined with more general attitudes towards healthcare, and that the two factor model employed in model 2 is preferable.

Whilst model 2 provides improved estimates of R-square for all variables, and does not require the correlation of error terms, there is still improvement to be made in terms of the amount of variance explained for the variables of *Risk*. Whilst this may in part be explained by highly skewed and polarised views regarding the risks and benefits of screening it may also be an area that would benefit from further research in terms of the influences on perceptions of risks and benefits with newborn screening.

Despite these findings several caveats must be made. When compared to the population in Merseyside and Cheshire, parents appeared to be older and more educated. This may present a bias in the results, although no significant differences were found when scores on knowledge indicators were compared by income or education. The sample showed marginal differences from the regional demographics in terms of ethnicity. It is likely that the relatively small sample size contributed to these differences in ethnic demographics. The response rate of 32% is also relatively low, indicating a potential bias. However,a lack of information with which to compare responders to non-responders means that a response bias cannot be confirmed or denied. Whilst the response rate was relatively low, it does compare well with other survey research relating to parental experiences of newborn screening which have seen response rates as low as 23% (Mischler et al., 1998), 28.6% (Ciske et al., 2001), and 32.5% (Davey et al., 2005).

Despite these cautions, the data does provide significant insights regarding parental decision-making, in particular the distinction between general attitudes and specific attitudes. The role of trust appears to be significant and through the disaggregation of the general and specific attitudes, the significant role of perceived choice is shown. Both of these elements suggest that the context and presentation of screening are key determinants of parental decisional-quality.

Chapter 9

Decision-making in the context of newborn bloodspot screening

This study took a mixed methods approach to exploring parental perceptions of decision-making within the context of newborn bloodspot screening, with initial exploratory interviews informing a larger scale quantitative survey. This process may be considered a process of triangulation, with the questionnaire data providing evidence as to the pervasiveness of attitudes or experiences indicated within the interviews. The interviews may also be considered to provide additional explanation of the trends seen within the questionnaire. Consequently, whilst the research design and protocol present the data collection and analysis in a linear fashion, the actualities of the analysis and interpretation is much more of an iterative cycle.

One limitation that may be levelled at the study is the relatively low response rate. It is unclear why there was a lack of response, although one may hypothesise that parents felt unable to commit the time to an interview or to complete a questionnaire, particularly for research that would provide no discernable health benefit for their child. This level of response is consistent with a number of previous studies on parental attitudes to newborn screening (Mischler et al., 1998; Ciske et al., 2001; Davey et al., 2005) and may explain the heterogeneous samples

noted in other studies (Dankert-Roelse et al., 1990; Dudding et al., 2000; Firth et al., 1983; Muchamore et al., 2006b; Hildes et al., 1993; Merelle et al., 2003). The responses to the postal questionnaire suggest that respondents may have a higher level of income than the general population and this should be borne in mind when interpreting the results. The lack of respondents who *declined* consent to newborn bloodspot screening does mean that the conclusions drawn cannot be generalised to those who do not consent. Whilst a deficit to the study, the small number of parents who did refuse - in the region of 0.025% for the period from which the sample was drawn - means that even with a high response rate there would not be enough respondents to conduct any reliable statistical analyses. As a result information relating to the decision-making of parents who decline newborn bloodspot screening remains an area in need of further research.

Despite these limitations the data from both phases clearly shows the importance of context as well as content in parental decision-making about newborn screening. Parents talked of their experiences in terms of their perceived ability to make an informed decision but also what they considered to be the availability of choice; that is the possibility of actually being allowed to make a decision rather than being routinely processed along a pre-defined trajectory. Equally the analysis through the structural equation modelling indicated a significant deterministic relationship between parental perceived choice and both their attitudes towards screening and perceptions regarding the quality of the decision made. Perceived choice was also positively associated with improved decisional quality indicated through increases in perceived effectiveness of decision and reduced decision uncertainty.

Trust was also found to be important. Parents talked of trust not only in the midwife, but also in the health service. Attitudes towards screening could be shown to be causally attributable, at least in part, to trust in these same staff and systems.

Surprisingly, there was a lack of evidence to support the proposed relationships between information-seeking and knowledge or experience and knowledge. Discussions with parents suggested that trust is again significant in terms of limiting the information-seeking process to a few trusted sources, particularly as screening occurs at a time when parents are principally occupied with the care of their baby.

In the remainder of this chapter I place these findings into the wider health context and discuss the new insights the study brings with it.

9.1 Choice and decision quality

The results of the structural equation modelling analysis presented evidence to support the modelled causal relationship between perceived choice and parental perceptions of decisional quality. This indicated that with increasing perceptions of choice there was a significant improvement in the perceived quality of the decision to accept newborn bloodspot screening. The effect of perceived choice on perceived decisional quality was found to be greater than the effect of specific attitudes towards the screening, with a path coefficient three times as large.

Perceived choice, and its relationship with decisional quality, is an area that is sorely lacking within the newborn screening literature. Those studies that have considered choice generally do so within the limited context of parental attitudes to mandatory or voluntary screening (Faden et al., 1982; Campbell and Ross, 2005; Davis et al., 2006; Detmar et al., 2007). This body of work presents highly variable results with some studies indicating that parents feel that screening should be mandatory (Faden et al., 1982), whilst others suggest that parents strongly support voluntary screening (Muchamore et al., 2006a; Detmar et al., 2007). What is generally missing is any analysis that considers the role that choice - or at least perceived choice - may play with regard to decision-making.

Despite this, research does exist which support that the current findings. Research by Parsons et al. (2005), which sought to evaluate an intervention which emphasised an optional screen for Duchenne muscular dystrophy, found that parents who received the intervention were more likely to feel that the midwife had given them a choice and were significantly more satisfied. Consequently the presentation of the information, and choice, appears to play a crucial role in parental experiences of decision-making for newborn bloodspot screening.

9.1.1 Perceptions of choice

The impact on perceived choice appears to largely rest with the perceptions relating to the availability of choice as opposed to parental concerns regarding their ability to make a choice. The data presented in this study shows that experiences in relation to choice are highly variable and on average parents perceived a low availability of choice. The highly variable responses covered the full spectrum of possibilities, with some parents rating the availability of choice (Avch) as zero whilst others scored 100 indicating that they felt that they had made an independent and free choice. The average scores were also low, below 50, indicating that on average parents viewed the availability of choice as distinctly limited. Over 80% of parents responding to the questionnaire felt that it was expected that their child had the heelprick and 40% disagreed to some extent that the heel prick was presented as an optional test. Perhaps more worryingly, over 30% of respondents felt that they did not have a choice to decline the test. When compared to the results of Parsons et al. (2005) these figures are considerably higher. Parsons et al. (2005) found that 15% of parents were not aware that testing was optional and 4% felt that they had not been given a choice. However, their study related to an additional screening for Duchenne muscular dystrophy that was explicitly noted as optional. The lower figures perhaps reflect this explicit nature of the offer.

The routine and implied choice

Almost all programmes of newborn bloodspot screening show high uptake rates (Bradley et al., 1993; Parsons et al., 2002; Campbell and Ross, 2003; Metz et al., 2003; Dhondt, 2005). High uptake, it has been argued, may reflect a routinisation of screening and consequently may be indicative of a programme where informed consent may be lacking (Murray and Clarke, 2002). This may also be reflected in the low levels of perceived choice within the current study. Whilst the context here was that of newborn bloodspot screening, the lack of perceived choice has also been reported with parents undergoing prenatal screening, suggesting that the issues at work may be more general and relating to the presentation of the screening as opposed to the specific details within each situation. In one study of parental experience of Duchenne muscular dystrophy screening (Parsons et al., 2005) the authors concluded that:

> "It would seem that, although the majority of mothers felt they were being given a choice about screening, for some it was a choice about a test they thought was always done. This, in reality, is no choice at all." (Parsons et al., 2005, p168)

Whilst the authors concluded that these parents didn't experience a choice, the causes of this lack of choice weren't explored. The present study, and in particular the interviews, supports the implication that the way in which screening is delivered is a factor in this lack of perceived choice; something which has been raised in the context of prenatal testing (Clarke, 1991). In particular the 'proceduralisation' of the process was noted within the interviews. This is consistent with ethnographical work conducted within the antenatal booking visit in which McCourt (2006) notes that whilst choice is affirmed the language used emphasises the almost inevitability of testing. As she notes:

> "Two main rhetorical patterns were identified in the use of language relating to choices. We have termed these 'routine as choice' and 'choice

> as routine'. Routine as choice implies that what is routine is the normal, common and therefore right choice to make. This commonly included terms in describing the visit such as 'you will have' or 'and then you have', or the language of a product for consumption such as 'we do', 'we offer' or 'everyone has'. Choice as routine implies more openness, but still presents certain choices as the routine ones, which most people are likely to make. It also commonly uses consumerist terms, for example, 'what we offer here', 'you can have' or 'it's your choice'. The language around choice in the majority of visits appeared to promote what Kirkham and colleagues, in their study of informed choice leaflets in maternity care, termed 'informed compliance' (Stapleton, Kirkham, & Thomas, 2002) since it was difficult for women (and could have appeared disruptive) to interject in the flow or to challenge it." (McCourt, 2006, p1315)

The same use of rhetoric was found in the present study when parents talked about how they were told that the midwife 'needed to come and do a couple of routine tests'. The latter rhetoric of routine as choice is sometimes emphasised through what Pilnick (2008) refers to as the tests' 'sequential location'; the way it is closely related in proximity to other more routine tests or maternal checks. Again this was identified within the interviews with parents talking about how the screening was just one of a range of tests.

Indeed some authors have argued that the mere existence of screening may in itself be seen by parents as a reason to assume that this screening is worthwhile (Clarke, 1991; Pilnick, 2002; Nijsingh, 2007). This implicit endorsement has been observed within prenatal screening (Potter et al., 2008) and appeared to be the case here. For some mothers the perception of the screening as routine was seen to verify the acceptability of it. This was not an irrational acceptance. For example, one mother discussed the financial constraints within which the NHS operates and this

was used to suggest that the fact that screening was offered meant that it was both worthwhile and efficient. This approach falls between the two principal processes of decision-making in that it is not a systematic processing of the immediate information about screening yet is not as simplistic as employing a heuristic such as 'experts can be trusted' (Marteau and Anionwu, 1999). That parents accept screening on the basis that it is offered by the healthcare system is consistent with what Porter and Macintyre (1984) refer to as 'what is, must be best'. This acceptance of routines because they are routine (Green and Statham, 1996) suggests that parents are not necessarily making an informed choice on the basis of the immediate information about the screening tests. This assertion finds support through the lack of evidence for a relationship between information-seeking and perceived decisional quality.

9.2 Choice and knowledge of screening

As discussed previously, knowledge and understanding are important aspects to informed consent. If parents do not understand the information provided then this can be seen to vitiate their consent. However, the framing of the information is key; information presented in a manner that is not conducive to decision-making is unlikely to be used or retained by the individual. In the present study perception of choice was strongly associated with knowledge. This evidence supports the notion of a positive causal relationship through which increases in perceived choice lead to increases in perceived knowledge. This is again found within the screening literature. Parsons et al. (2005) found that mothers who received an intervention to emphasise parental choice had a greater knowledge about the process of the optional screen for Duchenne Muscular Dystrophy. This confirmed earlier work in the US where it was found that a disclosure and consent process led to improved knowledge in mothers (Faden et al., 1982). This, it was argued in Chapter 8, may be attributable to greater discussion with the midwife or deliberation and awareness of screening brought about by an increase in perceived choice.

9.2.1 Parental knowledge

Parental knowledge is a common theme in newborn bloodspot screening research, with numerous studies suggesting that parents have a poor knowledge of newborn bloodspot screening (Smith et al., 1990; Tluczek et al., 1992; Zeuner et al., 1999; Tluczek et al., 2005; Lang et al., 2009). Despite these results, few studies have sought to disaggregate different aspects of knowledge about newborn bloodspot screening. In the present study, and in line with the results of the interviews, perceived knowledge of screening was divided into three broad domains: knowledge relating to the motivation for screening, knowledge of the procedures for conducting screening, and condition specific knowledge. Analysing parental responses showed higher average knowledge scores for the motivational and procedural aspects of screening when compared to condition specific knowledge.

The parental focus on practical aspects is in contrast to the majority of published research on recall which has tended to focus on technical details of conditions such as the ability to recall specific terms or condition prevalence (e.g. Warren et al., 1982; Holtzman et al., 1983; Lewis et al., 2006). The selection of the information to be assessed is integral to the responses one achieves and the subsequent inferences one can draw from the results. As Levitt argues:

> "If the aim in consulting the public is to show that more public education is needed then one method is to ask specific questions, keeping the focus in the researchers/funding bodies area of expertise" (Levitt, 2003, 20)

Consequently the selection of questions on which to assess knowledge is intrinsically tied to the results obtained. The results of the current study suggest that the selection of knowledge questions relating to motivational or procedural aspects of the programme are likely to yield results suggesting better knowledge than studies which focus on technical details or condition specific knowledge. However,

few studies disclose the process by which questions are selected or the reasoning behind their inclusion (Nicholls, 2010).

9.2.2 Knowledge saliency

The survey results are consistent with the interview findings where some parents lacked knowledge of condition names, yet knew why screening was conducted and understood the implications. Parental recall was consistent with stated interest. For parents who took part in the interviews it was the ability to act on information in order to benefit the health of their child which was important. The few studies which have considered different aspects of knowledge suggests that the present finding is not unique. Holtzman et al. (1983) found that a higher percentage of parents recalled how the screening was conducted than did the inherited nature of the disorders. Indeed the lowest levels of recollection was for a question asking about how often babies were born with the conditions. The authors also found that there was no significant difference between pre-disclosure and post-consent groups regarding this latter question. The parental responses within interviews for the present study suggest that this information is not as salient to parents as other aspects, and consequently not remembered. This observation has been referred to as "framing failures" in which it is noted that "the context of the individuals previous beliefs, assumptions, and experiences that frame the information provided" (Dawson, 2009, p104). Other studies have found that this pattern is replicated when parents are asked about information priorities. When parents requested more information this was about the nature and purpose of screening as opposed to technical details or aspects relating to prevalence (Campbell and Ross, 2004). More recent work has found that the saliency of the information may even extend to the recognition of conditions. One study found that 96% of mothers had heard of sickle cell disease, yet only 33% had heard of cystic fibrosis. The authors suggest that "This may be due in part to the high percentage (74%) of African American women in our survey. SCD is more common in per-

sons of African ancestry" (Lang et al., 2009, p2427). The suggestion being that as CF was less common, parents were less inclined to know about it. However, it remains unclear whether this difference is due to parents or to the practice of healthcare professionals, who may not present information they feel is not relevant.

The variation in areas of knowledge and interest is also found in other screening contexts. Studies in prenatal screening have found parents to have a greater knowledge of practical aspects of screening (Smith et al., 1994; Green et al., 2004). In particular, one study found that the meaning of a high risk result and the risk of miscarriage are important pieces of information, whilst the percentage of women who are likely to have a low risk result is seen to be unimportant (Michie et al., 2003). Again this differed by the immediate relevance. Unsurprisingly, information as to the percentage of women with a high risk result who went on to have a baby with Down's syndrome was more important to mothers who had a high risk result than to those who received a low risk result (Michie et al., 2003).

Similar knowledge patterns and information preferences are seen in cancer screening with Jepson et al. (2007) noting that parents wanted information on the risks and consequences of screening. In colorectal cancer screening, patients have been shown to have a greater knowledge of the procedures (colonoscopy) than the risk factors or symptoms (Koo et al., 2010), whilst a study of prostate cancer screening in African American men revealed a greater knowledge of the implications of screening than the function of the prostate or potential symptoms (Davis et al., 2010).

Not only does this have important implications for those assessing screening, but it is central to those providing information. Providing information that is not salient to parents will at worst fail to promote informed choices but may actually be detrimental. The consistency of findings across settings suggests that the areas

of parental concern are relatively well defined and discernable. By making these aspects central to the education given to parents it is not only likely that parental satisfaction with the information provided will be increased, but that this will also act to facilitate parental perceptions as to whether they have made an informed choice.

9.2.3 Knowledge through experience

Further to the lack of relationship between experience and information-seeking behaviour reported in the results of the structural equation modelling, there was also no evidence of any relationship between experience and knowledge. Parents were hypothesised to be more knowledgable if they were multiparous than if they were first time mothers. This supposition was made on the basis of interview data which suggested that parental understanding with their first child may be undermined by a lack of familiarity with healthcare systems. It seems that the converse is not to be found; parents with multiple children did not perceive themselves to be significantly more knowledgeable on any of the domains evaluated here.

Evidence regarding this within the newborn screening literature is minimal with only two studies identified which have considered the role of parity on knowledge. In a Welsh study Smith et al. (1990) found that parental awareness of screening was higher in multiparous compared to primaparous mothers. However, parental awareness of the screened for conditions did not differ with both groups showing a poor level of awareness. This result has been replicated in a further study which found that multiparous parents were no more knowledgeable of the screened for conditions than were primaparous parents (Statham et al., 1993).

This can be seen within the interview data, where parents recalled in greater detail their experiences with their first child. Decisions for subsequent children appeared less deliberated which may be why there was no association found between number

of children and knowledge. The interviews also suggest that the potential assumption of knowledge with multiparous mothers could lead to a plateau of knowledge or relatively small increase over those primaparous mothers. Furthermore, the present study did not include parents who had a screened for condition in their most recent child. The fact that these parents had not been confronted with a condition may be implicated in the failure to find an association between parity and knowledge. Time may also be a factor; for most parents childbirth will be a relatively rare event. It may be that a lack of impetus to remember details results in parents being no more knowledgeable in future pregnancies or after additional children than they were during their first.

9.2.4 Seeking information, gaining knowledge?

Parents discussed seeking information from a range of sources. Whilst sources did vary, both the interviews and questionnaires indicated that the midwife and health service dominated information provision. This is in keeping with previous research which has found that parents consistently cite the midwife as a key source of information (Tymstra, 1986; Hargreaves et al., 2005a; Davey et al., 2005; Tluczek et al., 2006; Parsons et al., 2006, 2007). Written information materials have been the focus of several studies (Hargreaves et al., 2004, 2005a), with the suggestion that in some cases these may not facilitate informed choice (Hargreaves et al., 2005b). Prior personal experience and the experiences of families and friends have also been found to be important (Davey et al., 2005). In a Welsh study one family's negative experience of screening for DMD was felt to have been the cause of a much higher refusal rate in one geographic area (Bradley et al., 1993). Despite these findings there is a paucity of evidence relating to information-seeking practices and how these impact on knowledge of newborn bloodspot screening.

The initial interview phase of the study had suggested that parents employed differing information-seeking practices which were broadly classed as "active seeking"

or "passive receiving". This grouping can be seen to show some similarities to a monitoring/blunting coping style (Miller, 1987). However, whilst those considered to be active seekers of information showed an information-seeking style akin to the traditional monitoring dimension, those parents labelled as passive receivers were inconsistent with the blunting aspects. Passive receivers did not provide demonstrable blunting techniques, such as information avoidance or distraction, and were found to accept information when this was provided to them. Instead, parental information seeking appeared to be consistent with the formulation of Wilson in which other modes of searching take place (Wilson, 1999; Case et al., 2005). Wilson's model identifies four methods of information search and acquisition:

> **"passive attention:** such as listening to the radio or watching television programmes, where information acquisition may take place without intentional seeking;
>
> **passive search:** signifies those occasions when one type of search (or other behaviour) results in the acquisition of information that happens to be relevant to the individual;
>
> **active search:** where an individual actively seeks out information; and
>
> **ongoing search:** where active searching has already established the basic framework of knowledge, ideas, beliefs or values, but where occasional continuing search is carried out to update or expand one's framework. In consumer research, Bloch et al. (1986) define ongoing search as that which is independent of specific purchase needs or decisions and that the motives are to build knowledge for future purchase decisions and simply to engage in a pleasurable activity." (Wilson, 1999, p562)

The parents within the interviews could, therefore, be considered to fulfil two of these categories with the active seekers equating to the active search within Wilsons

model, whilst the passive receivers can be considered to undertake a form of passive search, or what may also be referred to as "active scanning" which may involve practices such as semi-directed browsing and the active listening to conversations or questions in likely locations (McKenzie, 2002). Consequently, the attendance of antenatal appointments and the receipt of information from the midwife may be seen as a passive search occasioned by their attendance.

Questionnaire responses concurred with the different information-seeking practices with parents varying in terms of both their purported and actual information-seeking behaviour, indicated through the number of sources used. This variation is consistent with reported practices within newborn bloodspot screening internationally which has found that parents vary in terms of when, how and to what extent they collect or seek information (Tymstra, 1986; Parsons et al., 2006; Detmar et al., 2007; Tluczek et al., 2009). In particular, recent research has explicitly noted that "Some parents actively sought out information, for example, reading books, while others learned about NBS incidentally as part of their circumstances" (Tluczek et al., 2009, p329). Such a finding is consistent with the passive receiver or passive search strategies identified here.

Information-seeking behaviour scores indicated an active-seeking population with almost 40% of parents scoring highly in terms of stated behaviour. This wasn't borne out by actual behaviour, with the majority of parents (74%) using either one or two sources of information only and less than 10% using 4 or more sources. Cross-tabulations (not shown) revealed a non-significant relationship between stated information-seeking behaviour and the number of sources used. This may suggest that the differing information-seeking practices are not manifest in the number of sources used, but the way in which they are used.

Despite the identification of different information-seeking preferences, the present

study failed to find evidence of a relationship between information-seeking, assessed through either stated behaviour or the number of information sources used, and knowledge. Whilst there are no studies within the context of newborn bloodspot screening that consider such a relationship, work in prostate cancer screening has found that independent active seekers of information are significantly more knowledgeable about prostate cancer than doctor dependent seekers or passive recipients (Williams-Piehota et al., 2008). However, the analysis did not consider the information-seeking process *per se* as it did not clearly demarcate the active/passive or seeking/scanning distinctions.

The potential lack of association between both stated and actual information-seeking and knowledge is significant when one considers the education of parents. If the aim of the education is to improve parental knowledge of screening then giving parents more information, in the form of more materials from different sources, may not be effective. Instead resources may be better spent honing and refining a limited range of information sources. Furthermore, experience cannot be considered as knowledge. The finding that multiparous parents are not significantly more knowledgeable than primaparous parents demonstrates the need for consistent information irrespective of number of children. There is, however, a paucity of information relating to both information-seeking practices and the relationship with knowledge and it remains an area that is vastly under researched despite the extensive range of materials produced by the NHS and newborn screening programme. Specifically research is required which considers how the different information sources are used and the role that each of these play in the decision-making process. Whilst knowledge has been assessed in a variety of ways, the failure to consider the relationships with educational materials and information-seeking is to be lamented. More work is needed to consider what information materials are deemed accessible and useful and how these affect the information-seeking process. Each of these need to be related to knowledge so long as the consent given

by parents is required to be an informed one and not merely an assent.

9.3 Attitudes towards screening

Both the interviews and the questionnaire data indicate that trust is an important factor when making decisions to accept newborn bloodspot screening. Whilst both models 2 and 3 were supported by the quantitative data gathered through the questionnaire, model 2, in which general attitudes to medicine and specific attitudes to screening were disaggregated, was found to be theoretically more robust. Whilst attitudes are widely reported within both the newborn and prenatal screening literature, these attitudes tend to focus purely on the perceptions of risks and benefits of the immediate test (Al-Jader et al., 1990; Campbell and Ross, 2003, 2005; Davey et al., 2005). The findings here suggest that much more of the variability in perceived decisional quality may be accounted for if one also includes the wider attitudes towards healthcare. This finding further implicates the context of the screening in the responses which one achieves. Those parents who have more negative attitudes towards healthcare generally, are likely to have more negative attitudes towards screening specifically. The disaggregation of the general and specific attitudes may also in part explain the greater influence of choice on perceived decisional quality and may indicate that existing studies may over emphasise the role that attitudes play in parental decisions regarding screening.

Parents demonstrated the requirement for interpersonal trust, that is a trusting relationship between two individuals, and in particular singled out the relationship with the midwife. This was based on trust in her abilities and knowledge, demonstrated not only through objective sources of reference - such as qualifications - but also her demeanour. This finding is in keeping with previous studies on physician-patient trust which have found that "it was repeated experience of caring, compassion, and commitment that underpinned trust" (Guthrie, 2008; Brownlie, 2008). Parents also showed a more general trust in hospitals and the

NHS demonstrated by confidence in regulation and governance. The finding that both of these aspects loaded onto a single latent construct, and that this was a significant determinant of parental attitudes towards screening, is consistent with existing theories of trust relationships. As Hall et al. (2002) state:

> "General trust depends to some extent on patients' previous experiences with their own doctors. Also, patients who have greater general trust are expected to more readily trust individual physicians they meet for the first time. This is because, early in a treatment relationship, interpersonal trust is likely to be based primarily on general system features, but as the relationship continues, a divergence (either higher or lower) is more likely between general and interpersonal trust, as patients learn more about the particular characteristics of a provider." (Hall et al., 2002, p1422)

Trust, therefore, is not distinct between the healthcare professionals and the institutions in which they work. As Brownlie (2008, p21) suggests:

> "In reality, the relationships are two-way: health professionals are given a 'warrant for trust' (Misztal 1996, 121) from their association with a health profession or an organization such as a particular clinic; and, at the same time, trust in these institutions is 'built up', almost in a symbolic interactionist sense, through the recurrent actions between people within these settings."

Multiple-layers are in play with a reciprocal interaction between layers and confidence being established through individual interactions. Such a relationship was identified within the present study, suggesting that both the attitudes towards the key healthcare professionals - here the midwife - and the wider health service are important.

9.3.1 Established trust: parental relationships with the midwife

Trust in the midwife was a specific finding within the current study. This trusting relationship arose within the interviews and was confirmed through the quantitative analysis where it was found that there was a significant and positive relationship between the frequency with which parents saw the same midwife and the trust placed in them. This suggests that familiarity may be an important part of this trusting relationship.

The role of the parent-midwife relationship was noted by Parsons et al. (2007) and Muchamore et al. (2006b). In both studies trust in the midwife or healthcare professionals was an important factor in parental decision-making, even overriding the need for information in some parents (Parsons et al., 2007). The present study confirms this but also offers an insight into the causal determinants of this trust. The quantitative analysis indicates that increasing familiarity was associated with increased trust. This is potentially explained through a build up of trust over time. The discussions with parents suggests that characteristics of the midwife, such as honesty, confidentiality and not being patronised were important in establishing a trusting relationship.

This role of familiarity is a feature within trust literature, with authors claiming that an ongoing relationship not only engenders greater trust within an individual but may also mean that the individual may act in a more trustworthy manner (Solbjør, 2008). The qualitative data from the present study suggests that increasing contact with the same midwife, and the associated familiarity, led parents to perceive that they are given a greater deal of attention. Confidence in abstract systems of governance also plays a role. The parental interviews indicated that qualifications and experience together with the midwife's personal skills and demonstrable expertise were key to developing trust relations. This is consistent

with previous studies into trust relationships which have found that items relating to patient-centred care e.g. whether patients felt they were given enough attention, and perceptions of professional expertise are ranked highest in terms of explaining trust (Calnan and Rowe, 2006; Rowe and Calnan, 2006).

9.3.2 Institutional confidence

Whilst both interpersonal trust and institutional confidence were relatively high with mean scores of 69.7 and 65.4 respectively, confidence in healthcare institutions was lower than the interpersonal trust in the midwife. This is again consistent with the existing literature regarding trust relations which, although complex, tends to show a decline in institutional confidence whilst individual trust remains higher (Brownlie, 2008). Explanation can again be found within the interview data, specifically in relation to the receipt of results which some parents noted were either not received or were not given on the basis that "no news is good news".

This can be seen to tie in with parental acceptance of screening on the basis that it is offered by the health service, and this service is trusted. The role of trust has been noted in the context of prenatal screening decisions where the trust in the healthcare system led to the perception that screening was a helpful technology (Santalahti et al., 1998; Chiang et al., 2006). The same was found within the parental interviews conducted here, in which parents talked about the review and vetting processes they felt tests went through, indicating a trust in the systems of accountability in place to govern the NHS.

9.3.3 Knowledge, attitudes and parental decisions

The present study found an association between knowledge and attitudes which in turn had an effect on parental perceptions of decisional quality. This was specifically postulated as a causal relationship; with increased knowledge leading to more positive attitudes leading to a greater perceived quality of decision. This thinking

is consistent with the study of Holtzman et al. (1983) where parents with higher knowledge scores were significantly more in favour of mandatory screening.

This can be compared to studies in prenatal screening where inconsistent results have been found with some failing to find a relationship (Michie et al., 2003) whilst others suggest that those mothers accepting screening, and with positive attitudes, tend to be more knowledgeable than those declining (Michie et al., 1999). Studies in cancer screening have found the converse with increased knowledge being associated with decreased intention to screen for prostate cancer, (Coulter and Fitzpatrick, 2000).

It may, therefore, be that the specific circumstances of newborn screening, and the central location of parental attitudes towards the 'ability to act' on information, are contributing factors in this identified association between perceived knowledge and attitudes. However, more research is required to identify whether this association can be extrapolated to parents who decline screening, or if it is an artefact of the present sample which consisted only of parents who had accepted newborn screening.

9.4 Parental decision-making and informed consent to newborn bloodspot screening

In the UK there is a policy of informed consent (UK National Screening Committee, 2000; Campbell and Ross, 2004; Kenner and Moran, 2005). This is spelt out clearly in the National Screening Programme's own literature which states that:

> "It is important to offer parents an informed choice about screening for their baby, to gain consent and to prepare them for the blood sampling procedure." (UK Newborn Screening Programme Centre, 2008a, p2).

In January 2008 updated guidelines were issued by the UK Newborn Screening Programme Centre for those professionals involved in the collection of the blood spot sample. These guidelines confirm the initial step in the screening process, stating that in order to enable parents to make an informed decision about screening they should have the 'Screening tests for you and your baby' booklet (UK Newborn Screening Programme Centre, 2008a). Parents are given this information at some point during the 3rd trimester of pregnancy but at least 24 hours before screening. No stipulation is provided for exactly when the information is given and so this may vary. Staff are then expected to:

> "Explain fully to parents and then record in the maternity record that newborn blood spot screening has been discussed and recommended, booklet given and consent sought." (UK Newborn Screening Programme Centre, 2008a, p2).

Despite advocating choice there is no data to assess whether an informed consent is being given; none of the standards for assessing screening relate to the consent process. Standard three; that regions should account for 100% of the screening tests offered, in no way ensures that the recorded decisions constitute an informed consent, or even an informed refusal. It does, however, suggest that the data should be available to identify all those parents who have accepted, given a partial acceptance or have declined screening. As such, analysis could, theoretically, be conducted on the percentage of parents who refuse screening for all or some conditions as well as those who accept all conditions.

Whilst this data would be of huge value, the present study suggests that audit data would only capture a small proportion of the elements which play a role in parental-decision making. The results here clearly show that context is key; that the way in which screening is presented and the experiences of parents in their interactions with healthcare staff and the system more widely, are crucial to the determination of both their attitudes towards screening and their retrospective

attitudes towards the decisions taken.

The disaggregation of attitudes has provided insights into the strength of effect of different constructs. When attitudes towards the staff and healthcare system were separated from attitudes towards the specific test the influence on decisional quality by these specific attitudes was reduced. This suggests that the failure to account for these aspects separately overestimates the specific influence of test attitudes on parental decisional quality. When separated, the results indicated that the direct effect of perception of choice had a greater effect on parental perceptions of decisional quality than attitudes towards the screening test itself. This finding implicates the presentation of screening as a key determinant of decisional quality.

The study also highlights areas that are under-researched. More work is required to consider the mechanisms through which perceived choice affects decisional quality and the role of choice in developing parental knowledge. The lack of association between experience, information-seeking, and knowledge is also one that deserves greater attention, and would be extremely pertinent to those involved in the areas of health promotion and parental education. More research is also needed to establish the generalisability of the effects of the identified variables within the context of parental declines.

Appendix A

Invitation letter (interviews)

3rd February 2009

Dear

Invitation to take part in a study of newborn screening experiences

I am writing to invite you to take part in a research study to find out about your experiences and views about newborn blood spot screening, or what you may know as the 'heel prick test'. You are receiving this letter because you have recently had a child. The study is being conducted by Stuart Nicholls at Lancaster University.

In this study you will be asked to take part in one interview, and this should take roughly one hour of your time. Interviews are a good way to find out what you think in a way that allows you to explain in your words what you feel. There is very little research about how parents experience these tests and how they are offered and I hope that this research may be used to develop this process.

Please read the attached information sheet that explains the purpose of this research.

If, in the meantime, you have any questions please do not hesitate to contact Stuart Nicholls, by telephone on 07892721041 or by email at s.nicholls@lancaster.ac.uk.

Thank you for taking the time to read this letter

Yours Sincerely

Dr Kevin Southern
Senior Lecturer and Consultant in Paediatric Respiratory Medicine

Please return the reply slip below in the envelop included. If you do not wish to take part please indicate on the form below, this will ensure you receive no further letters.

Name:..... Ref:.....

Yes, I would like to take part ☐

No, I would not like to take part ☐

Contact details:

Telephone:.....

Address:.....

.....

.....

Postcode:.....

Appendix B

Patient information sheet (interviews)

18 December 2008: version 1.5

Participant Information sheet

Parental experiences of newborn screening

You are being invited to take part in a study to explore the experiences of parents whose children have been offered newborn blood spot screening (or what may have been referred to as the heel prick or Guthrie test) as part of the care for their newborn baby. This leaflet gives you information about this study and how it involves you. Before you decide to take part it is important for you to understand why the research is being done and what it will involve. The research to be carried out has been approved by the Multi Centre Research Ethics Committee (MREC Reference: 08/H1005/85) as well as the ethics committee of Lancaster University.

Purpose of research

The aim of this study is to find out how parents experience newborn blood spot screening, and how they decide whether or not to have their children tested. To do this you have been invited to take part in an interview. Whilst the main outcome will be research to achieve an educational qualification (PhD), I hope that the information from this study will be used to help develop the way that newborn blood spot screening is conducted.

Who is doing the study?

The project is being conducted by Stuart Nicholls, a PhD student at Lancaster University and who is being supervised by Dr Mairi Levitt and Professor Paul Fearnhead, both of whom are also at Lancaster University. For more information about Lancaster University you can visit their website at www.lancs.ac.uk or by contacting Stuart Nicholls. This project forms part of the doctoral research for Stuart Nicholls which is to be completed by September 2010. Stuart is funded by a studentship from the Economic and Social Research Council (ESRC). As part of this studentship Stuart has received training in a range of research methods. In addition to this Stuart has conducted previous research interviewing patients of the health service and this work has been published in international medical journals. If you require any further information please contact Stuart Nicholls on 07892721041 or by email at s.nicholls@lancaster.ac.uk

Why have I been chosen?

You have been chosen because you have had a child during the last twelve months. As such you should have been offered the newborn blood spot screening tests as part of the National Screening Programme for newborn babies.

What will taking part involve?

If you agree to help with the research you will be asked to take part in one interview. This will be informal and will take approximately one hour, although it may be shorter or longer. This interview will explore different aspects of how the newborn blood spot screening tests were offered to you, what you felt about them, and what influenced your decision-making.

Interviews are a good way to find out about peoples experiences and views as they allow the person to put their thoughts in their own words and are less restrictive than a questionnaire.

Risks and benefits of taking part

There are no risks to you in taking part in this research. You will only be asked about your experiences and views. Any comments you make will remain anonymous. This may, however, allow for the possibility of discussing issues that may be upsetting. You do not need to talk about anything which you do not wish to, and you will not be asked to do so. The research will provide information about how parents experience the tests and may identify ways in which this process can be developed. If you would like to, you can receive a copy of the interview as well as a summary of the research. The research will also help to design future studies.

What if I do not wish to take part?

Taking part is completely optional and you can choose not to take part. This will not affect your future treatment in any way. If you do not wish to take part please inform Stuart Nicholls by completing the enclosed reply slip to say this, or contact him by email or telephone.

Confidentiality

All interviews will be conducted with the strictest confidence and your name will not be used in any publication or shown to any person. Only Stuart Nicholls will be able to identify you. Occasionally direct quotations will be used for publication but these will have your name and any identifying comments removed from them. All information will be stored in secure locations as required by the Data Protection Act 1998 and processed under data protection registration Z6328653. Data will be held for a required period of 5 years after which it will be destroyed.

Will I be paid for taking part?

Unfortunately I will not be able to pay you for helping with this study. If you have incurred any expenses as a result of taking part in this study these will be reimbursed.

What will happen to the results?

The results will be compiled as part of a doctoral thesis for submission for the qualification of PhD for Stuart Nicholls. The results may also be published in academic and professional journals and at conferences. No individuals taking part in the study will be identifiable from these results.

Future research

You will not automatically be expected to take part in any future research

What if something goes wrong?

In the unlikely event that you are harmed by taking art in this research project, there are no special compensation arrangements. If you are harmed due to someone's negligence, then you may have grounds for legal action but you may have to pay for it.

Regardless of this, If you have a concern about any aspect of this study, you should ask to speak to Stuart Nicholls who will do his best to answer your questions (tel: 07892721041). If you remain unhappy and wish to complain formally, you can do this through the NHS Complaints Procedure or through Lancaster University. Details of how to do this are available on request.

For further information or comments about the study please contact:

Stuart Nicholls
c/o Mathematics and Statistics,
Fylde College,
Lancaster University,
Lancaster
LA1 4YW
Tel: 07892721041
Email: s.nicholls@lancaster.ac.uk

Alternatively you may contact your local NHS R&D department for further information.

Thank you for taking the time to read this. If you have any further questions please contact Stuart Nicholls

You should keep this information sheet for future reference

Appendix C

Reminder letter (interviews)

6 March 2009

Dear

Invitation to take part in a study of newborn screening experiences

I recently wrote to you regarding the above study and invited you to take part in research to find out about your experiences and views about newborn blood spot screening, or what you may know as the 'heel prick test'. If you have recently replied then please discard this letter.

You are receiving this letter because you have recently had a child. The study is being conducted by Stuart Nicholls at Lancaster University. In this study you will be asked to take part in one interview, and this should take roughly one hour of your time. Interviews are a good way to find out what you think in a way that allows you to explain in your words what you feel. There is very little research about how parents experience these tests and how they are offered and I hope that this research may be used to develop this process.

Please read the attached information sheet that explains the purpose of this research. If you wish to take part please return the enclosed reply slip.

If, in the meantime, you have any questions please do not hesitate to contact Stuart Nicholls, by telephone on 07892721041 or by email at s.nicholls@lancaster.ac.uk.

Thank you for taking the time to read this letter

Yours Sincerely

Dr Kevin Southern
Senior Lecturer and Consultant in Paediatric Respiratory Medicine

Please return the reply slip below in the envelop included. If you do not wish to take part please indicate on the form below, this will ensure you receive no further letters.

Name:..... Ref:.....

Yes, I would like to take part ☐

No, I would not like to take part ☐

Contact details:

Telephone:.....

Address:.....

.....

.....

Postcode:.....

Appendix D

R code for sample size calculation

R code for generating SEM Sample size

```
# Calculate required sample size for test of close fit
#   (RMSEA)
#
# @author Timo Gnambs <timo@gnambs.at>
# @version 2008-09-10
#
# @source MacCallum, R. C., Browne, M. W. & Sugawara, H. M.
#         (1996). Power analysis and determination of
#         sample size for covariance structure modeling.
#         Psychological Methods, 1(2), 130-149.
#




##################### SETTINGS ####################

df <- 109               # Degrees of freedom
alpha <- 0.05           # Significance level
power <- 0.80           # Desired power
rmsea0 <- 0.05          # RMSEA under H0
rmseaa <- 0.08          # RMSEA under H1

###################################################




#initialize values
powa <- 0.0
n0 <- 0
#begin loop for finding initial level of n ;
while(!(powa > power)) {
  n0 <- n0 + 100
  ncp0 <- (n0-1)*df*rmsea0**2
  ncpa <- (n0-1)*df*rmseaa**2
  #compute power
  if(rmsea0 > rmseaa) {
    cval <- qchisq(alpha,df=df,ncp=ncp0)
    powa <- pchisq(cval,df=df,ncp=ncpa)
  } else {
    cval <- qchisq(1-alpha,df=df,ncp=ncp0)
    powa <- 1 - pchisq(cval,df=df,ncp=ncpa)
  }
}
#begin loop for interval halving
dir <- -1
newn <- n0
intv <- 200
powdiff <- powa - power
while(!(powdiff < .001)){
```

```
  intv <- intv*0.5
  newn <- newn + dir*intv*.5
  #compute new power
  ncp0 <- (newn-1)*df*rmsea0**2
  ncpa <- (newn-1)*df*rmseaa**2
  #compute power
  if(rmsea0 > rmseaa) {
    cval <- qchisq(alpha,df=df,ncp=ncp0)
    powa <- pchisq(cval,df=df,ncp=ncpa)
  } else {
    cval = qchisq(1-alpha,df=df,ncp=ncp0)
    powa = 1 - pchisq(cval,df=df,ncp=ncpa)
  }
  powdiff <- abs(powa - power)
  if(powa < power) dir <- 1 else dir <- -1
}
nrmsea <- newn
rm(powa, n0, newn, dir, intv, powdiff, ncp0, ncpa, cval)
print(c('Required N for test of close of fit (McCallum et al., 1996)', ceiling(nrmsea)))
```

Appendix E

Invitation letter (questionnaire)

Alder Hey Children's NHS
NHS Foundation Trust

Alder Hey
Eaton Road
Liverpool
L12 2AP

www.alderhey.com

January 2010

Dear

I am writing to you as the Paediatric Doctor supporting a study to find out about your experiences of the heel prick. This will have been conducted shortly after your baby was born. You have been selected at random from a sample of parents who had a child born in 2008.

The study, conducted by Stuart Nicholls at Lancaster University, aims to explore how you made your decision and what affected this.

If you would like to take part please complete the questionnaire included with this letter and return it in the freepost (no stamp required) envelope enclosed. As a thank you all completed questionnaires will be entered into a draw to win **£20 of high street vouchers**.

Thank you for taking the time to read this letter and I sincerely hope you can spare the time to take part. You should read the information sheet that explains more about the study, and if you have any questions please contact Stuart directly by telephone on 07892721041 or by email at s.nicholls@lancaster.ac.uk.

You have been chosen at random and without sight of medical records. As such we are not aware of an circumstances that may prevent you from taking part. If this is the case please accept my apologies for any inconvenience or distress caused.

Once again, thank you for your time.

Yours Sincerely

Dr Kevin Southern MBChB, MRCP, PhD
Senior Lecturer and Consultant in Paediatric Respiratory Medicine,
member of the Cystic Fibrosis Board, National Newborn Screening Committee.

Appendix F

Patient information sheet (questionnaire)

October 2009: version 1.2

Participant Information sheet: Experiences of the heel prick

You are being invited to take part in a study to explore the experiences of parents whose children have been offered the heel prick as part of the care for their newborn baby. This leaflet gives you information about this study and how it involves you. Before you decide to take part it is important for you to understand why the research is being done and what it will involve. The research to be carried out has been approved by the Multi Centre Research Ethics Committee (MREC Reference: 09/H1005/66) as well as the ethics committee of Lancaster University.

Purpose of research

The aim of this study is to find out how you, as parents, feel about the heel prick and and how you made your decision about whether to have the test or not. To do this you have been invited to complete a short survey. Whilst the main outcome will be research to achieve an educational qualification (PhD), I aim to use the information you provide to help develop the way that heel prick is conducted.

Who is doing the study?

I am Stuart Nicholls, a PhD student at Lancaster University. I am supervised by Dr Mairi Levitt and Professor Paul Fearnhead, both of whom are also at Lancaster University. For more information about Lancaster University you can visit the website at www.lancs.ac.uk or contact me. This project forms part of my doctoral research which is to be completed by September 2010. I am funded by a studentship from the Economic and Social Research Council (ESRC). As part of this studentship I have received training in a range of research methods but have also conducted previous research which has been published in international medical journals. If you require any further information please contact me on 07892721041 or by email at s.nicholls@lancaster.ac.uk

Why have I been chosen?

You have been chosen at random from a sample of parents who have had a child during 2008 and whose tests were analysed by the Merseyside and Cheshire screening laboratory. As such you should have been offered the newborn blood spot screening tests as part of the National Screening Programme for newborn babies.

Continued on the next page....

What will taking part involve?

If you agree to help with the research you will be asked to complete a short questionnaire. This should take roughly 15 minutes. The questionnaire will ask you about the heel prick and how you felt about different things to do with this.

The questionnaire is completely confidential and no one will be able to identify you from the results.

Risks and benefits of taking part

There are no risks to you in taking part in this research. You will only be asked about your experiences and views. Any comments you make will remain anonymous. The research will provide information about how parents experience the tests and may identify ways in which this process can be developed. If you would like to, you can receive a summary of the research.

What if I do not wish to take part?

Taking part is completely optional and you can choose not to take part. If you do not wish to take part please inform Stuart Nicholls by completing the tick box on the questionnaire to say this, or contacting him by email or telephone. This will mean that you receive no further letters.

Confidentiality

All questionnaires will be analysed with the strictest confidence and your name will not be used in any publication or shown to any person. Only Stuart Nicholls will be able to identify you. All results published will be averages from all the people who respond and so no one will see your results. All information will be stored in secure locations as required by the Data Protection Act 1998 and processed under data protection registration Z6328653. Data will be held for a required period of 5 years after which it will be destroyed.

Will I be paid for taking part?

Unfortunately I will not be able to pay you for helping with this study. However, as a thank you, all completed questionnaires will be entered into a draw to win £20 of High Street vouchers.

What will happen to the results?

The results will be compiled as part of a doctoral thesis for submission for the qualification of PhD for Stuart Nicholls. The results may also be published in academic and professional journals and at conferences. No individuals taking part in the study will be identifiable from these results.

Continued over the page....

What if something goes wrong?

It is extremely unlikely that you are harmed by taking part in this research project. However, there are no special compensation arrangements. If you are harmed due to someone's negligence, then you may have grounds for legal action but you may have to pay for it.

Regardless of this, If you have a concern about any aspect of this study, you should ask to speak to Stuart Nicholls who will do his best to answer your questions (tel: 07892721041). If you remain unhappy and wish to complain formally, you can do this through the NHS Complaints Procedure or through Lancaster University. Details of how to do this are available on request.

For further information or comments about the study please contact:

Stuart Nicholls
c/o Mathematics and Statistics,
Fylde College,
Lancaster University,
Lancaster
LA1 4YF
Tel: 07892721041
Email: s.nicholls@lancaster.ac.uk

You should keep this information sheet for future reference

Appendix G

Reminder letter (questionnaire)

Alder Hey Children's NHS
NHS Foundation Trust

Alder Hey
Eaton Road
Liverpool
L12 2AP

www.alderhey.com

January 2010

Dear

I recently wrote to you regarding a study about your experiences of the heel prick and invited you to take part in this research.

If you have recently replied then please discard this letter.

The study, conducted by Stuart Nicholls at Lancaster University aims to explore how you made your decision about the heel prick and what affected this. If you would like to take part please complete the questionnaire included with this letter and return it in the freepost (no stamp required) envelope enclosed. As a thank you all completed questionnaires will be entered into a draw to win **£20 of high street vouchers**.

Thank you for taking the time to read this letter and I sincerely hope you can spare the time to take part. You should read the information sheet included which explains more about the study, and if you have any questions please contact Stuart directly by telephone on 07892721041 or by email at s.nicholls@lancaster.ac.uk.

You have been chosen at random and without sight of medical records. As such we are not aware of an circumstances that may prevent you from taking part. If this is the case please accept my apologies for any inconvenience or distress caused.

Once again, thank you for your time.

Yours Sincerely

Dr Kevin Southern MBChB, MRCP, PhD
Senior Lecturer and Consultant in Paediatric Respiratory Medicine,
member of the Cystic Fibrosis Board, National Newborn Screening Committee.

Appendix H

Questionnaire & coding scheme

Reference Number: _____

LANCASTER UNIVERSITY

Experiences of the heel prick

This survey asks you about your experiences of newborn bloodspot screening, or what may have been referred to as the heel prick (the procedure whereby a small amount of blood is taken from your baby's heel for testing). In answering please think about how you felt at the time and, if you have more than one child, please answer for your most recent child. If you wish to complete the survey on line please visit www.maths.lancs.ac.uk/~nichols1

Thank you for your time and for completing this questionnaire. Please remember that all completed questionnaires will be entered into a prize draw to **win £20 of high street vouchers**.

Please tick this box and return the survey if you do not wish to take part: ☐

Information

In this section you will be asked about your general experiences with your doctor or nurse before moving on to how you found out about the heel prick and your views about the information you found.

1. Please tell me about your usual experiences, such as when you go to see your own doctor. Please tick whether you agree or disagree:

	Agree	Disagree
Instead of waiting, I usually ask the doctor or nurse immediately after an examination about my health	1	0
I usually ask the doctor or nurse lots of questions about the procedures during a medical examination	1	0
I'd rather be given many choices about what's best for my health than to have the doctor make the decisions for me	1	0
I usually don't ask the doctor or nurse many questions about what he or she is doing during a medical examination	1	0
I'd rather have doctors and nurses make the decisions about what's best than for them to give me a whole lot of choices	1	0
It is better to trust doctor or nurse in charge of a medical procedure than to question what she or he is doing	1	0
I usually wait for the doctor or nurse to tell me about the results of a medical exam rather than asking him or her immediately	1	0

Continued on the next page...

2. Thinking now about the heel prick, please tick any sources of information that you used when finding out about the heel prick (*please tick **all** that apply*):

The midwife....	1
Friends....	1
Family....	1
NHS leaflet/book....	1
Other leaflet/book....	1
Television/radio....	1
Internet....	1
Other (please state)_____	1

3. Which would you say was the most important source of information about the heel prick? (*please tick **one** only*):

The midwife....	1
Friends....	2
Family....	3
NHS leaflet/book....	4
Other leaflet/book....	5
Television/radio....	6
Internet....	7
Other (please state)_____	8

4. To what extent did you see the same midwife during your antenatal visits? (*please tick **one** only*):

Never....	1
Rarely....	2
Seldom....	3
Sometimes....	4
Occasionally....	5
Most of the time....	6
Always....	7

5. Do you have a friend or member of the family with a condition diagnosed through the heel prick?

Yes....	1
No....	0

Continued on the next page...

6. Thinking back to when you had your baby; please answer the following:

	Yes	No
Before your first baby was born had you heard about the heel prick ?	1	0
Before your baby was born had you heard about testing for phenylketonuria (PKU)?	1	0
Before your baby was born had you heard about testing for congenital hypothyroidism (CH)?	1	0
Before your baby was born had you heard about testing for cystic fibrosis (CF?)	1	0
Before your baby was born had you heard about testing for sickle cell disease (SCD)?	1	0
Before your baby was born had you heard about testing for medium chain co-acyl dehydrogenase deficiency (MCADD)?	1	0

7. For each of the following statements please tick the extent to which you agree or disagree:

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree
I feel I understand *why* the heel prick is done	4	3	2	1	0
I feel I understand why the test is done at the time it is	4	3	2	1	0
I feel I understand what the test results mean	4	3	2	1	0
I feel I understand how the test is done	4	3	2	1	0
I feel I understand when the test is done	4	3	2	1	0
I feel I understand when the results will be available	4	3	2	1	0
I feel I understand what the conditions are that the heel prick tests for	4	3	2	1	0
I feel I understand how the conditions could affect my child	4	3	2	1	0
I feel I understand how the conditions would be dealt with if found	4	3	2	1	0

Continued on the next page...

Attitudes to screening

In this next section I would like to ask you about your views regarding the heel prick.

8. For each of the following statements please tick the extent to which you agree or disagree:

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree
I feel my child is at risk of having a condition which the heel prick tests for	4	3	2	1	0
The heel prick is an important test	4	3	2	1	0
The conditions screened for are rare	4	3	2	1	0
The heel prick is likely to cause a lot of pain to my child	0	1	2	3	4
The heel prick causes long lasting pain to my child	0	1	2	3	4
The heel prick is likely to have a long term impact on my child	0	1	2	3	4
The heel prick exposes my child to the risk of infection	0	1	2	3	4
The heel prick is a quick test	4	3	2	1	0
The heel prick can provide useful information.	4	3	2	1	0
The heel prick will allow treatment of an identified health problem	4	3	2	1	0
The conditions tested for in the heel prick are serious	4	3	2	1	0
Testing earlier is better than testing later	4	3	2	1	0

Continued on the next page...

Decision-making

In this next section I would like to ask about how you felt about aspects of making the decision about the heel prick.

9. For each of the following statements please tick the extent to which you agree or disagree:

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree
I felt I had enough time to make a decision about the heel prick	4	3	2	1	0
I was too tired to make a decision about the heel prick	0	1	2	3	4
I was too emotional to make a decision about the heel prick	0	1	2	3	4
I did not feel able to make a decision about the heel prick	0	1	2	3	4
The heel prick is a routine test for all babies	0	1	2	3	4
It was expected that my child had the heel prick	0	1	2	3	4
The heel prick was presented as an optional test	4	3	2	1	0
I felt I had a choice to decline the test	4	3	2	1	0
I am clear about the best choice from me	0	1	2	3	4
I feel sure about what to choose	0	1	2	3	4
This decision is easy for me to make	0	1	2	3	4
I feel I have made an informed choice	0	1	2	3	4
My decision shows what is important to me	0	1	2	3	4
I expect to stick with my decision	0	1	2	3	4
I am satisfied with my decision	0	1	2	3	4

Continued on the next page...

General attitudes towards medicine

In this section you will be asked bout your views more generally about the National Health Service, hospitals generally and your specific views about your midwifery care.

10. In the following statements please rate the extent to which you agree or disagree:

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree
I want to have all the tests offered by the midwife or hospital	4	3	2	1	0
I completely trust my midwife's decisions about which medical tests are best for my child	4	3	2	1	0
The midwife is totally honest in telling you about all of the different options available	4	3	2	1	0
The midwife will answer my questions to the best of her abilities	4	3	2	1	0
I am confident in abilities of the midwife	4	3	2	1	0
All in all I trust the midwife completely	4	3	2	1	0
I am confident in the accuracy of test results from the hospital	4	3	2	1	0
I always receive test results from the hospital	4	3	2	1	0
I feel like I have to double check everything the hospital does	0	1	2	3	4
The National Health Service (NHS) only provides tests that are important	4	3	2	1	0
The National Health Service (NHS) provides unbiased information	4	3	2	1	0
The National Health Service (NHS) ensures tests are safe	4	3	2	1	0

About you

This next section asks you for some background information.

This information will only be used to compare the answers from different people to this questionnaire and not be used by anybody else.

11. Age (*please tick **one** only*):

Under 21....	1
21-30....	2
31-40....	3
41-50....	4
51-60....	5
61 or above....	6

12. Total number of children (*please tick **one** only*):

1....	1
2....	2
3....	3
4....	4
5 or more....	5

13. Where was your *most recent* child born? (*please tick **one** only*):

NHS Hospital.... (please give hospital name)	1
Private Hospital... (please give hospital name)	2
Home....	3
Other (please state).....	4

14. Ethnic group (*please tick **one** only*):

White....	1
Black African....	2
Black Caribbean....	3
Black Other....	4
Indian....	5
Pakistani....	6
Bangladeshi....	7
Chinese....	8
Other (please state)	9

15. Highest level of educational attainment (*please tick **one** only*):

School/GCSE....	1
College/A-Level....	2
Undergraduate degree....	3
Postgraduate degree....	4
Professional qualification....	5
Other (please state)....	6

16. What is your estimated household income (including benefits) before tax? (*please tick **one** only*):

Less than £11500....	1
£11501 – £18500....	2
£18501 - £28000....	3
£28001 – 41000....	4
£41001 -75000....	5
Over £75000....	6

Please tick the box below if you would like a summary of the findings when complete:

1

Thank you for your time and for helping with this research.

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