



September 2005

**Survey of information
resources on newborn blood
spot screening for parents
and health professionals:
a systematic review**

Katrina Hargreaves, Ruth Stewart and Sandy Oliver

This review has been produced by members of the Social Science Research Unit within the Perspectives, Participation and Research stream. It has used methods developed in SSRU's Evidence for Policy and Practice Information and Co-ordinating Centre (EPPI-Centre).

This report should be cited as:

Hargreaves K, Stewart R and Oliver S (2005). *Survey of information resources on newborn blood spot screening for parents and health professionals: a systematic review*. London: Social Science Research Unit, Institute of Education, University of London, pp 1-76. ISBN: 0-9550487-2-9

Katrina Hargreaves, Ruth Stewart and Sandy Oliver are researchers at the Social Science Research Unit, the Institute of Education, University of London, funded by the Department of Health as part of the UK Newborn Screening Programme Centre; the views expressed in this publication are those of the authors and not necessarily those of the Department of Health

A glossary of terms used in this report is attached as Appendix 8.

Acknowledgements

We would like to acknowledge the invaluable help of the many people who gave us leaflets or told us how to find them, including staff in the following organisations: the NHS Sickle Cell and Thalassaemia Screening Programme; St James's University Hospital, Leeds; University Hospital of Wales; Brent Sickle Cell & Thalassaemia Centre, Birmingham Children's Hospital; NHS, Scotland; United Hospitals, Northern Ireland; Addenbrooke's NHS Trust, Cambridge; The Royal Hospitals Trust, Belfast, Northern Ireland; Royal Berkshire Hospital; St Helier Hospital, Surrey; Sheffield Children's Hospital NHS Trust; Northhampton General Hospital; The Ipswich Hospital NHS Trust; Southend Hospital NHS Trust; Maidstone and Tunbridge Wells NHS Trust; South East Thames Newborn Screening Laboratory Partnership; Peterborough Hospitals NHS Trust; Cystic Fibrosis Trust; Sickle Cell Society; National Society for Phenylketonuria (NSPKU); and the Children Living with Inherited Metabolic Diseases (Climb).

© SSRU 2005

This systematic review contributed data that is included on the EPPI-Centre website. The EPPI-Centre owns the copyright for all material on the website it has developed, including the contents of the databases, manuals, and keywording and data-extraction systems. The centre and authors give permission for users of the site to display and print the contents of the site for their own non-commercial use, providing that the materials are not modified, copyright and other proprietary notices contained in the materials are retained, and the source of the material is cited clearly following the citation details provided. Otherwise users are not permitted to duplicate, reproduce, re-publish, distribute, or store material from this website without express written permission.

TABLE OF CONTENTS

- EXECUTIVE SUMMARY 1
- Background and aims 1
- Methods 1
- Findings 2
- Conclusions 3
- Recommendations 3
- 1. BACKGROUND AND AIMS 4
- 2. METHODS 6
 - 2.1 Research question and inclusion criteria 6
 - 2.2 Searching for information on newborn blood spot screening 6
 - 2.3 Cataloguing leaflets and information sheets 8
 - 2.4 Identifying relevant leaflets 8
 - 2.5 Developing an appraisal tool 8
 - 2.6 Appraising the leaflets 10
- 3. RESULTS: APPRAISAL OF LEAFLETS 11
 - 3.1. Purpose of the leaflet 13
 - 3.2 Reasons for screening 14
 - 3.3 Process of screening 15
 - 3.4 Consequences of screening 17
 - 3.5 Follow-up to screening 18
 - 3.6 Information about Carriers 19
 - 3.7 Production of the leaflet 19
- 4. RESULTS: APPLYING STANDARDS FOR PERSONAL DECISIONS 21
 - 4.1 Standards for personal decisions 22
- 5. RESULTS: APPLYING STANDARDS FOR POLICY DECISIONS 25
 - 5.1 Standards for policy decisions 26
- 6. DISCUSSION AND CONCLUSIONS 31
 - 6.1 Summary of findings 32
 - 6.2 Strengths and weaknesses of the study 32
 - 6.3 Other surveys about patient information 33
 - 6.4 How do leaflets match parents' information needs? 34
 - 6.5 What has this survey achieved? 35
 - 6.6 Conclusions and recommendations 35
- REFERENCES 37
- Appendix 1: The DISCERN Instrument 39
- Appendix 2: NSC criteria for mounting a screening programme 44

Appendix 3: Bringing together DISCERN and NSC criteria to develop our appraisal tool	46
Appendix 4: Appraisal tool for leaflets	50
Appendix 5: Leaflets and Information Sheets included in the Survey.....	53
Appendix 6: Applying principles underpinning DISCERN criteria to information about screening	59
Appendix 7: Applying principles underpinning NSC criteria to information available to parents.....	61
Appendix 8: Glossary.....	64

EXECUTIVE SUMMARY

Background and aims

In the United Kingdom, newborn screening for phenylketonuria (PKU) was introduced on a national level in 1969 and for congenital hypothyroidism (CHT) in 1981. Recent developments in newborn blood spot screening in the UK, however, have highlighted the need to re-examine information and communication needs for parents and health professionals in this area. These developments include: policy decisions in 2000 and 2001 to introduce screening for cystic fibrosis and sickle cell disorders respectively; innovations in screening technologies that have increased the number of disorders that can be detected from dried blood spot specimens; and raised expectations for informed consent.

A recent systematic review of existing reviews on newborn blood spot screening and communication about screening revealed gaps in the research literature on communication for newborn screening.¹ It also highlighted the lack of primary research on the information and communication needs of parents and health professionals in the newborn period.

Research carried out by the Parent Support Research Team of the UK Newborn Screening Programme Centre has aimed to address some of the gaps in the literature. A qualitative study was undertaken to assess parents' and health professionals' information needs. In addition, the team carried out this survey of existing information resources on newborn blood spot screening for parents and health professionals, which is the focus of this report.

The aims of the survey were to:

- Identify existing information resources on newborn blood spot screening available to parents and health professionals internationally
- Develop an appraisal tool to evaluate the quality of the existing information resources, with a view to identifying strengths and weaknesses of the available information
- Draw on the survey of leaflets to inform the development of our own information resources: initially, a standardised pre-screening leaflet for parents throughout the UK.

Methods

Leaflets and other information resources on newborn blood spot screening were sourced through internet websites, health service providers and support organisations in the UK.

Information resources on newborn blood spot screening and the conditions screened for were catalogued electronically for easy retrieval and analysis. To identify leaflets that were relevant to the task of developing a general parent

leaflet, all leaflets that described the heel-prick test were included in the survey of leaflets. Those that merely mentioned the procedure and/or contained information about the conditions only were excluded.

An appraisal tool was developed by adapting DISCERN criteria for judging the quality of patient information on treatment choices, and National Screening Committee (NSC) criteria for making policy decisions about the implementation of screening programmes. Two researchers tested the tool by applying it to a limited number of leaflets, compared their answers and achieved agreement on what information was required to meet our criteria. The finalised tool contained questions grouped under the following sub-headings: purpose of the leaflet; reasons for screening; process and consequences of screening; follow-up to screening; production of the leaflet.

Findings

Some of the key findings include:

- Of the 106 leaflets included in the survey (see Appendix 5), most were for parents (79/106) and this was evident in the title of about half of them (42/79) and in the text of the great majority (67/79).
- The vast majority of the leaflets (91/106) explained the aims of screening; three-quarters provided some information on the natural history of at least one of the conditions (79/106). Less than half of the leaflets (43/106), however, mentioned prevention or the difficulties of prevention.
- The great majority of leaflets described the procedures for doing the blood spot screening test particularly how the blood sample is taken (93/106) and the possibility of repeat testing (76/106)
- Two-thirds of leaflets (68/106) did not indicate whether or not screening was compulsory or mandatory, though many of these implied that screening was routine.
- Less than half of the leaflets indicated when parents would receive screening results (45/106).
- About half the leaflets contained information about the limitations or possible harms of screening (55/106).
- The majority of leaflets indicated the need for follow-up tests to confirm diagnosis after a positive screening result (71/106).
- The vast majority of leaflets cited no source of evidence for the information provided (89/106).
- Over half the leaflets indicated the date of development and/or review (56/106), but the great majority (90/106) did not mention how the information was developed.

- Overall, the leaflets/information sheets rated better on issues rooted in DISCERN criteria for the provision of information on treatment choices, than on NSC criteria for choices regarding whether or not to introduce a screening programme.

Conclusions

Most leaflets provided information on their purpose, the aims of screening, the conditions screened for, the screening procedure, the need for follow-up to confirm positive screening results, and treatment for the conditions. The appraisers, however, were generous when using the appraisal tool. In some cases the information provided in the leaflets barely met the requirement. Whilst most leaflets mentioned the benefits of screening, few addressed the issue of the possible harms, particularly false results, indicated that screening was a choice, discussed alternatives to screening, or gave information about when and how parents would receive results. Fewer still provided any sources of evidence for the information provided or how the information was developed.

Recommendations

We recommend:

- the use of DISCERN with topic specific guidelines as a starting point for developing patient information;
- appraising patient information with DISCERN and a topic specific tool to access good practice in information provision.

1. BACKGROUND AND AIMS

Newborn screening programmes are an important part of health care services because they enable early detection and treatment of certain disorders associated with significant morbidity or mortality.^(1, 2) In the United Kingdom, newborn screening for phenylketonuria (PKU) was introduced on a national level in 1969 and for congenital hypothyroidism (CHT) in 1981.

Recent developments in newborn screening, however, have led to the need to re-examine information and communication needs of parents and health professionals in this area. Policies to introduce newborn screening for sickle cell disorders and cystic fibrosis using the same Guthrie card blood sample were announced in 2000 and 2001 respectively. Innovations in DNA technologies and tandem mass spectrometry, which increase the number of disorders that can be detected from dried blood spot specimens, have introduced further complexity. Moreover, raised expectations for informed consent for treatment, screening, diagnosis, and research have led to the need to communicate clearly with parents about their child's health and the choices that they face in relation to testing and its possible consequences. Furthermore, with the expansion of screening services to include cystic fibrosis and sickle cell disorders, research is needed to understand how to provide parents and health professionals with information relevant to all four conditions.

Our plan, therefore, was to develop and pilot information resources for parents and health professionals and communication guidelines for health professionals. Three separate but related pieces of work have informed this process.

First, a systematic review of existing reviews on newborn blood spot screening and on communication about screening commissioned by the Cochrane Collaboration and the NHS Health Technology Assessment programme revealed gaps in the research literature on communication for newborn screening.⁽³⁾ Most striking is the lack of primary research that considers the information and communication needs of parents and health professionals in the newborn period. The review also highlighted the need for good-quality standardised information resources about screening.

Second, focus groups and interviews with parents and health professionals have also helped us identify information needs of these groups.

Finally, to draw on the strengths of existing materials, we carried out an international survey of information resources (leaflets and information sheets) currently available through the internet and health service providers. This work is reported here, and the implications considered for the development of a leaflet on newborn blood spot screening and other information resources available to both parents and health professionals. This work is also reported in a paper in a peer-reviewed journal.²

The aims of the survey were to:

- Identify existing information resources on newborn blood spot screening available to parents and health professionals internationally

- Develop an appraisal tool to evaluate the quality of the existing information resources, with a view to identifying strengths and weaknesses of the available information
- Draw on the survey of leaflets to inform the development of our own information resources: initially, a standardised pre-screening leaflet for parents throughout the UK.

2. METHODS

Outline of Chapter

This chapter describes the methods used in the survey: namely, identification of leaflets and www pages, development of the survey instrument, and the methods of analysing the data.

2.1 Research question and inclusion criteria

We sought to answer the research question: what written information is publicly available about routine newborn blood spot screening for phenylketonuria, congenital hypothyroidism, cystic fibrosis and sickle cell disorders? The purpose of identifying these documents was to inform the development of a pre-screening leaflet by building on existing materials. Documents to be included needed to:

- i. DESCRIBE newborn blood spot screening (i.e. included reference to taking blood from the baby's heel for screening tests) or, as it is sometimes known, the heel-prick or the Guthrie test.

Documents were to be excluded if they:

- ii. MENTIONED newborn screening but did not include information about testing blood taken from the baby's heel; or
- iii. referred to one or more of the four conditions screened for but contained NO information on newborn screening itself.

2.2 Searching for information on newborn blood spot screening

Leaflets and information sheets on newborn blood spot screening were sought through

- Internet websites
- Health service providers
- Support organisations

2.2.1 Internet websites

The following categories of website were searched using the Google search engine:

- i. Children's hospitals in the UK, USA, Canada and Australia. These were selected as leading providers of health care for children affected by the screened conditions;

- ii. Support organisations and charities in the UK (see Section 1.3);
- iii. Links provided on the above websites to specialist websites with information on newborn blood spot screening and/or the particular conditions;
- iv. Professional bodies and health organisations (e.g. Department of Health, National Health Service (NHS), American Association of Paediatricians (AAP)).

Sections on the websites referring to patient/consumer/parent information or health professional information were searched to identify information on newborn blood spot screening generally, and information or fact sheets on the specific conditions phenylketonuria (PKU), congenital hypothyroidism (CHT) cystic fibrosis (CF) and sickle cell disorders (SC).

The search facility for each website was used to search for the following words and phrases:

- i. 'newborn screening'
- ii. 'newborn screening program(me)'
- iii. 'neonatal screening'
- iv. 'in-born errors of metabolism'
- v. 'phenylketonuria' and 'PKU'
- vi. 'hypothyroidism'
- vii. 'cystic fibrosis'
- viii. 'sickle cell'

Many websites did not appear to contain any information on newborn blood spot screening. Each site was searched thoroughly before it was determined that no relevant information could be found on the site. All relevant leaflets/information sheets found on the internet were printed out and catalogued.

2.2.2 Health service providers

Leaflets and information sheets were sought from health service providers, including all laboratories screening for sickle cell disorders and cystic fibrosis in England and Wales, specialists other individuals involved in wider Programme Centre networks such as sickle cell counsellors, cystic fibrosis nurses, midwives, and screening programme co-ordinators. Leaflets and information sheets were also provided by the Sickle Cell and Thalassaemia Screening Programme who undertook a scoping exercise to determine the range of resources available to parents on newborn blood spot screening.⁽⁴⁾

2.2.3 Support organisations

We contacted the following support organisations for leaflets on newborn screening and the four conditions to be screened for nationally:

- Cystic Fibrosis Trust (CF Trust)
- Sickle Cell Society
- British Thyroid Foundation
- National Society for Phenylketonuria (NSPKU)
- Contact a Family
- Children Living with Inherited Metabolic Diseases (Climb)
- National Childbirth Trust (NCT)

2.3 Cataloguing leaflets and information sheets

All leaflets on newborn screening and/or the four conditions were numbered, to facilitate accurate retrieval and analysis, and catalogued on a Reference Manager database with their citation details. They were coded for: the condition being screened (PKU, CHT, SCD, CF, or a general leaflet on newborn blood spot screening); their target audience (e.g. parent, health professional); and country of origin (e.g. UK, USA, AUS, etc.).

2.4 Identifying relevant leaflets

One researcher conducted the search and was over inclusive in applying the inclusion criteria. Two researchers independently applied the inclusion criteria to all documents initially identified. Discrepancies were resolved through discussion.

2.5 Developing an appraisal tool

We drafted an appraisal tool to evaluate the leaflets. We drew on two existing sets of criteria: the DISCERN instrument, which was developed to evaluate the quality of consumer health information on treatment choices (see Appendix 1),⁽⁵⁾ and the NSC criteria for determining the appropriateness of potential screening programmes (see Appendix 2).⁽⁶⁾

Whilst the intervention(s) considered by DISCERN involves treatment, the intervention for the NSC is screening itself. To draw together these criteria they were first grouped into the following themes:

- The conditions (treated / screened for)
- Different options
- Decision-making

- The intervention process
- What happens next
- The effectiveness of the intervention

Grouping the criteria in this way, enabled cross cutting issues to be identified, and a set of synthesised criteria drafted that took into account DISCERN's focus on high quality information for treatment choices and the NSC's focus on screening. Whilst not all DISCERN or NSC criteria were appropriate for inclusion in a tool for appraising parent information on newborn screening, most of these criteria could be adapted to the screening context.

To refine the tool, two researchers independently applied the questions to four separate leaflets/web pages on newborn blood spot screening. Responses to the questions were compared and discussed between the two researchers; where necessary the questions were clarified in order to achieve more consistent responses. The refined appraisal tool was applied to ten more leaflets/web pages and then a further five for a total of three rounds of comparing independent appraisals and refining the tool as necessary. The resulting appraisal tool focused on five criteria for assessment of content comprising purpose of leaflet, reasons for screening, process and consequences of screening, follow-up to screening, and production of the leaflet. The appraisal tool is attached as Appendix 3.

The finalised questions included in the appraisal tool were organised into subheadings to prepare a template for high-quality newborn screening information for parents and health professionals (see Box 1).

Box 1: Issues addressed when appraising leaflets**Purpose of Leaflet**

Is it clear whom it is for?

If so, how is this indicated in the leaflet?

Is it clear when the information would be given?

Reasons for Screening

Aims/reasons for screening

Natural history of the conditions screened for

Prevention programmes/difficulties of prevention

Process and Consequences of Screening

Description of the heel-prick test

Indication of when parents will receive results

Information on whether screening is not compulsory or mandatory

The limitations of screening (e.g. false-negative, false-positive results)

Costs of screening

Cost-effectiveness of screening

Follow-up to Screening

Information on need for further testing for diagnosis

Treatment for the conditions screened for

Any related services

Information about Carriers

Babies may be identified as carriers

Babies' DNA may be tested

Mutations tested for

Implications of carrier status for babies' health

Parents may be identified as carriers

Wider implications for families

Production of the Leaflet

Sources of evidence

Is specific information linked to evidence?

Level of complexity of the leaflet

Indication of how the leaflet was developed

2.6 Appraising the leaflets

Having finalised the set of questions, the appraisal tool was then entered into specialist software (EPPI Reviewer) and used to evaluate each individual leaflet. Some minor modifications were made to the set of questions at this stage as we gained a greater understanding of the leaflets. Two researchers independently evaluated each leaflet. Discrepancies were discussed and resolved.

3. RESULTS: APPRAISAL OF LEAFLETS

Outline of Chapter

This chapter describes the findings of the survey: namely, how well the leaflets and information sheets reported their purpose, the reasons for screening, the process and consequences of screening, follow-up care, and how the leaflet was produced.

Key findings

Purpose of the leaflet

- *Most leaflets were for parents (79/106) and this was evident in the title of about half of them (42/79) and in the text of the great majority (67/79).*
- *About half of the leaflets (50/106) were developed to provide information prior to screening.*

Reasons for screening

- *The vast majority of the leaflets (91/106) explained the aims of screening generally, for a specific population, or both. The 15 leaflets that did not contain this information, were guidelines for health professionals or condition-specific leaflets, rather than general information resources for parents.*
- *Three-quarters of the leaflets provided some information on the natural history of at least one of the conditions (79/106).*
- *Less than half of the leaflets (43/106) mentioned prevention or the difficulties of prevention; almost all of these mentioned whether or not the conditions were inherited (39/43), and some mentioned reproductive choice or other means of prevention (11/43).*

Process of screening

- *The great majority of leaflets describe the procedures for doing the blood spot screening test particularly how the blood sample is taken (93/106), the possibility of repeat testing (76/106), what happens to the card once the blood spots are taken (72/106). Few leaflets indicate that the test will be uncomfortable for the baby (19) or how the pain might be eased (5).*
- *Two-thirds of leaflets (68/106) did not indicate whether or not screening was compulsory or mandatory, though many of these implied that screening was routine. A quarter of the leaflets state that screening is not compulsory (25/106), but several of these also emphasise the importance of screening by recommending screening (10) or outlining the implications of not screening (4).*
- *All 16/106 leaflets stating that screening is mandatory were from the United States where it is mandated by law.*

Consequences of screening

- *Less than half of the leaflets indicated when parents would receive screening results (45/106). Less than half of these mentioned both negative and positive results (17/45).*

- *Less than a quarter of the leaflets contained information about the limitations or possible harms of screening in terms of the risk of false negative (15/106) or false positive results (22/106). More leaflets, however, mentioned other possible harms such as anxiety associated with the need for repeat tests (41/106).*
- *Few of the leaflets mentioned the costs of screening (14/106) and fewer still the cost-effectiveness (5/106). Those mentioning costs or cost-effectiveness were almost all from the United States.*

Follow-up to screening

- *The majority of leaflets indicated the need for follow-up tests to confirm diagnosis after a positive screening result (71/106). Almost all of these (69/71) informed about the circumstances in which follow-up would occur, but few leaflets indicated when parents would be told about the need for follow-up tests (9), when such tests might occur (12), or when parents would receive results of follow-up tests (1).*
- *Most leaflets mentioned treatment for at least one of the four conditions (73/106) and almost all of those mentioning treatment also gave some information about the natural history of the conditions.*
- *Of the 66 leaflets that referred to a service related to newborn screening, the majority (48/66) referred to health professionals or specialists, and over a third referred to support organisations (23/66).*

Information about Carriers

- *Almost half of the leaflets (49/106) referred to carriers. About half of these (23/49) indicated that, as an outcome of screening, babies may be identified as carriers of the conditions screened for, or that parents might be carriers (24/49).*

Production of the leaflet

- *The vast majority of leaflets cited no source of evidence for the information provided (89/106). Seventeen of the leaflets included reference to policy documents, research papers or named specialist reviewers.*
- *Over half the leaflets indicated the date of development and/or review (56/106), but the great majority (90/106) did not discuss how the information was developed.*
- *Over half the leaflets were appraised as 'easy to read' (56/106); about a third contained technical terms that were unexplained (36/106) and 14 leaflets required some expert knowledge to understand.*

Over 300 leaflets were found in our initial search for information sheets/leaflets on newborn blood spot screening. 106 leaflets met our inclusion criteria of describing newborn blood spot screening: thirty-four from the USA (33%), 68 from the UK (64%) and four from Australia (4%).

The results of the survey are set out below in three sub-sections. The first reports the results from applying our appraisal tool, the second and third sections report how well the leaflets matched DISCERN and NSC criteria respectively.

The questions selected for inclusion in our appraisal tool were grouped under the following sub-headings: the purpose of the leaflet; reasons for screening; process of screening; consequences of screening; follow-up to screening, and the production of the leaflet.

3.1. Purpose of the leaflet

The purpose of each leaflet was considered in terms of who it was for and for when it was provided.

Seventy-nine (75%) of the 106 leaflets clearly indicated that they were intended to provide information for parents or carers of babies. Nineteen of the leaflets indicated that they were specifically written for health professionals. One leaflet was intended for both parents and health professionals, one was for parents and wider family members and one was for parents and adults more generally. For eight of the leaflets, the intended audience was not clearly specified (see Table 1.1).

Table 1.1: Is it clear whom the leaflet is for?*

Target audience	Number
Parent/carer	79
Families	1
Adults	2
Health professionals	19
Not clear for whom leaflet intended	8
Total number of leaflets	106

* Some documents appear in more than one category

We then looked at whether the intended audience was indicated in the title of the leaflet or in the text. Of the 98/106 leaflets that were clear about the intended audience, this was more likely to be indicated in the text (82/98) than in the title of the leaflet (45/98). In over a third of the leaflets clear about their intended audience (36/98) this was indicated in both the title and the text. Most leaflets were for parents (79/106) and this was evident in the title of over half (42/79) and the text of four-fifths of the leaflets (67/79). To be indicated in the text, the leaflet needed to contain phrases such as 'your baby' or 'my baby'. Illustrating this point, some leaflets intended for parents provided information by posing and answering questions commonly asked by parents about screening, for example,

“Q1. Did I do something wrong in my pregnancy which resulted in my baby having congenital hypothyroidism?” “A. The answer is most certainly NO” [128].*

In 45/106 leaflets it was not clear at what stage in the screening pathway the leaflet was to be given. In 50, it was clear that the information was to be given before screening took place (antenatally or postnatally). Three of these 50 leaflets were also given as part of a consent process by screening programmes in

* A list of numbered leaflets included in this survey is attached as Appendix 1.

the USA that were carrying out pilot research for supplemental screening programmes, and one was part of a consent process for newborn screening in Scotland. The majority of the leaflets to be given before screening took place also described how the blood sample was taken from the baby's heel (45/50). 39/50 discussed the possibility of a repeat screening test, and 32/50 described how results were reported to parents.

Other leaflets provided information on conditions which was clearly to be given to parents with positive screening results (8/106), after a confirmed positive diagnosis (2/106), or with a request for a repeat blood sample (1/106).

Table 1.2: Is it clear from the leaflet when in the screening pathway this information would be given?*

Timing of information giving	Number
Yes, Before screening takes place (antenatally or postnatally)	50
Yes, As part of the consent process	4
Yes, With screening results	8
Yes, After confirmed positive diagnosis	2
Yes, at other time (e.g. when repeat blood sample requested)	1
No	45
Total number of leaflets	106

* Some documents appear in more than one category

3.2 Reasons for screening

Over four-fifths of the leaflets (91/106) explained the aims or reasons for screening (see Table 1.3). Researchers noted that during the appraisal process, however, reasons were often implied rather than the aims of screening being explicit. Fifteen of the 106 leaflets gave no reasons for screening. These included leaflets describing screening protocols for health professionals, or providing information on a specific condition screened for rather than newborn screening generally (e.g. 'Information for parents/carers for cystic fibrosis' [46], 'Hypothyroidism and your infant' [160]). Others gave results information to parents whose babies received a positive screening result. One US leaflet for health professionals provided bullet-point information to be passed on to patients about ensuring that newborn babies were screened before leaving hospital.

Eighty-eight out of the 106 leaflets explained why screening tests were recommended. Twenty-four explained the importance of screening a particular population, often providing statistics about the frequency of occurrence of one or more of the disorders in that population. For example, one leaflet for parents describes PKU as a "rare disorder that affects 1 in every 8,000 babies born in Scotland" [268]. Twenty-one of the 106 leaflets explained both the reasons for screening generally and the importance of screening a particular population for a certain disorder.

Table 1.3: Does the leaflet give the aims of/reasons for screening?*

Aims of screening	Number
Yes, Explains why the tests are recommended/important (list conditions covered)	88
Yes, Explains importance to screen a particular population (list conditions covered)	24
No	15
Total number of leaflets	106

* Some documents appear in more than one category

Seventy-nine leaflets (75%) provided some information on the natural history of at least one of the conditions; 27 did not provide information about any of the four conditions, though one of these provided internet hyperlinks for condition-specific information. The conditions most likely to be described were CHT (62/106) and PKU (58/106). About a quarter of the leaflets described sickle cell disorders (29/106) or cystic fibrosis (27/106), and 22 described other conditions.

We were interested in the extent to which leaflets explained the importance of screening for these conditions and the lack of other prevention programmes. Over half the leaflets (63/106) made no mention of whether or not these conditions could be prevented or the difficulties of prevention. Of the 43/106 leaflets that did mention prevention or difficulties of prevention of the conditions, most (39/43) mentioned whether or not the disorders were inherited. A few mentioned preconception and/or antenatal screening or reproductive choice (7/43) as methods of prevention, provided other information about prevention programmes (3/43) or indicated that, apart from reproductive choice, no prevention programmes existed (1/43).

Table 1.5: Does the leaflet discuss available prevention programmes and/or the difficulties of prevention?*

Prevention/difficulties of prevention	Number
Yes, Indicates whether or not disorders are inherited	39
Yes, Mentions preconception and/or antenatal screening and reproductive choice	7
Yes, Provides other information about prevention programmes	3
Yes, Specifies that apart from reproductive choice, no prevention programmes exist	1
No	63
Number of leaflets	106

* Some documents appear in more than one category

3.3 Process of screening

We examined in detail the amount of information provided in each leaflet about the actual screening process. Given our initial selection criteria to include only leaflets that described some aspect of the screening test, most of the leaflets described how the blood sample was taken from the heel (93/106). Less than a

fifth, however, indicated that this might be uncomfortable for the baby (19/106); and few suggested how the pain might be eased (5/109). Leaflets that mentioned discomfort for the baby were produced in the UK for parents: 7/19 since 2000, though the rest were undated. Statements about discomfort tended to minimise the significance of the baby's pain in relation to the potentially significant health benefits of screening. For example, they contained statements such as:

It will only cause a moment of discomfort, which your baby will soon forget.
The heel-prick mark will disappear in a few days [195];

and

Most babies cry a bit when the heel is pricked but the discomfort is only for a moment and the possible benefits are for a lifetime [194].

Over half of the leaflets (57/106) explained how the blood spots were placed on the Guthrie card. Seventy-two leaflets described what happened next to the blood sample, and 76 of the 106 leaflets discussed the possibility of a repeated screening test. About half (54/106), described how results were reported to health professionals and then made available to parents. Few leaflets discussed the storage or later use of the blood spots (3/106). Thirteen leaflets addressed other issues related to performing the heel-prick test, such as the existence of registers and the option to opt out of research on blood spots (1); the risks associated with puncturing the heel (1); protocols for health professionals to follow (5); anonymous HIV testing (2) quality assurance mechanisms (1); repeat blood tests to rule out thalassaemia in older children (1); genetic tests for cystic fibrosis (1); and testing parents in cases of suspected haemoglobinopathy (1).

Table 1.6: Does the leaflet describe the procedures for doing the heel-prick test?*

Information about heel-prick test	Number
Yes, Describes how the blood sample is taken	93
Yes, Indicates that it may be uncomfortable for the baby	19
Yes, Describes how pain may be eased for baby	5
Yes, Describes how blood spots placed on the Guthrie card	57
Yes, Describes what happens next to the card/blood sample	72
Yes, Describes how results are reported	54
Yes, Discusses the possibility of a repeated test	76
Yes, Indicates storage/possible later use of cards	3
Yes, Other (please specify)	13
No	3
Total number of leaflets	106

* Some documents appear in more than one category

3.4 Consequences of screening

Screening programmes have immediate consequences in terms of offering (or withholding) choice to parents, reporting results of screening, and the costs of screening.

More than half of the leaflets (61/106) gave no information about when parents would receive or be asked to collect screening results. Of the 45 that indicated when parents would receive results, 17 specified this for both positive and negative results.

With the increased focus on informed choice for treatments and screening, we were interested to find out whether leaflets indicated that parents could choose whether or not their baby was screened. The situation differs in the USA where newborn screening is mandated by law. In this context, we were interested in how US leaflets addressed the issue that screening is compulsory. Our analysis showed that almost two-thirds of the leaflets (68/106) did not state directly whether or not screening was compulsory. Many leaflets implied, however, that screening all babies took place as a matter of course. Illustrating this point, one US leaflet states:

While your newborn baby is in the hospital or shortly afterward, he or she will have a blood sample taken to screen for certain birth defects [36].

Similarly, a UK leaflet begins:

The heel prick test is carried out on all babies born in the UK at 6 days of age. [303]

Leaflets specifying that screening was not obligatory (25/106) provided varied information around the concept of 'choice'. Some leaflets presented screening as a choice but also emphasised the importance of screening by recommending it (10) or outlining the implications of not screening (4). For example, one UK leaflet for parents stipulates that:

All babies born in Scotland are eligible.... It is important that you realise that a delayed diagnosis of any of the conditions below may lead to permanent damage to your baby" [268].

Similarly, another UK leaflet informs parents that

In this country the newborn screening test is not a legal requirement (unless a baby is up for adoption) but it is very important, since a delayed diagnosis may cause your baby permanent damage" [194].

Twelve leaflets that indicated that screening was not compulsory outlined the processes of accepting or refusing screening for a baby. Some of these leaflets indicated that refusal might have negative consequences for newborns. For example, an Australian leaflet informed that: "Parents may refuse the newborn screening test on behalf of their baby. However the programme diagnoses 70-80 babies each year for whom treatment is urgently needed, and refusal of the test might unnecessarily risk the baby's health" [43]. Two UK leaflets took a more neutral position: one leaflet for parents commented that "If you do not want these screening tests to be undertaken your wishes will be respected" [107]; and

another, for health professionals, merely outlined the recording protocol for refusals. Three of these 12 leaflets stated that screening was optional only in the context of pilot screening programmes in the USA.

Understandably, because of its legal position, all the leaflets/sheets stating that newborn blood spot screening was mandatory (16/106) were from the USA. Six of these leaflets stipulated the reasons for refusal of the tests. One leaflet informed parents that:

In Massachusetts you may refuse for religious reasons. If you do so, you may be asked to sign a refusal form. This form relieves your doctor of any liability for damages that result from a disorder that could have been detected by screening [157].

Another US leaflet indicated that people could also refuse:

in many states, because of their personal beliefs [12].

Neither leaflet explains what constitutes 'religious beliefs' or 'personal beliefs'.

About half of the leaflets (55/106) discussed the limitations or possible harms of screening, which meant that a significant proportion of the leaflets did not address these issues. Over half of the leaflets written for parents (43/79), and half of those intended health professionals (8/19) contained this information, including the risk of false-negative (19/55), or false-positive (22/55) results. Forty-one of the 55 leaflets that addressed the limitations of screening discussed other possible harms such as parental anxiety associated with the need for repeated tests because of insufficient blood or unclear results.

The issue of the costs of screening was not addressed in most of the leaflets (92/106). Those including information on costs were almost all from the USA, and mentioned costs to parents (12/14), health care services (7/14) or health insurers (5/14). Very few leaflets addressed the issue of the cost-effectiveness of screening (5/106).

3.5 Follow-up to screening

The need for further testing to confirm a diagnosis after a positive screening result was mentioned in the majority of the leaflets (71/106). Almost all of these leaflets (69/71) identified the circumstances in which follow-up would occur. For example, one UK leaflet states that: "In the unlikely event that a test is abnormal there will be no delay in arranging through your family doctor for your baby to be seen by a paediatrician" [106]. Few leaflets, however, contained any information about when parents would hear about the necessity for follow-up tests (9/106), when such tests might occur (12/106), and only one indicated when parents would receive the results of any follow-up tests.

Treatment for any of the four conditions screened for was mentioned in most of the leaflets (73/106). Almost all leaflets that mentioned treatment for PKU also mentioned the natural history of the condition (52/59). Similarly, 53 of the 58 leaflets that mentioned treatment for CHT and 16 of the 18 leaflets mentioning treatment for sickle cell disorders also discussed the natural history of the

condition. All of the leaflets mentioning treatment for CF (15/15) also discussed the natural history of the condition.

Sixty-six of the leaflets referred to some form of service related to newborn screening. This was primarily to health professionals or specialists (48/66). Other related services indicated in leaflets were support organisations within the health service or charities (23/66), counselling, including genetic counselling (16/66) and other services such as supplementary screening programmes (16/66).

3.6 Information about Carriers

Almost half of the leaflets (49/106) referred to carriers. About half of these (23/49) indicated that, as an outcome of screening, babies may be identified as carriers of cystic fibrosis (10), sickle cell disorders (9), PKU (2) or other disorders (3). Twelve leaflets mentioned that babies' DNA might be tested, and 19 mentioned the implications of carrier status for babies' health. Almost half of the leaflets mentioning carriers (24/49) indicated that parents might be carriers of sickle cell disorders (10), PKU (9), cystic fibrosis (8) or other disorders (4). The following information relating to carrier status also appeared in some of the leaflets: the implications of being a carrier for family planning and reproductive choice (19); wider carrier testing for family members (10); uncertainty regarding diagnosis (8); the number of carriers in the population (8); the psychological implications (e.g. anxiety) (6); not all mutations are tested for (4); and the number of mutations being tested (3).

3.7 Production of the leaflet

Over four-fifths of the leaflets (89/106) contained no sources of evidence for the information provided. Sources of evidence we were looking for included reference to policy documents, research papers or the names of specialist reviewers. Seventeen of the leaflets cited any of these types of evidence. About half of these were produced in the USA, primarily as public or health professional educational materials (8/16). 9/16 leaflets were produced in the UK, and included: health professional guidelines (4), a leaflet for parents including technical details of screening for CF (1) leaflets for parents whose babies have received a positive screening result (2), or needed a repeat test (1), a commercially-sponsored leaflet on newborn screening (1), and a booklet on thyroid disorders (1). Only four of the 17 leaflets gave dates for the evidence provided. One of these was a summary for health professionals produced in the UK, appearing on a website and providing a list of additional reading resources and a glossary of terms related to screening, rather than a list of references per se. Six of the 17 leaflets providing evidence indicated using more than one type of source in its development. This included one information resource produced by an on-line medical encyclopaedia [209].

Two of the 106 leaflets appraised gave information linked to the evidence. One North American leaflet reported case studies of babies whose conditions were not picked up by routine newborn screening but might have done had their parents chosen supplemental screening for their baby. The other leaflet referred to UK Department of Health guidelines for good practice.

Over half (56/106) of the leaflets indicated the date when it was developed and/or updated, but the great majority (90/106) did not indicate how the information was developed. All but one leaflet providing this information (16/106) indicated that the leaflet was developed through clinical specialist knowledge or collaboration between specialists and committees of experts. One UK leaflet stated that the leaflet was produced taking into account the views of parents.

In terms of the complexity of the leaflets and the ease of reading them, over half (56/106) were appraised as easy to read, with technical terms explained or stated in lay language. About a third (36/106) included technical terms that were unexplained or confusing, and 14 required expert or medical knowledge to understand. Forty-nine of the 79 leaflets designed for parents were easy to read, with many carefully explaining technical terms in lay language. Four of the 19 leaflets for health professionals were easy to read. Of the 14/106 leaflets that required medical expertise to understand, 10 were for health professionals, two appeared to be for parents/carers, and for two the target audience was unclear. Two parent leaflets were appraised as requiring expert knowledge to understand. For example, one of these leaflets explained that:

Expanded newborn screening using tandem mass spectrometry (TMS) commences in 2002. TMS detects changed levels of metabolites in the blood spot, which may indicate disorders of metabolism of fatty acids and amino acids [41].

4. RESULTS: APPLYING STANDARDS FOR PERSONAL DECISIONS

Outline of Chapter

This chapter considers the challenge of encapsulating standards for evidence-based treatment choice in information for screening. It reflects on the process of interpreting and applying appraisal criteria, and discusses how well screening information appears to reflect the principles underpinning the DISCERN instrument that address the reliability of the publication to inform decisions (is the purpose clear, the information relevant and up-to-date, and is how it was prepared reported?) and the quality of information provided (balanced and unbiased, acknowledging uncertainty, referring to sources of additional information, support and research evidence).

Key findings

Interpreting evidence-informed treatment choice for patients for screening decisions:

- *Translating DISCERN criteria into a tool for discriminating information relatively more able to support evidence-informed parent choice required us to be generous in making judgements*

Most leaflets reflected the following principles expressed by DISCERN:

- *The aims are clear, including what the publication is about and who is the intended audience*
- *The publication is relevant to the intended audience*
- *The information describes why babies are screened and the importance of early detection*

Less than half of the leaflets reflected the following principles expressed by DISCERN:

- *Sources and dates of evidence are given for the information provided*
- *Additional sources of support and information appear in the publication*
- *The importance of screening a specific population is mentioned*
- *The information describes the limitations and uncertainties of screening as well as the benefits*
- *Screening is presented as a choice or mandatory; alternatives to screening to screening are described*
- *Information about support for shared decision-making is provided.*

4.1 Standards for personal decisions

4.1.1 Applying DISCERN criteria

The DISCERN criteria address the reliability of the publication to inform decisions and the quality of information provided. The instrument asks: is the purpose clear, the information relevant and up-to-date, and is how it was prepared reported? It asks is the information provided balanced and unbiased; does it acknowledge uncertainty, and refer to sources of additional information, support and research evidence?

The details for how we did this is recorded in appendix 5 where we match the DISCERN criteria to acceptable answers from our tool that would indicate meeting those criteria. In order to discriminate between screening information that might provide any evidence to support parent choice, and screening information that did not, we applied a low threshold when applying these criteria. For instance, in considering whether the information was balanced and unbiased, we accepted mere mention of false positive AND false negative results OR other harms, such as the anxiety that parents may experience when newborns undergo blood spot screening. We did not appraise the quality of the explanations of these concepts. For this reason, our analysis of how well screening leaflets meet DISCERN criteria for quality of patient information must be considered to be a description of minimum requirements rather than what might be accepted as good practice.

4.1.2 Reliability of the publication

To support informed choices about screening, leaflets need to be explicit in their purpose of informing parents prior to screening. We found that 58/79 (73%) of the leaflets for parents were clear about when the information should be provided.

We judged whether leaflets were explicitly relevant to parents facing screening if they contained information about the importance of newborn screening generally and for a specific population. We found that 91/106 of the leaflets provided information on the importance of newborn screening generally **or** the importance of screening a particular population, but only 21/106 (20%) provided information on both. Eleven of the latter were from the UK, nine from the USA and one from Australia.

Only four of the 106 leaflets provided evidence to support the information provided and the dates of the publication of this evidence. Three were from the US and one from the UK. Two were public health education information sheets and two were fact sheets or reference materials for health professionals.

4.1.3 Quality of the information

We judged that leaflets were balanced, unbiased and open about areas of uncertainty if they mentioned some of the limitations or uncertainties of screening, such as the existence of false-positive or false-negative results, or other possible harms, such as anxiety for parents (although this was often expressed as exhortations not to worry). Over a half of the leaflets (55/106) discussed the

limitations or uncertainties of screening. However, only 27/106 specifically referred to false results, whilst 41/106 referred to other possible harms

Additional sources of support or information are provided in 23/106 of the leaflets. They referred to charitable or health service organisations providing support and further information.

We judged information on the natural history of the conditions screened for and information about the importance of early detection through screening as an indication of the effectiveness of screening. About two-thirds of the leaflets (70/106) contained this information, although the amount of information about the condition itself varied depending on whether it was a general leaflet outlining several conditions or was condition-specific. For example, a general leaflet about screening includes the following information about congenital hypothyroidism:

It is caused by an absent, small or improperly functioning thyroid gland. Lack of thyroid hormone slows brain development and growth. Early treatment with daily thyroid hormone leads to normal mental and physical development [43].

In contrast, a leaflet [175] devoted entirely to congenital hypothyroidism describes in more detail, the condition, its causes, and how it is determined and treated.

We found that 88/106 leaflets mentioned the benefits of screening generally; and 24/106 mentioned the benefits to a particular population (often providing information about the incidence of each condition in the population). Only a fifth 21/106 of the leaflets, however, mentioned the benefits of screening generally *and* within a particular population. A recent Scottish leaflet describes the benefits of screening as follows:

Happily, most babies are perfectly healthy when they are born. A small number however are born each year with problems. This is why all babies are examined carefully after birth so that any problem identified in this way can be assessed and treated as soon as possible. Some problems of body chemistry will not show up on the head to toe check but can be detected through a blood test...It is important that you realise that a delayed diagnosis of any of the conditions below may lead to permanent damage to your baby. [268]

The leaflet then goes on to provide information on each of the conditions screened for. For cystic fibrosis it says:

Cystic fibrosis is a serious inherited condition that affects 1 in every 2,500 babies born in Scotland. The organs that are the most severely affected are the pancreas and the lungs, causing poor digestion and chest infections.

Early treatment may help affected children to maintain good nutrition and minimise chest infections, leading to improved quality of life. The majority of affected babies will be picked up by the screening test [268].

Less than a quarter of the leaflets contained information about the limitations or possible harms of screening in terms of the risk of false negative (19/106) or false positive results (22/106). Most of the leaflets therefore did not meet DISCERN's

criteria for providing information about the risks. One UK information sheet includes the following information about false results:

For most tests, some people with a positive result will turn out not to have the disorder ('false positive') and some with a negative result will subsequently be shown to have the disorder ('false negative'). Those people who have a positive result on the screening test are usually offered a further test ('diagnostic test'), which will pick out much more accurately those who have the disorder [186].

The DISCERN tool asks whether the leaflet describes what would happen if no treatment were given. To answer this question, we assessed leaflets according to whether they described the risks of not screening. Analysis showed that 91 of the 106 leaflets explained the importance of early detection and treatment for their child's health. However, only four leaflets specifically outlined the implications of not screening. For example, one UK leaflet comments that "[screening] is very important since delayed diagnosis may cause your baby permanent brain damage" [194]. Similarly, a US leaflet warns parents that:

A newborn baby can look healthy, but have a serious disease that cannot be seen. If not treated, these diseases can lead to slow growth, blindness, brain damage, or possibly death. Early treatment can help prevent these serious problems. [35]

The DISCERN tool indicates that health information should ideally contain information about treatment choices. We were therefore interested in whether newborn screening leaflets mentioned that screening was a choice or was mandatory. We found that 41/106 leaflets contained this information (39%) and therefore met this criterion.

Finally, the DISCERN tool asks whether the leaflet provides support for shared decision-making, such as questions to ask a health professional, or suggesting who to approach to discuss screening. Our analysis showed that no leaflets we appraised addressed this issue.

5. RESULTS: APPLYING STANDARDS FOR POLICY DECISIONS

Outline of Chapter

This chapter considers the challenge of encapsulating criteria to inform population level decisions about screening to personal/ family level decisions about screening. It reflects on the process of interpreting and applying criteria for implementing screening programmes, and discusses how well screening information appears to reflect the principles underpinning the work of the National Screening Committee, with its explicit consideration of the viability, effectiveness and appropriateness of screening programmes.

Key findings

Interpreting policy making principles for personal/ family decisions

- *NSC criteria for policy making are specific and demanding, and have changed over the years. Translating them into criteria to judge information to be used for parents' decisions was very challenging.*

Most leaflets included information about screening corresponding to the following principles of the NSC:

- *The screened condition should be an important health problem*
- *The natural history of the condition should be adequately understood*
- *There should be an agreed policy for follow-up of positive screening results*
- *Adequate staffing and facilities for testing, diagnosis and treatment should be available*

Fewer than half the leaflets included information relating the following principles of the NSC:

- *All available prevention programmes should have been implemented as far as possible*
- *Clinical management should be optimised prior to screening*
- *The benefits of the screening programme outweighs the physical and possible psychological harm caused by screening*
- *If carriers of a mutation are identified as a result of screening, the natural history of people with this status should be understood, including the psychological implications*

Approximately 1 in 10 or fewer included information relating the following principles of the NSC:

- *There exists a safe, precise and validated screening test*
- *Effective early treatment is available, with evidence that early detection and treatment leads to better outcomes*

- *There is an agreed evidence-based policy about who should be offered treatment*
- *Evidence exists from high-quality RCTs that the programme is effective in reducing mortality and morbidity; evidence also exists that the test accurately measures risk; or the information provided should be of value and readily understood*
- *Opportunity costs should be balanced with other health care costs*
- *All other options should have been considered to ensure no more cost-effective intervention could be introduced*
- *There must be a plan for managing and monitoring screening and an agreed set of quality assurance standards*
- *Evidence-based information for parents should be available explaining the consequences of testing, investigation and treatment.*
- *If the test is for mutations the criteria used to select the subset of mutations to be covered by screening, if all possible mutations are not being tested, should be clearly set out*

5.1 Standards for policy decisions

5.1.1 Applying NSC criteria

We sought to determine the extent to which leaflets addressed the issues raised in the criteria set out by the UK National Screening Committee for appraising the viability, effectiveness and appropriateness of a screening programme.⁽⁶⁾ The details of this work are presented in appendix 6. We included questions in our appraisal tool relating to the NSC's criteria because of their specific focus on screening, despite their being related to policy decisions about implementing a particular health service rather than the provision of health information to consumers.

A changing balance in policy between supporting public health priorities and encouraging realistic public expectations or informed choice for screening is evident since the National Screening Committee for England (NSC) published its first criteria for appraising the viability, effectiveness and appropriateness of a screening programme.⁽¹⁾ The NSC's additional criteria in the second report are included to assist people further in making informed choices about screening.⁽²⁾ Criteria added in a more recent report of the NSC's Director relate specifically to the identification of unaffected carriers of the conditions and genetic testing.⁽⁶⁾

Whilst the DISCERN criteria for information for patients on treatment choices required some adaptation when developing our own appraisal tool, there was even greater leap when developing our tool to answer questions relating to the NSC's stringent criteria for the implementation of screening programmes. We did not include all NSC criteria in our leaflet appraisal tool because such criteria did not appear to be appropriate in the context of information leaflets or difficult to measure (e.g. evidence that the complete screening programme is clinically, socially and ethically acceptable to health professionals and the public; and anticipation of increased pressure for more screening), and were generous in our

evaluations of whether or not the information materials appraised conformed with the NSC criteria for implementing a screening programme.

According to the NSC, ideally all the following criteria should be met before screening for a condition is initiated:

5.1.2 The condition

One of the National Screening Committee criteria is that the condition to be screened for should be an important health problem. Analysis of our survey data showed that 91/106 leaflets (86%) provided information on the importance of screening newborns for particular conditions. The Committee also maintains that the natural history of the condition should be adequately understood and that there should be a clear means of identifying the condition in an early asymptomatic phase. Seventy of 106 leaflets (66%) contained information about the natural history of at least one of the four conditions, and the importance of screening.

Another NSC criterion is that all available prevention programmes should have been implemented as far as possible. To address this issue, we included questions in our appraisal tool about whether leaflets specified that apart from reproductive choice no prevention programmes exist for these conditions or provided other information about prevention programmes. Most leaflets did not mention prevention programmes or reproductive choice as a means of prevention (64/106). Of the 42 (40%) that did, 39/42 leaflets provided information on whether or not the conditions were inherited. Seven leaflets mentioned preconception and/or antenatal screening and reproductive choice. One leaflet specified that apart from reproductive choice no prevention programmes exist. Three leaflets provided other information about prevention programmes. For example, one information sheet published by a US children's hospital claims that:

Four methods have worked to decrease the number of deaths due to birth defects by half since 1960: taking 400 micrograms of the B-vitamin folic acid prior to and in the early weeks of pregnancy; new corrective surgical procedures; the specialized care and advanced technology of neonatal intensive care units; and new tests (screening) to detect and treat these defects at birth [33].

The NSC stipulates that if carriers of a mutation are identified as a result of screening the natural history of people with this status should be understood, including the psychological implications. About half of the leaflets in this survey mentioned that babies might be identified as carriers (23/49) or that parents might be carriers (24/49). Nineteen leaflets mentioned the implications of carrier status for babies' health. Other leaflets mentioned the wider implications for families, including implications for family planning/reproductive choice (19/49), wider carrier testing for family members (10), how many carriers there are in the population (8), any uncertainty regarding diagnosis (8) and the psychological implications, such as anxiety (6).

5.1.3 The test

We judged descriptions about how the blood was taken, the possibility of repeated tests, and information about false-positive and false-negative results as indicators of a safe, precise and validated screening test. Nine of the 106 leaflets (8%), all from the United States, contained this information. With regard to NSC criteria that the distribution of test values in the target population should be known and a suitable cut-off level defined and agreed, our analysis showed that only five leaflets (5%) met these criteria, again from the USA, by providing information on the importance of screening a particular population for the conditions, the possibilities of false-negative and false-positive results and of a repeated test.

Seventy-one (67%) of the leaflets described the circumstances in which follow-up would occur, indicated when the parents would hear about the necessity for further tests, stated when follow-up tests would occur, or indicated when parents would receive results of follow-up tests. We determined that two-thirds of the leaflets therefore met the NSC criteria that there should be an agreed policy for follow-up for positive screening results (i.e. thought to be affected by the condition), and any further choices that might need to be made.

The NSC stipulates that if the test is for mutations the criteria used to select the subset of mutations to be covered, if not all mutations are being tested, should be clearly set out. We found that almost half of the leaflets mentioned carriers (49/106). However less than half of these mentioned that babies could be identified as carriers (23/49), and only 12 of these mentioned that babies' DNA might be tested. Few leaflets mentioned the number of mutations tested (3) or that not all mutations were tested (4).

5.1.4 The treatment

The specification that there should be an effective early treatment for patients, with evidence that early detection and treatment leads to better outcomes was met by only 9/106 leaflets primarily because few cited evidence. Similarly, only 11/106 leaflets included information related to an agreed evidence-based policy about who should be offered treatment.

Thirty-two leaflets (30%) provided information relating to NSC criteria that clinical management is optimised prior to implementing screening, in that they mentioned treatment for at least one of the four conditions and cited particular health professionals or specialists (e.g. paediatricians or metabolic specialists) who might become involved if treatment were indicated.

5.1.5 The screening programme

Thirteen leaflets (12%) met the criterion that there should be evidence from high quality randomised controlled trials (RCTs) that the screening programme is effective in reducing mortality and morbidity, that there should be evidence that the test accurately measures risk, or that information is provided about the test and its outcome must be of value and readily understood. In this instance, we appraised leaflets according to whether they provided evidence and explained why the screening tests are important, or explained the importance of screening a particular population, provided evidence, and were easy to read/understand.

Less than half of the leaflets (47/106) provided information about the advantages and limitations of screening, including explaining why screening was recommended/important and information about false-negatives, false-positives and any other limitations.

Another of the NSC's criteria is that the opportunity cost of the screening programme should be economically balanced in relation to other medical costs. To address this issue we looked for information in leaflets pertaining to the cost-effectiveness of a newborn screening programme, and found this information in five leaflets. Four of these leaflets were from the USA; the one UK information sheet referred to the cost-effectiveness of screening in the context of raising funds for the purchase of purchasing new equipment for screening laboratories [207].

Our analysis showed that only four leaflets contained information that addressed the NSC's criteria that there must be a plan for managing and monitoring screening and an agreed set of quality assurance standards. Three of the leaflets were from the USA, and one from Scotland. A comprehensive US overview for parents, states:

The Program has numerous quality improvement, educational and monitoring mechanisms in place to assure that all infants are screened and that the results are valid. However, biological variability, transfusions, and human error can result in missed cases [18].

In the case of meeting NSC criteria that adequate staffing and facilities for testing, diagnosis and treatment were concerned, we found that 97/106 (92%) of leaflets either mentioned follow-up to testing for diagnosis of a condition, or mentioned treatment for at least one of the four conditions. To determine whether the leaflets fulfilled NSC criteria that all other options should have been considered to ensure no more cost-effective intervention could be introduced, we included questions in our appraisal tool relating to information about prevention programmes or the difficulties of preventing of the conditions. Analysis of the leaflets showed that 42/106 (40%) of the leaflets specified either that no prevention programme was available, mentioned whether or not the conditions were inherited, preconception and/or antenatal screening and reproductive choice, or provided information about other means of prevention. For example a UK information sheet for parents whose baby needs a repeat test for cystic fibrosis states that:

Cystic fibrosis is an inherited disease....Approximately 1:25 of the population are carriers of the faulty gene which causes CF in their children [263].

Another UK leaflet on congenital hypothyroidism contains information on the causes of different types of hypothyroidism and whether or not they are inherited. Describing one form that is not inherited, and the cause unknown, the leaflet explains:

Very early in your baby's development in your uterus, their thyroid gland moves from the back of the tongue to its normal position in the neck. In some babies this does not happen, which means that the gland cannot work properly... If you have one child with this type of congenital hypothyroidism, the chance of having another baby who is affected is very low [49].

The NSC criteria stipulate that evidence-based information, explaining the consequences of testing and treatment should be made available to potential participants to help them make an informed choice. To meet these criteria we determined that leaflets for parents should contain information on: the importance of screening for the four conditions; the importance of screening this population; the possibilities of receiving false-negative or false-positive results or incurring other possible harms; identify the circumstances in which follow-up would occur; treatment for at least one of the four conditions; and provide sources of evidence. Only two information sheets met all these criteria (2%). One was a public health education information sheet on newborn screening produced in the USA [137], the other was fact sheets produced for health professionals in the United States [204].

6. DISCUSSION AND CONCLUSIONS

Outline of Chapter

This chapter summarises and discusses the findings of the survey in terms of its methodological strengths and weaknesses; how it compares with other surveys of evidence-based patient information; and how the leaflets found match information needs identified in a qualitative needs assessment. It ends with our conclusions about the next steps required to prepare evidence-based parent information about newborn blood spot screening.

Key messages

Summary of findings

- *Most leaflets met at least basic criteria for providing information on the purpose of the leaflet, the aims of screening, conditions screened for, screening procedure, need for follow-up to confirm positive screening results, and treatment for the conditions.*
- *Whilst most leaflets mentioned the benefits of screening, few addressed the issue of the possible harms, indicated that screening was a choice, discussed alternatives to screening, or gave information about when and how parents would receive results.*
- *Few leaflets provided any sources of evidence for the information provided or how the information was developed.*
- *The leaflets/information sheets did better on issues rooted in the principles of DISCERN for the provision of information on treatment choices, than on principles underpinning NSC decisions for the implementation of screening programmes.*

Strengths and weaknesses of the study

- *The 106 leaflets included in the survey were sourced through the internet and a selection of health service and support organisations in the UK; our searching strategy, however, was not exhaustive.*
- *Two researchers performed double data extraction to ensure reliability of the appraisal tool, which, unlike the DISCERN instrument is not a validated tool.*

Relating to other surveys of patient information

- *Like other surveys of patient information, this study has found that information for parents is variable in quality and length and often biased in favour of screening.*

Matching the information needs identified in a qualitative needs assessment

- *Most leaflets met the criteria for providing 'basic' information and therefore met the needs of parents, most of whom perceived screening as routine for babies.*

Recommendations

- *We recommend the use of DISCERN with topic specific guidelines as a starting point for developing patient information.*
- *We recommend appraising patient information with a topic specific tool to access good practice in information provision.*

6.1 Summary of findings

Most leaflets provided information on the purpose of the leaflet, the aims of screening, conditions screened for, the screening procedure, the need for follow-up to confirm positive screening results, and treatment for the conditions. Even though the majority of leaflets might have met these criteria, however, the appraisers were generous when using the tool, and in some cases the information provided in the leaflets was scant and barely met the requirement. In many cases, the majority of the leaflets did not meet the specific criteria. For example, whilst most leaflets mentioned the benefits of screening, few addressed the issue of the possible harms, particularly false results, indicated that screening was a choice, discussed alternatives to screening, or gave information about when and how parents would receive results. Fewer still provided any sources of evidence for the information provided or how the information was developed.

The leaflets/information sheets did better on issues rooted in DISCERN criteria for the provision of information on treatment choices, than on NSC criteria for the implementation of a screening programme. This is perhaps not surprising, considering that the former relates to patient information and the latter to screening policy criteria. It also reflects previous policy and practice when newborn screening was performed as a routine test, and informed consent not sought from parents, who were told that ‘no news is good news’.

6.2 Strengths and weaknesses of the study

The study involved carrying out a wide search for information resources through the internet, and through a selection of health service and support organisations throughout the UK. Our searching strategy, however, was not exhaustive. Rather, we sought information that was accessible to UK maternity service users. To gather the full range of materials we purposely sought materials through UK maternity services and English language web sites of leading children’s hospitals.

In developing our tool, we drew on both NSC criteria, which are topic specific and policy focussed, and DISCERN criteria, which are broader and more focussed on the patient/individual. The researchers were generous in applying the criteria. Some leaflets were judged to have met the criteria even if the information provided was extremely brief.

In developing and using the tool, two researchers performed double data extraction independently and compared results. This provided a quality check and highlighted issues for discussion.

Some contradictions were evident in categorising information as either presenting the benefits or difficulties/harms of screening. For example, we have listed the possible need for repeat testing as a limitation of screening. This is because repeat testing can cause worry for parents, and highlights some of the uncertainties of screening. However, repeat testing could also be considered a benefit, in that it provides evidence of follow-up procedures to ensure that screening is done correctly and all affected babies detected.

Nevertheless, the appraisal tool was able to distinguish parent information in terms of the type and style of information provided, and to indicate its reliability in terms of how and when it had been produced.

6.3 Other surveys about patient information

6.3.1 Surveys of screening and diagnosis information

Other studies that have examined and evaluated patient information about screening have also concluded that the risks, limitations or difficulties were not openly discussed. An Australian survey evaluating 58 leaflets on screening for breast cancer found that the benefits of screening were reported only ever as relative risk reduction and never as absolute risk reduction, thereby exaggerating the benefit.⁴

Another study of 27 websites compared information on the possible benefits and harms of screening for breast cancer provided by different groups⁵. The authors concluded that information provided by professional advocacy groups and government organisations were biased in favour of screening. In contrast, information on consumer websites was more balanced and comprehensive.

Other small qualitative studies have been carried out to evaluate existing patient information. One such study of information on gastroscopy procedures in seven different hospitals, found that the quality of information varied between units, most leaflets lacked vital information, or included information that was confusing or ambiguous, and that information about risks was included in only one leaflet⁶. An audit of informed consent procedures prior to surgical procedures found that the poorest area of information-giving was related to the potential complications of various procedures⁷.

6.3.2 Surveys employing DISCERN

The Royal College of Anaesthetists (RCA) has also evaluated patient information materials with the specific aim of developing new information resources^{8,9}. Like us, they carried out an in-depth review of patient information with a view to developing new, better-quality patient information. In the process of evaluating the leaflets, the RCA also developed an appraisal tool based on the DISCERN tool for evaluating patient information, as well as other tools for evaluating health information⁸.

Whilst our survey relied on adapting the original DISCERN instrument,³ and combining it with very focused topic specific criteria, since we completed our work, the DISCERN team has developed another instrument, DISCERN for

genetic testing, which would be applicable to some, but not all, of the conditions screened by the heel prick test.

6.4 How do leaflets match parents' information needs?

A qualitative study we carried out simultaneously with this survey identified parents' information needs. It highlighted the need for brief, non-technical information for parents, who largely perceived newborn blood spot screening as a routine procedure for all babies, and wanted only 'basic' information.¹¹ Whilst many parents were not specific about what this might include, some said they would like to know the reasons for screening, the conditions they are screened for, when parents will receive the results, and what would happen if a baby tested positive for one of the conditions. Most of the leaflets in this survey provided this type of 'basic' information. For example, almost all of the leaflets (91/106) explained the aims of screening and three-quarters of them gave some description of the screened conditions (79/106). The great majority of leaflets described the procedures for carry out the screening test. Less than half, however indicated when parents would receive screening results, which reflects a finding of the qualitative study that most parents were told that the screening test was nothing to worry about, and that "no news was good news".

Most leaflets also mentioned the need for follow-up tests to check abnormal results (71/106), and most also mentioned treatment for at least one of the screened conditions (73/106). Just over half of the leaflets were appraised as 'easy to read' (56/106), which indicated that a significant number of leaflets did not meet parents' requested criteria of 'basic' easily understood information.

That two-thirds of the leaflets did not indicate whether or not screening was compulsory, and implied that screening was routine, supported the finding of our qualitative study that parents perceived screening as not subject to choice. Results from our qualitative study showed that many parents and health professionals supported screening as a routine procedure and expressed concerns that an 'informed choice' model would lead to a lower screening uptake.

Whilst the model of informed choice promulgated by the National Screening Committee indicates the need to provide parents with more detailed information about the limitations as well as the benefits of screening, parents were generally not in favour of receiving detailed information about the limitations of screening. Many parents and health professionals regarded this as potentially reducing uptake of screening, which both, and particularly health professionals and parents of affected children regarded as undesirable. Few parents told us they were interested in knowing the sources of information contained in a leaflet. This is matched by our finding that the great majority of the leaflets provided no source of evidence for the information given (89/106). Those that did (17/106) were generally intended for health professionals.

It should be noted that the research evidence is in favour of screening either to improve health outcomes through early treatment, or to improve parents' experiences of the diagnostic period¹¹⁻¹⁵, and that informed choice for screening is a relatively new phenomenon. It is not surprising, therefore, that the leaflets

favour screening, and many do not support informed choice. With or without informed choice, in an increasingly legalistic climate there may be increased pressure to provide patients with detailed information to facilitate realistic expectations. The question also arises whether in the current climate of informed choice and with increasing patient information available some parents may begin to demand more information about screening⁷.

6.5 What has this survey achieved?

This survey of currently available information on newborn blood spot screening forms part of the work we have undertaken to inform the development of parent and health professional information resources. The other streams of work have included a systematic review of the literature on newborn screening and communication about it,³ an information needs assessment,¹¹ and convening an expert advisory group to help develop information resources.

This survey provided a list of topics, adapted from the appraisal tool, for members of the expert advisory group to debate. Structured discussion of these topics, around a table at its first meeting, and subsequently by email, led to a template for a pre-screening parent information leaflet. Inspection of the leaflets addressing topics within this template provided examples of wording, brief and extensive, for the expert advisory group to discuss at its second meeting. This discussion informed the first draft of a leaflet. A subsequent consultation of members and parents and health professionals interviewed in the parallel qualitative needs assessment refined this draft, making it ready for piloting with parents and their midwives between May and July 2004.

Feedback on the resources was subsequently sought from parents and midwives (and other health professionals involved in taking the blood spots) through self-administered questionnaires. Feedback on these resources was also collected through the Programme Centre's consultation during the summer of 2004. Data received was analysed and the information resources modified in light of the feedback received.

Final versions of the pre-screening leaflet and communication guidelines have been available for national distribution since October 2004. The same methods have been applied to develop 'results' leaflets to be given to parents if their baby has a positive screening result (i.e. likely to be affected by one of the conditions). These provide more information about the implications of receiving a positive screening result, follow-up procedures and testing, and support services.

6.6 Conclusions and recommendations

We found combining the DISCERN criteria for patient information about treatment choices with NSC criteria for implementing screening programmes provided a practical route for structuring topics to be considered in parent information. **We recommend the use of DISCERN with topic specific guidelines as a starting point for developing patient information.**

We found appraising leaflets with a tool developed for the specific topic valuable for identifying the variation in parent information in terms of content and style. **We recommend appraising patient information with a topic specific tool to access good practice in information provision.**

REFERENCES

1. Stewart R, Oliver S (2003) *What is known about communication with parents about newborn bloodspot screening?* London, UK: Newborn Screening Programme Centre.
2. Hargreaves K, Stewart R, Oliver S (2005) Newborn screening information supports public health more than informed choice. *Health Education Journal* **64** (2): 110-119.
3. Charnock D, Shepperd S, Needham G, Gann R (1998) DISCERN: an instrument for judging the quality of written consumer health information on treatment choices. *Journal of Epidemiology & Community Health* **53**: 105–11.
4. Slaytor EK, Ward JE (1998) How risks of breast cancer and benefits of screening are communicated to women: analysis of 58 pamphlets. *British Medical Journal* **317**: 263–5.
5. Jorgensen K, Gotzsche P (2004) Presentation on websites of possible benefits and harms from screening for breast cancer: cross sectional study. *British Medical Journal* **328**: 148–61.
6. Parahoo K, Ridley T, Thompson K, Melby V, Humphreys G (2003) A qualitative evaluation of information leaflets for gastroscopy procedure. *Journal of Evaluation in Clinical Practice* **9**: 423–31.
7. Ferguson DJ (2001) Informed Consent - who knows? Results of a hospital audit project. *Health Education Journal* **60**: 371–7.
8. Lack J, Rollin A, Thoms G, White L, Williamson C (2003) *Raising the Standard: Information for Patients*. London: The Royal College of Anaesthetists.
9. Paul F, Hendry C, Cabrelli L (2004) Meeting patient and relatives' information needs upon transfer from an intensive care unit; the development and evaluation of an information booklet. *Journal of Clinical Nursing* **13**: 396–405.
10. Hargreaves K, Stewart R, Oliver S (2004) Informed choice and public health screening for children: the case of blood spot screening. *Health Expectations* **8**: 161-171.
11. Barratt A, Irwig L, Glasziou P, Cumming RG, Raffle AE, Hicks N, Gray JA, Guyatt GH. (1999) Users' guides to medical literature: XVII. How to use guidelines and recommendations about screening. Evidence-Based Medicine Working Group. *Journal of the American Medical Association* **281**: 2029–34.
12. Davies SC, Cronin E, Gill M, Greengross P, Hickman M, Normand C (2000) Screening for sickle cell disease and thalassaemia: a systematic review with supplementary research. *Health Technology Assessment* **4**: 1-76.

13. Gray M (2003) *Screening in England. NSC Programme Director's Report*. Oxford: National Screening Committee.
14. Merelle ME, Nagelkerke AF, Lees CM, Dezateux C (2002) Newborn screening for cystic fibrosis. *The Cochrane Database of Systematic Reviews*, 2002: Issue 1. <http://www.cochrane.org/>
15. Pollitt RJ, Green A, McCabe A, Booth A, Cooper NJ, Leonard JV, Nicholl J, Nicholson P, Tunaley JR, Viridi NK (1997) Neonatal screening for inborn errors of metabolism: cost, yield and outcome. *Health Technology Assessment* 1: 1–203.

Appendix 1: The DISCERN Instrument

SECTION 1. Is the publication reliable?

1. Are the aims clear?

No		Partially		Yes
1	2	3	4	5

HINT Look for a clear indication at the beginning of the publication of:

- what it is about
- what it is meant to cover (and what topics are meant to be excluded)
- who might find it useful

If the answer to Question 1 is 'No', go directly to Question 3.

2. Does it achieve its aims?

No		Partially		Yes
1	2	3	4	5

HINT Consider whether the publication provides the information it aimed to as outlined in Question 1

3. Is it relevant?

No		Partially		Yes
1	2	3	4	5

HINT Consider whether:

- the publication addresses the questions that readers might ask
- recommendations and suggestions concerning treatment choices are realistic or appropriate.

4. Is it clear what sources of information were used to compile the publication (other than the author or producer)?

No		Partially		Yes
1	2	3	4	5

HINT:

- Check whether the main claims or statements made about treatment choices are accompanied by a reference to the sources used as evidence, e.g. a research study or expert opinion.

- Look for a means of checking the sources used such as a bibliography/reference list or the addresses of the experts or organisations quoted, or external links to the online sources.

Rating note: In order to score a full '5' the publication should fulfil both hints. Lists of **additional** sources of support and information (Question 7) are not necessarily sources of **evidence** for the current publication.

5. Is it clear when the information used or reported in the publication was produced?

No		Partially		Yes
1	2	3	4	5

HINT Look for:

- dates of the main sources of information used to compile the publication
- date of any revisions of the publication (but not dates of reprinting in the case of print publications)
- date of publication (copyright date).

Rating note: The hints are placed in order of importance - in order to score a full '5' the dates relating to the first hint should be found.

6. Is it balanced and unbiased?

No		Partially		Yes
1	2	3	4	5

HINT Look for:

- a clear indication of whether the publication is written from a personal or objective point of view
- evidence that a **range** of sources of information was used to compile the publication, e.g. more than one research study or expert
- evidence of an external assessment of the publication.

Be wary if:

- the publication focuses on the advantages or disadvantages of one particular treatment choice without reference to other possible choices
- the publication relies primarily on evidence from single cases (which may not be typical of people with this condition or of responses to a particular treatment)
- the information is presented in a sensational, emotive or alarmist way.

7. Does it provide details of additional sources of support and information?

No		Partially		Yes
1	2	3	4	5

HINT Look for suggestions for further reading or for details of other organisations providing advice and information about the condition and treatment choices.

8. Does it refer to areas of uncertainty?

No		Partially		Yes
1	2	3	4	5

HINT:

- Look for discussion of the gaps in knowledge or differences in expert opinion concerning treatment choices.
- Be wary if the publication implies that a treatment choice affects everyone in the same way, e.g. 100% success rate with a particular treatment.

SECTION 2. How good is the quality of information on treatment choices?

N.B. The questions apply to the treatment (or treatments) described **in the publication**. Self-care is considered a form of treatment throughout this section.

9. Does it describe how each treatment works?

No		Partially		Yes
1	2	3	4	5

HINT Look for a description of how a treatment acts on the body to achieve its effect.

10. Does it describe the benefits of each treatment?

No		Partially		Yes
1	2	3	4	5

HINT Benefits can include controlling or getting rid of symptoms, preventing recurrence of the condition and eliminating the condition, both short-term and long-term.

11. Does it describe the risks of each treatment?

No		Partially		Yes
1	2	3	4	5

HINT Risks can include side-effects, complications and adverse reactions to treatment, both short-term and long-term.

12. Does it describe what would happen if no treatment is used?

No		Partially		Yes
1	2	3	4	5

HINT: Look for a description of the risks and benefits of postponing treatment, of watchful waiting (i.e. monitoring how the condition progresses without treatment) or of permanently forgoing treatment.

13. Does it describe how the treatment choices affect overall quality of life?

No	Partially		Yes
1	2	3	4
			5

HINT Look for:

- description of the effects of the treatment choices on day-to-day activity
- description of the effects of the treatment choices on relationships with family, friends and carers.

14. Is it clear that there may be more than one possible treatment choice

No	Partially		Yes
1	2	3	4
			5

HINT Look for:

- a description of who is most likely to benefit from each treatment choice mentioned, and under what circumstances
- suggestions of alternatives to consider or investigate further (including choices not fully described in the publication) before deciding whether to select or reject a particular treatment choice.

15. Does it provide support for shared decision-making?

No	Partially		Yes
1	2	3	4
			5

HINT Look for suggestions of things to discuss with family, friends, doctors or other health professionals concerning treatment choices.

SECTION 3. Overall Rating of the Publication

16. Based on the answers to all of the above questions, rate the overall quality of the publication as a source of information about treatment choices

Low	Moderate		High
<i>Serious or extensive shortcomings</i>	<i>Potentially important but not serious shortcomings</i>		<i>Minimal shortcomings</i>
1	2	3	4
			5

Copyright British Library and the University of Oxford 1997

Appendix 2: NSC criteria for mounting a screening programme

UK National Screening Committee

Criteria for appraising the viability, effectiveness and appropriateness of a screening programme

Ideally all the following criteria should be met before screening for a condition is initiated:

The Condition

1. The condition should be an important health problem
2. The epidemiology and natural history of the condition, including development from latent to declared disease, should be adequately understood and there should be a detectable risk factor, disease marker, latent period or early symptomatic stage
3. All the cost-effective primary prevention interventions should have been implemented as far as practicable
4. If the carriers of a mutation are identified as a result of screening the natural history of people with this status should be understood, including the psychological implications.

The Test

5. There should be a simple, safe, precise and validated screening test
6. The distribution of test values in the target population should be known and a suitable cut-off level defined and agreed
7. The test should be acceptable to the population
8. There should be an agreed policy on the further diagnostic investigation of individuals with a positive test result and on the choices available to those individuals
9. If the test is for mutations the criteria used to select the subset of mutations to be covered by screening, if all possible mutations are not being tested, should be clearly set out.

The Treatment

10. There should be an effective treatment or intervention for patients identified through early detection, with evidence of early treatment leading to better outcomes than late treatment
11. There should be agreed evidence based policies covering which individuals should be offered treatment and the appropriate treatment to be offered
12. Clinical management of the condition and patient outcomes should be optimised in all health care providers to participation in a screening programme.

The screening programme

13. There should be evidence from high quality Randomised Controlled Trials that the screening programme is effective in reducing mortality or morbidity.
Where screening is aimed solely at providing information to allow the person being screened to make an “informed choice” (e.g. Down syndrome, cystic fibrosis carrier screening), there must be evidence from high quality trials that the test accurately measures risk. The information that is provided about the test and its outcome must be of value and readily understood by the individual being screened.
14. There should be evidence that the complete screening programme (test, diagnostic procedures, treatment/ intervention) is clinically, socially and ethically acceptable to health professionals and the public.
15. The benefit from the screening programme should outweigh the physical and psychological harm (caused by the test, diagnostic procedures and treatment).
16. The opportunity cost of the screening programme (including testing, diagnosis and treatment, administration, training and quality assurance) should be economically balanced in relation to expenditure on medical care as a whole (i.e. value for money)
17. There should be a plan for managing and monitoring the screening programme and an agreed set of quality assurance standards
18. Adequate staff and facilities for testing, diagnosis, treatment and programme management should be available prior to the commencement of the screening programme.
19. All other options for managing the condition should have been considered (e.g. improving treatment, providing other services), to ensure that no more cost effective intervention could be introduced or current interventions increased within the resources available.
20. Evidence-based information, explaining the consequences of testing, investigation and treatment, should be made available to potential participants to assist them in making an informed choice.
21. Public pressure for widening the eligibility criteria for reducing the screening interval, and for increasing the sensitivity of the testing process, should be anticipated. Decisions about these parameters should be scientifically justifiable to the public.
22. If screening is for a mutation the programme should be acceptable to people identified as carriers and to other family members.

Appendix 3: Bringing together DISCERN and NSC criteria to develop our appraisal tool

This table illustrates how we brought together the DISCERN and NSC criteria, grouped in the following themes:

- The conditions
- Different options
- Decision-making
- The intervention process
- What happens next
- The effectiveness of the intervention

This enabled us to identify a list of criteria on which we could judge parent information about newborn blood spot screening.

Types of information	NSC criteria for implementing screening programmes	DISCERN criteria for patient information about treatment outcomes	Criteria on which existing leaflets might be judged
A. The conditions	<ul style="list-style-type: none"> • the importance of the health problems • the natural history of the conditions • the natural history of people with carrier status (if the genetic carriers of a condition are identified as a result of screening) 	<ul style="list-style-type: none"> • who it's relevant to and in what context 	<ol style="list-style-type: none"> 1. clarity about who this information is for and in what context 2. the importance of the 4 conditions screened for and a summary of their natural history (including CF and sickle carrier babies) 3. The aims of screening
B. Different options	<ul style="list-style-type: none"> • any primary prevention interventions which have been implemented 	<ul style="list-style-type: none"> • information which is balanced and unbiased • presenting evidence from a range of sources 	<ol style="list-style-type: none"> 4. an outline of any prevention programmes for these 4 conditions (or why there are no other prevention programmes) 5. a clear and unbiased description of what the alternatives to screening are for parents and their babies 6. ensure that evidence is cited from a range of different sources in order to provide balanced information

Appendix 3: Bringing together DISCERN and NSC criteria to develop our appraisal tool

Types of information	NSC criteria for implementing screening programmes	DISCERN criteria for patient information about treatment outcomes	Criteria on which existing leaflets might be judged
C. Decision-making	<ul style="list-style-type: none"> • NSC's definition of screening specifies that the target population is "offered a test", and that "the individual to whom it is offered is helped to make an informed choice" (ref - second report). • the distribution of test values in the target population, and the defined and agreed cut-off level 	<ul style="list-style-type: none"> • the fact that parents have a choice (and support for shared decision making) • the aims of the information leaflet • the relevance of the content to the intended audience • the date of publication • the fact that (and why?) this is evidence-based information • information which is balanced and unbiased • the sources of the information contained in it (dated) • additional sources of support and information • suggestions of alternatives to consider before deciding whether or not to have your baby screened • what would happen if your baby was not screened 	<ol style="list-style-type: none"> 7. a clear description of the choice parents are being offered 8. a clear description of the accuracy of the test results (the distribution of test values and the agreed cut off levels) 9. a clear description of the alternatives to screening 10. a clear description of what would happen if your baby wasn't screened 11. the aims of the leaflet: in providing information to inform parents' choice 12. how the information provided is based on evidence 13. when the information provided was developed 14. the sources (and dates) of the information included 15. additional sources of support and information to assist with the decision-making process
D. The intervention process	<ul style="list-style-type: none"> • the value for money, balancing the costs of screening and expenditure on health care in general • the provision of adequate staffing and facilities of testing, diagnosis, treatment and programme management 		<ol style="list-style-type: none"> 16. the costs of screening and subsequent diagnosis and treatment to parents 17. the cost-effectiveness of screening 18. an outline of the services provided within the screening programme
E. What happens next	<ul style="list-style-type: none"> • the provision of adequate staffing and facilities of testing, diagnosis, treatment and programme management • the agreed policy on the further diagnostic investigation of babies with a positive test result and the choices which those parents will have. • the agreed policies covering which individuals are offered treatment and the appropriate treatment to be offered. 	<ul style="list-style-type: none"> • additional sources of support and information 	<ol style="list-style-type: none"> 19. a continuation of the outline in (4), including the services available for testing, diagnosis and treatment 20. the processes followed for different screening outcomes, and any subsequent choices parents will have. 21. specifically, the process of following up affected babies, and how these cases are managed on a clinical level 22. additional sources of support and information (and about different outcomes???)

Types of information	NSC criteria for implementing screening programmes	DISCERN criteria for patient information about treatment outcomes	Criteria on which existing leaflets might be judged
F. Effectiveness of intervention	<ul style="list-style-type: none"> • the situation re clinical management of those with the condition <hr/> • the evidence that there is an effective treatment for babies identified through early detection, with evidence of early treatment leading to better outcomes than late treatment <hr/> • the evidence from high quality randomised controlled trials that the screening programme is effective in reducing mortality and morbidity • if the test is for mutations the criteria used to select the subset of mutations to be covered by screening if all possible mutations are not being tested • where screening is aimed solely at providing information to allow the person being screened to make an 'informed choice' (eg Downs screening) there must be evidence from high quality trials that the test accurately measures risk; and that the information is of value and readily understood by the individual being screened <hr/> • the evidence that the complete screening programme (including screening test, diagnostic procedures, treatment/intervention) is clinically, socially and ethically acceptable to health professionals and the public • if screening is for a mutation the programme should be acceptable to people identified as carriers and to other family members <hr/> • the safety and validation of the screening tests <hr/> • evidence that the test is acceptable to the population 	<ul style="list-style-type: none"> • that (and how) screening works <hr/> • any areas of uncertainty (either within the evidence or due to lack of evidence) <hr/> • the benefits of screening • description of who is most likely to benefit from screening (or not screening) <hr/> • the risks of screening <hr/> • the risks of not screening 	<ul style="list-style-type: none"> 23. the effectiveness of screening on affected babies 24. the effectiveness of screening on affected babies compared to later diagnosis based on symptoms 25. the effects of screening on the screened population in general: <ul style="list-style-type: none"> • the benefits of screening • the risks of screening • the benefits of not screening • the risks of not screening 26. the evidence that the complete screening programme (information, screening test, diagnostic procedures, treatment) is clinically, socially and ethically acceptable to health professionals and the public NB might include the effectiveness of: <ul style="list-style-type: none"> • receiving information about the screening test • being offered a choice (or not) about screening • having the test performed on your baby • receiving a true positive result • receiving a true negative result • receiving a false positive result • following a false negative result • receiving a carrier result on the following range of outcomes: <ul style="list-style-type: none"> • the physical health of the baby (in particular as measured by number of hospital admissions AND/OR the incidence of infections) • growth (height and weight) of the child • the physical health of the parent(s)

Appendix 3: Bringing together DISCERN and NSC criteria to develop our appraisal tool

Types of information	NSC criteria for implementing screening programmes	DISCERN criteria for patient information about treatment outcomes	Criteria on which existing leaflets might be judged
	<ul style="list-style-type: none"> • evidence that the benefits of the screening programme outweigh the physical and psychological harm (caused by the test, diagnostic procedures and treatment) 	<ul style="list-style-type: none"> • how screening affects overall quality of life • the effects of screening on day to day activity • the effects of screening on relationships with family friends and carers 	<ul style="list-style-type: none"> • family relationships • the relationship between the family and the health service • the psychological health of the parent(s), including anxiety (as well as other emotional responses) • measures of quality of life (? various) • the response of the community or wider family, including stigma • future reproductive choices by the parents • future choices about screening for a) other family members AND b) subsequent newborns • parents' understanding and knowledge, including retention of knowledge • parents' satisfaction with the screening service • parents' communication and decision-making skills • financial costs to the health service

Appendix 4: Appraisal tool for leaflets

Newborn bloodspot screening data extraction April 2003

Section A: Purpose of Leaflet

A.1 Is it clear whom the leaflet is for?	A.1.1 Yes, Parent/Carer A.1.2 Yes, Family A.1.3 Yes, Adult A.1.4 Yes, Child A.1.5 Yes, Health Professional A.1.6 No
A.2 If the leaflet states whom it's for, how is this indicated?	A.2.1 In the title of the leaflet A.2.2 In the text <i>e.g. says 'your baby' or 'my baby'</i>
A.3 Is it clear from the leaflet when in the screening pathway this information would be given?	A.3.1 Yes, Before screening takes place (antenatally or postnatally) A.3.2 Yes, As part of the consent process A.3.3 Yes, At time of the test A.3.4 Yes, With screening results A.3.5 Yes, After confirmed positive diagnosis A.3.6 Yes, Other timing in which information is given (please specify) A.3.7 No

Section B: Background to screening programme

B.1 Does the leaflet give the aims of/reasons for screening Includes statistical information on number of cases of particular condition in a specific population (ethnic or geographic)	B.1.1 Yes, Explains why the tests are recommended/important (list conditions covered) B.1.2 Yes, Explains importance to screen a particular population (list conditions covered) B.1.3 No
B.2 Does the leaflet describe the natural history of the conditions?	B.2.1 Yes, for PKU B.2.2 Yes, for CHT B.2.3 Yes, for SC disorders B.2.4 Yes, for CF B.2.5 Yes, for other disorders B.2.6 No
B.3 Does the leaflet discuss available prevention programmes and/or the difficulties of prevention?	B.3.1 Yes, Specifies that apart from reproductive choice, no prevention programmes exist B.3.2 Yes, Indicates whether or not disorders are inherited B.3.3 Yes, Mentions preconception and/or antenatal screening and reproductive choice B.3.4 Yes, Provides other information about prevention programmes B.3.5 No

Section C: Process of screening

C.1 Does the leaflet describe the procedures for doing the heel-prick test?	<p>C.1.1 Yes, Describes how the blood sample is taken</p> <p>C.1.2 Yes, Indicates that it may be uncomfortable for the baby</p> <p>C.1.3 Yes, Describes how pain may be eased for baby</p> <p>C.1.4 Yes, Describes how bloodspots placed on the Guthrie card</p> <p>C.1.5 Yes, Describes what happens next to the card/blood sample</p> <p>C.1.6 Yes, Describes how results are reported</p> <p>C.1.7 Yes, Discusses the possibility of a repeated test</p> <p>C.1.8 Yes, Indicates storage/possible later use of cards</p> <p>C.1.9 Yes, Other (please specify)</p> <p>C.1.10 No</p>
C.2 Does the leaflet indicate when the parents will receive or be asked to collect the screening results?	<p>C.2.1 Yes, Positive (specify time)</p> <p>C.2.2 Yes, Negative (specify time)</p> <p>C.2.3 No</p>
C.3 Does the leaflet indicate that screening is not compulsory/mandatory?	<p>C.3.1 Yes, Outlines choices/alternatives to screening</p> <p>C.3.2 Yes, Recommends screening</p> <p>C.3.3 Yes, Discusses possible implications of not screening</p> <p>C.3.4 Yes, Outlines processes for accepting/refusing</p> <p>C.3.5 No, Indicates that it is compulsory/mandatory (e.g. in USA)</p> <p>C.3.6 No, Specifies circumstances where refusal is acceptable</p> <p>C.3.7 No, Nothing specified</p>
C.4 Does the leaflet discuss the limitations of screening and the possible harms, e.g. false negatives, false positives? <i>Refers to screening results only, not diagnostic test results.</i>	<p>C.4.1 Yes, False negative</p> <p>C.4.2 Yes, False positive</p> <p>C.4.3 Other (please specify) e.g. "Don't worry"</p> <p>C.4.4 No</p>
C.5 Does the leaflet mention the costs of screening?	<p>C.5.1 Yes, Costs to parents</p> <p>C.5.2 Yes, Costs to the health service</p> <p>C.5.3 Yes, Costs to health insurers (e.g. in USA)</p> <p>C.5.4 No</p>
C.6 Does the leaflet discuss the cost-effectiveness of screening?	<p>C.6.1 Yes</p> <p>C.6.2 No</p>

Section D: Follow-up to screening

D.1 Does the leaflet mention the need for further testing to confirm diagnosis?	D.1.1 Yes, Identifies circumstances in which follow-up would occur D.1.2 Yes, Indicates when parents will hear about necessity for follow-up tests D.1.3 Yes, States when tests might occur D.1.4 Yes, Indicates when parents will receive results of follow-up tests D.1.5 No
D.2 Does the leaflet mention treatment for any of the four conditions?	D.2.1 Yes, PKU D.2.2 Yes, CHT D.2.3 Yes, SC D.2.4 Yes, CF D.2.5 No
D.3 Does the leaflet mention any related services?	D.3.1 Yes, support organisations (within health service or charity) D.3.2 Yes, Support re decision-making D.3.3 Yes, Counselling D.3.4 Yes, Health professional/specialist D.3.5 Yes, Other (e.g. quality assurance, supplementary screening) D.3.6 No

Section E: Production of the leaflet

E.1 Are sources of evidence indicated on the leaflet? <i>For example, included named specialist or specialist organisation.</i>	E.1.1 Yes, evidence provided E.1.2 Yes, dates of evidence provided E.1.3 Yes, more than one type of source used (specify type, e.g. policy document, research paper, specialist reviewer) E.1.4 No
E.2 Is specific information provided linked to evidence?	E.2.1 Yes (please specify) E.2.2 No
E.3 Does the leaflet indicate when the leaflet was developed?	E.3.1 Yes (give details) E.3.2 No
E.4 What is the level of complexity of the leaflet?	E.4.1 Easy to read (technical terms explained or stated in lay language) E.4.2 Some technical terms not explained or confusing E.4.3 Requires some expert/medical knowledge to understand
E.5 Does the leaflet indicate how the information was developed?	E.5.1 Yes E.5.2 No

Appendix 5: Leaflets and Information Sheets included in the Survey

1. Gilham, P. (2002) Notification letter. Reading, UK, Sickle Cell and Thalassaemia Service, Haematology Department, Reading
5. Gilham, P. (2002) Notification letter. Reading, UK, Sickle Cell and Thalassaemia Service, Haematology Department
12. Brayden, R. (1999) Newborn Screening Tests. Orange County, US, Children's Hospital of Orange County - <http://www.choc.com/>
13. (2003) Newborn Screening Program Overview. California, US, California Department of Health Services
14. (2001) Important Information for Parents about the Newborn Screening Test. California, US, California Department of Health Services - <http://www.dhs.cahwnet.gov/>
18. (2003) Newborn Screening Program Overview for Parents. California, US, California Department of Health Services - <http://www.dhs.ca.gov/>
33. (2002) The importance of newborn screening. US, Akron Children's Hospital - <http://www.akronchildrens.org/>
34. (2002) Newborn Screening: For Your Baby's Health. New York, US, NYS Department of Health - <http://www.wadsworth.org/>
35. (2003) Parents' Page: Newborn Screening in Wisconsin. Wisconsin, US, Wisconsin Division of Public Health Newborn Screening - <http://slh.wisc.edu/>
36. (2003) Most Important Health Messages for Patients. March of Dimes - <http://www.marchofdimes.com/>
38. (2003) Newborn Screening Tests. US, March of Dimes - <http://209.10.141.228/>
39. (2003) Professionals and Researchers, Quick Reference: Birth Defects and Genetics. US, March of Dimes - <http://209.10.141.228/>
41. (2003) Expanded Newborn Screening. Victoria, Australia, Genetic Health Services Victoria - <http://www.genetichealthvic.net.au/>
43. (2001) Tests to Protect Your baby. Why early diagnosis is important to your baby's future. New South Wales, Australia, NSW Newborn Screening Programme - <http://www.health.nsw.gov.au/>
45. (2000) Information for parents/carers. Further investigations requested for congenital hypothyroidism. New South Wales, Australia, New South Wales Newborn Screening Programme - <http://www.chw.edu.au/>
46. (2000) Information for parents / carers for cystic fibrosis. New South Wales, Australia, New South Wales Newborn Screening Programme - <http://www.chw.edu.au/>
49. (2002) Congenital Hypothyroidism. London, UK, Institute of Child Health and Great Ormond Street Hospital for Children - <http://www.ich.ucl.ac.uk/>

56. (2003) Overview of Newborn Screening for Defects. Stanford, US, Lucile Packard Children's Hospital, <http://www.lpch.org/>
71. (2001) Child Health A-Z: Sickle Cell Disease. Boston, USA, Children's Hospital Boston - <http://www.web1.tch.harvard.edu/>
89. (2003) Newborn Screening Tests. Pittsburg, US, Children's Hospital of Pittsburg - <http://www.chp.edu/>
104. (2000) The Blood Test on Newborn Babies. Leeds, UK, The Leeds Teaching Hospitals
106. Neonatal Biochemical Screening - The Guthrie Test. Addenbrooke's NHS Trust
107. (2000) Your baby's blood test. Northampton, UK, Northamptonshire Neonatal Screening Service
108. Neonatal Screening - A practical guide. Belfast, UK, The Royal Hospitals
128. Hulse, T. (1998) Hypothyroidism. London, UK, British Society for Paediatric Endocrinology and Diabetes - <http://www.bsped.org.uk/>
135. (2003) Kentucky's Newborn Screening Program. Kentucky, US, KY Cabinet for Health Services - <http://publichealth.state.ky.us/>
136. (1997) Ask NOAH About Pregnancy. PKU Public Health Educational Information Sheet, Genetic Series. March of Dimes Birth Defects Foundation - <http://www.noah-health.org/>
137. (1999) Newborn Screening Tests - Public Health Education Information Sheet. White Plains, US, March of Dimes - <http://www.noah-health.org/>
141. Beasley, J. (1997) Sickle Cell Newborn Screening - Frequently Asked Questions (FAQs). Georgia, US, The Georgia Sickle Cell Centre - <http://www.emory.edu/>
153. (2003) What Parents should Know - A Summary. New England, US, New England Newborn Screening Program - <http://www.umassmed.edu/>
157. (2003) Answers to Common Questions about Newborn Screening. USA, New England Newborn Screening Program
159. (2003) Newborn Screening Case Management Program. Texas, US, Texas Department of Health - Newborn Screening - <http://www.tdh.state.tx.us/>
160. (1998) Newborn Screening Case Management Program: Hypothyroidism and Your Infant. Texas, US, Texas Department of Health - <http://www.tdh.state.tx.us/>
161. (2003) Newborn Screening Case Management Program. Congenital Hypothyroidism. Texas, US, Texas Department of Health - <http://www.tdh.state.tx.us/>
167. (1999) Phenylketonuria (PKU). Texas, US, Texas Department of Health - <http://www.tdh.state.tx.us/>
174. (2003) Nebraska Newborn Screening Program - Practitioner's Manual. Nebraska, US, Nebraska Department of Health and Human Services - <http://www.hhs.state.ne.us/>
175. (2003) Congenital Hypothyroidism. New York, USA, Wadsworth Centre. New York State Department of Health - <http://www.wadsworth.org/>

177. Newborn Screening for Cystic Fibrosis. Pittsburg, US, University of Pittsburg, USA - <http://www.pitt.edu/>
183. A Simple Test Could Save Your Baby's Life - A Parent's Guide to Newborn Screening. Save Babies Through Screening - <http://www.savebabies.org/>
186. (2002) Antenatal, Neonatal and Childhood Screening: A summary for professionals. Contact a Family - <http://www.cafamily.org.uk/>
194. (2001) The Neonatal Screening Test - Guidance for Parents. Sheffield, UK, Trent Region Neonatal Metabolic Screening Service
195. The Heel-Prick Test. Blood Screening Tests for Newborn Babies. Surrey, UK, South West Thames Regional Infant Screening Service
200. (2002) Genetics, Carrier Testing, Tests during Pregnancy and for Newborn Babies. London, UK, Cystic Fibrosis Trust - <http://www.cftrust.org.uk/>
202. (1998) Overview: Newborn Screening. US, National Newborn Screening and Resource Center - <http://genes-r-us.uthscsa.edu/>
204. (1996) Newborn Screening Fact Sheets. US, American Academy of Pediatrics - <https://www.aap.org/>
205. Collins, J. (2000) Ask Dr Jane Collins... Newborn Screening - The Times, 2 April 2000. London, UK, GOSH/ICH - <http://www.ich.ucl.ac.uk/>
207. Chalmers, R. A. (2003) Screen Our Babies. Surrey, UK, Screen Our Babies - <http://www.sghms.ac.uk/>
209. Hait, E. (2001) Neonatal cystic fibrosis screening. US, United States National Library of Medicine - <http://www.nlm.nih.gov/>
213. Bartoszesky, L (2000) Newborn Screening Tests. Ohio, US, Children's Hospital Medical Center of Akron - <http://www.kidshealth.org/>
233. Guthrie Test. United Hospitals
262. (2002) A parents' guide to newborn screening for PKU, CH, and CF. Scotland, UK, Health Education Board for Scotland
263. Information sheet for parents whose baby needs a repeat test for cystic fibrosis. Leeds, UK, The Leeds Teaching Hospitals
265. (2002) Your baby's screening test result - what does it mean? Sheffield, UK, Trent Neonatal Screening Service, The Children's Hospital, Sheffield
267. Rotenstein, D. (2000) Thyroid Division - congenital hypothyroidism. Chicago, US, The Magic Foundation - <http://www.magicfoundation.org/>
269. The Heel-Prick Test. Birmingham, UK, Birmingham Children's Hospital NHS Trust
270. Hall, F. and Hanley, G. The Heel Prick Test on Newborn Babies. St James's University Hospital, Leeds, UK, Farley's Baby Milks
271. (2002) Congratulations on the birth of your baby. Ipswich, UK, Maternity Dept, The Ipswich Hospital NHS Trust

272. (1994) Policy for Screening of Phenylketonuria and Congenital Hypothyroidism. UK, North Tees Health NHS Trust
273. (2003) Neonatal screening blood tests - Guthrie Test. Peterborough, UK, Peterborough Hospitals NHS Trust
274. (2002) Guidelines for the Newborn Blood Spot Screening Programme in Hertfordshire. Hertfordshire, UK, Hertfordshire Phenylketonuria Working Party
275. The neonatal screening test - guidance for midwives. Sheffield, UK, Trent RHA
276. (1993) Blood Tests for Newborn Babies. Surrey, UK, The St Helier NHS Trust
277. To parents of new babies. Gloucester, UK, Gloucestershire Royal NHS Trust
278. Blood Test for New Born Babies. UK, South West Thames Regional Health Authority
279. (1994) To parents of new babies. Derriford, UK, Plymouth Health Authority
280. Your baby's blood test. North Durham, UK, Community Health Care: North Durham NHS Trust
281. Why is my baby having blood tests? London, UK, Brent Sickle Cell and Thalassaemia Centre
282. (2003) Congenital Hypothyroidism. New England, US, New England Newborn Screening Program - <http://www.umassmed.edu/>
283. (1993) Procedure Guidelines for Explaining PKU/Guthrie Tests to Patients. Walsall, UK, Walsall Hospitals NHS Trust
284. Guthrie Test. Maternity Unit, Southend Hospital. Patient Information Service.
285. (1993) Phenylketonuria and Hypothyroid Screening Test. Plymouth, UK, Plymouth Health Authority Child Health Directorate
286. (2002) Guideline for the Guthrie Test in the Neonate. Southend, UK, Southend Hospital NHS Trust
287. (1994) All about your baby's blood test. Wiltshire, UK, Cow & Gate Nutricia Limited
288. Routine Blood Tests for New-Born Babies. Information for Parents. Bath & West Community NHS Trust; Royal United Hospital NHS Trust; Wiltshire NHS Trust.
290. (1996) Neonatal Screening Protocol. Brighton, UK, Brighton Health Care NHS Trust; Directorate of Women and Children's Health
291. (1996) Blood Screening Tests for Babies - The Heel Prick Test. Middlesex, UK, Milupa Limited
292. Blood Screening Tests for Babies - The Heel Prick Test. UK, Bradford & Airedale Health Promotion Unit
293. (2001) Screening the Newborn. UK, Riverside Community Healthcare NHS Trust
294. (2003) The Guthrie Test. Information for Post Natal Mothers. UK, The Princess Alexandra Hospital NHS Trust

295. Green, A., Holmes, B., MacDonald, A., and Rylance, G. (1996) Blood Screening Tests for Babies in the UK - The Heel-Prick Test. Birmingham, UK, The Children's Hospital
296. Colbourne, C. (1996) Neonatal Screening for PKU, CHT, CF Generally Known as the Guthrie Test. Ipswich, UK, The Ipswich Hospital NHS Trust
298. Your baby's blood tests explained. London, UK, West Lambeth Community Care (NHS) Trust
299. The Heel Prick test and your baby. UK, Southeast Thames Newborn Screening Laboratory Partnership
300. Heel Prick Test. Maidstone and Tunbridge Wells, UK, Maidstone and Tunbridge Wells NHS Trust
301. (2001) Blood tests for your baby. London, UK, Hammersmith Hospitals NHS Trust
302. (2001) Your baby's Guthrie test. Peterborough Maternity Unit, Peterborough Hospitals NHS Trust
303. The Heel Prick Test - Testing for haemoglobin disorders in Birmingham. Birmingham, UK, Birmingham Sickle Cell and Thalassaemia Centre
304. All About Your Baby's Blood Test. London, UK, West London Healthcare NHS Trust
305. (1997) Baby's Blood Test. Grimsby, UK, Grimsby Maternity Hospital
306. Lepper, E. (1988) A blood test - on a well baby! Is it necessary? For mine? Peterborough, UK, Peterborough District Hospital
307. (2001) Routine Blood Tests for Newborn Babies. Poole, UK, Poole Hospital NHS Trust
309. (1995) Routine Blood Tests for Newborn Babies. Salisbury, UK, Salisbury District Hospital Maternity Services
310. (1996) Guthrie Testing. Surrey, UK, Frimley Park Hospital NHS Trust
311. (2000) Your baby's blood test for PKU and Hypothyroidism. Sidcup, UK, Queen Mary's Sidcup NHS Trust
312. (1994) All About Your Baby's Blood Test. Thanet, UK, Thanet Healthcare NHS Trust
313. (1995) The Guthrie Test - Information for Parents. South Tyneside, UK, South Tyneside Health Care Trust
314. (1996) Blood Tests for New Born Babies. UK, Wessex Health Authority
315. (1996) Heel Prick Test. Maidstone, UK, Midkent Healthcare Trust
316. A Guide to Maternity Care - Blood tests for newborn babies. Sussex, UK, St Richard's Hospital, The Royal West Sussex Trust
317. (1994) Your Babies Blood Test for PKU and Congenital Hypothyroidism. Unknown, Unknown
318. Hulse, T. (1998) Thyroid Disorders. Middlesex, UK, Serono Pharmaceuticals

319. Neonatal Screening Blood Specimen Collection and Handling Procedure. New Hampshire, US, Schleicher & Schuell
320. Screening for Cystic Fibrosis - Request for Repeat Test. Leeds, UK, St James's University Hospital
321. Hall, K., Asplin, D., and Green, A. (2000) Your baby has had a positive heel-prick blood test. Birmingham, UK, Birmingham Children's Hospital
322. Hall, K., Asplin, D., and Green, A. (2000) Your baby has had a positive heel-prick blood test. Birmingham, UK, Birmingham Children's Hospital
323. (2003) Blood Spot Screening Tests for Babies in the West Midlands. Birmingham, UK, Birmingham Children's Hospital NHS Trust

Appendix 6: Applying principles underpinning DISCERN criteria to information about screening

DISCERN is designed to appraise information about treatment choices. In applying DISCERN questions to screening information, we needed to replace treatment choices with choices about whether to not to screen. If leaflets were to provide parents with the same information that is required by the DISCERN they would include the following information:

DISCERN criteria	Acceptable answer to our question	Question/answer from our appraisal tool
1. Are the aims of the information clear?	It's clear about who the leaflet is for AND It's clear when the leaflet would be given	A.1.1 – A.1.5 AND A.3.1 – A.3.6
2. Does it achieve its aims?		Our appraisal tool does not address this question
3. Is it relevant?	Explains importance to the population	B.1.2 or B.1.2
4. Is it clear what sources of information were used to compile the publication (other than the author or producer)?	That evidence is provided and isn't just the author	E.1.3 OR E.1.1 (as long as this is not just the name of the author – check 'details' box)
5. Is it clear when the information used or reported in the publication was produced?	Dates of evidence provided AND not just author and date of last update	E.1.2 AND E.1.1 (as long as this is not just the name of the author – check 'details' box)
6. Is it balanced and unbiased?	Mentioned false-positive AND Mentioned false-negative OR Other possible harms (e.g. worry)	C.4.1 AND C.4.2 OR C.4.3
7. Does it provide details of additional sources of support and information?	Mentioned support organizations	D.3.1
8. Does it refer to areas of uncertainty?	Mentioned false-positive AND Mentioned false-negative OR Other possible harms (e.g. worry)	C.4.1 AND C.4.2 OR C.4.3

DISCERN criteria	Acceptable answer to our question	Question/answer from our appraisal tool
9. Does it describe how each treatment works?	Does it mention the natural history of the conditions AND The importance of screening	B.2.1 OR B.2.2 OR B.2.3. OR B.2.4 AND B.1.1
10. Does it describe the benefits of each treatment?	Does it mention the benefits of screening	B.1.1 OR B.1.2
11. Does it describe the risks of each treatment?	Does it describe the risks of screening (false positives and/or false negatives or anxiety) AND The risks of not screening (natural history and why tests are important)	C.4.1 OR C.4.2 OR C.4.3 AND B.2.1 AND B.2.2 OR B.2.3. OR B.2.4 AND B.1.1
12. Does it describe what would happen if no treatment were used?	The risks of not screening (natural history and why tests are important)	B.2.1 AND B.2.2 OR B.2.3. OR B.2.4 AND B.1.1
13. Does it describe how the treatment choices affect overall quality of life?		Our appraisal tool does not address this question
14. Is it clear that there may be more than one possible treatment choice	Mentions choices OR States that it's mandatory	C.3.1 OR C.3.5
15. Does it provide support for decision-making	Mentions support re decision-making	D.3.2

Appendix 7: Applying principles underpinning NSC criteria to information available to parents

If leaflets were to provide parents with the same information that is required by the NSC, as specified in the first report of the NSC (1998) (in order to decide that a screening programme should be introduced) they would include the following information:

	NSC criteria	Acceptable answer to our question	Question/answer from our appraisal tool
1.1	That the condition is an important health problem	Explains importance of tests	B.1.1 OR B.1.2
1.2	The natural history of the condition That there is a clear means of identifying the condition during an early asymptomatic phase	Natural history of at least one of the conditions AND Explains importance of tests	B.2.1 OR B.2.2 OR B.2.3 OR B.2.4 AND B.1.1 OR B.1.2
1.3	All available prevention programmes have been implemented as far as possible	Specifies that there are no prevention programmes OR provides other info about available prevention programmes	B.3.1 OR B.3.4
1.4	That there is a safe, precise and validated screening test	Description of how the blood is taken AND Description of the possibility of repeated tests AND False positive rates AND False negative rates	C1.1 AND C1.7 AND C.2.1 AND C.2.2
1.5	Distribution of test values and clear cut off levels	Our appraisal tool does not address this question	
1.6	Test is acceptable to population	Our appraisal tool does not address this question	
1.7	Agreed policy for follow up for positive screens, and any further choices which might need to be made	Circumstances in which follow-up would occur OR Indication when parents will hear about the necessity of further tests OR Statement about when follow-up tests might occur OR Indication of when parents will receive results of follow up tests.	D.1.1 OR D.1.2 OR D.1.3 OR D.1.4

NSC criteria	Acceptable answer to our question	Question/answer from our appraisal tool
1.8 Effective early treatment, with evidence that early detection and treatment leads to better outcomes	Mentions treatment for at least one of the conditions AND Explains why the tests are important AND Provides evidence	B.1.1 AND B.2.1 OR B.2.2 OR B.2.3 OR B.2.4 AND E.1.1
1.9 Agreed evidence-based policy about who should be offered treatment	Mentions treatment AND Provides the evidence	D.1.1 AND E.1.1
1.10 Clinical management optimized prior to screening	Mentions treatment of at least one of the four conditions AND Mentions a health professional / specialist	D.2.1 OR D.2.2 OR D.2.3 OR D.2.4 AND D.3.4
1.11 Evidence from high quality RCTs that the programme is effective in reducing mortality and morbidity	Provides evidence AND Explains why tests are important	E.1.1 AND B.1.1
1.12 Evidence that the complete screening programme is clinically, socially and ethically acceptable to health professionals and the public	Our appraisal tool does not address this question	
1.13 That the benefits of the screening programme outweighs the physical and psychological harm	Provides full details about the advantages and limitations of the screening programme, including: Explains why the tests are recommended/important AND Discusses false-negatives OR false positives OR other limitations	B.1.1 AND C.4.1 OR C.4.2 OR C.4.3
1.14 Opportunity costs should be balanced with other health care costs – value for money	Any mention of cost-effectiveness	C.6.1
1.15 Must be a plan for managing and monitoring screening and an agreed set of quality assurance standards	Refers to quality assurance mechanisms	D.3.5 – search for quality assurance

NSC criteria	Acceptable answer to our question	Question/answer from our appraisal tool
1.16 Adequate staffing and facilities for testing, diagnosis and treatment	Mention of follow up testing OR Mention of treatment	D.1.1 OR D.1.2 OR D.1.3 OR D.1.4 OR D.2.1 Or D.2.2 OR D.2.3 OR D.2.4
1.17 All other options for managing the condition should have been considered	Specifies no prevention programme available OR Indicates whether or not disorders are inherited OR Mentions preconception and/or antenatal screening and reproductive choice OR Provides other information about prevention programmes	B.3.1 OR B.3.2 OR B.3.3 OR B.3.4

Appendix 8: Glossary

affected	When someone has a condition, it is said that they are affected . A child, who is <i>affected</i> with CHT , is a child who has CHT .
antenatal screening	Antenatal screening , is screening which is carried out before a baby is born. This can include doing tests on the pregnant mother, her partner or the unborn baby. Antenatal screening includes tests for a wide range of conditions, including haemoglobinopathies ..
audit	This is the evaluation of clinical performance against standards or through comparing the same services in different settings, with the aim of improving health service delivery.
blood sampling	Blood sampling is the collecting of a small amount of blood referred to as a blood sample . In the case of newborn screening it refers to the collection of a small amount of blood from the baby's heel at about one week of age. This is done by pricking the heel with a lancet or similar device, to draw drops of blood which are then dropped onto a Guthrie card .
blood spot	When newborn babies are about a week old a blood sample is taken from their heel. This is sometimes referred to as the heel prick test. Blood from the baby's heel is dropped onto a special type of filter paper called a Guthrie card and stored as a series of bloodspots . A number of tests are then carried out on these <i>bloodspots</i> for the purposes of newborn screening . These tests are often called newborn bloodspot screening to distinguish them from other types of newborn screening, such as hearing screening , which don't involve this blood test. These bloodspots can be stored for very long periods of time.
carrier	Everyone inherits two copies of a gene – one from their father and one from their mother. One or both copies of a gene may have a change (mutation) that alters the function of the gene . For some gene pairs, disease only results if both members of that gene pair have a mutation . A carrier is someone who has one copy of a changed (mutated) gene in a gene pair, where disease occurs only if both members have a mutation, and therefore has no symptoms and no disease. The carrier can pass on the changed gene to their offspring. If a person inherits mutations in the same gene from both parents then they will have the disorder associated with mutations in two copies of that particular gene pair e.g. cystic fibrosis , sickle cell anaemia or phenylketonuria . Carriers are also referred to as heterozygotes .
Child Health Department (often referred to as 'child health')	The Child Health Department keeps records about each child who is born within their area of responsibility. When a mother gives birth the Child Health Department is notified of the birth. The results of newborn screening tests are also reported to the Child Health Department .

Child Health Subgroup	The Child Health Subgroup is a smaller group within the National Screening Committee, which concentrates on the screening of children.
condition	Many different words are used to describe illnesses. They are sometimes called diseases , or disorders , or conditions .
congenital hypothyroidism (CHT)	Congenital hypothyroidism - a condition which newborn babies are tested for. Hypothyroidism is a condition in which not enough thyroid hormone is produced. Thyroid hormone, or thyroxine (T4), is made by the thyroid gland, which is located in the front of the neck just beneath the Adam's apple. In most cases of hypothyroidism, the problem arises from an absent or under-functioning thyroid gland. In rare instances there can be a normally functioning gland, which does not make enough thyroid hormone because of insufficient thyroid stimulating hormone (TSH) from the pituitary. Babies born like this have congenital hypothyroidism (CHT) . By measuring thyroid hormone levels in all babies shortly after birth, newborn screening programmes are able to identify infants with low thyroid hormone levels who may have CHT before there are any signs or symptoms of hypothyroidism. Prompt and appropriate treatment of infants with CHT with manufactured thyroid hormone (thyroxine) allows normal growth and intellectual development. CHT has been screened for throughout the UK since the 1980s.
confirmed result	A confirmed result is used to refer to a positive screening result that has been confirmed by further diagnostic tests . Screening test results are often called presumptive results . Screening tests are not diagnostic tests; abnormal screening results need to be followed by further testing. A confirmed result could be a normal result, meaning that the individual is found to be unaffected by any of the conditions screened for.
consent	Consent is agreement to a plan of action or particular treatment. "Informed consent" is when a patient (or parent) is given information and then voluntarily agrees to continue. Another term used is "informed choice". Informed choice is when a patient (or parent) is given a choice and provided with information about the procedure, treatment or test to inform their decision.
coverage	When assessing screening programmes, one term used is coverage . This term describes the proportion of people eligible for screening who are actually screened. The success of screening programmes is sometimes measured by the coverage achieved. Within the newborn bloodspot screening programme, very high levels of coverage have been recorded in the UK but we need to continue to record this information to identify any problems at an early stage.
cystic fibrosis	Cystic fibrosis (CF) is a condition that affects the organs in the body, especially the lungs and pancreas, by clogging them with thick sticky mucus. Children who have cystic fibrosis are born with the disorder, but early recognition of the diagnosis may be delayed by either lack of symptoms or failure to recognise that symptoms may be due to CF. Hence newborn screening can help make the diagnosis earlier for some families allowing earlier access to treatment. There is no cure for cystic fibrosis, but early treatment appears to improve the health of affected

children. Cystic fibrosis is more common in some populations within the **UK** than others. Some areas have been screening for CF in the UK since the 1980s and more recently, newborn screening for CF has started in Scotland. Within the next few years all newborn babies born in the UK will be screened for cystic fibrosis.

diagnosis / diagnostic test	A diagnostic test is one which tests for a specific condition , and confirms whether or not someone has a condition. Several diagnostic tests may follow a positive screening result in order to obtain a confirmed result . For example a newborn baby might be screened for CHT and the screening result is positive. Further diagnostic tests will then be carried out to find out whether the child definitely has CHT . However, treatment will often be started before the confirmed result is available, as this may take some weeks.
disease	Different words are used to describe illnesses. They are sometimes called <i>diseases</i> , or <i>disorders</i> , or <i>conditions</i> .
disorder	Different words are used to describe illnesses. They are sometimes called <i>diseases</i> , or <i>disorders</i> , or <i>conditions</i> .
false-negative	A child with a false-negative screening result is one who really has the disorder even though the screening result was negative. For example, a child who has a false-negative result for CHT is one who is told that they don't have the condition, and then it turns out that they do have CHT . Depending on the condition, this can be very serious. All screening tests are associated with a variable risk of false- negative and false-positive results.
false-positive	A child with a false-positive screening result is one who is thought to have the disorder when in fact they do not. For example, a child who has a false-positive result for CHT is a child who has been told they have the condition, and then it turns out that this is not the case. For parents, receiving a false-positive result can mean that they think that their child is sick, when actually their child is healthy.
Guthrie Card	When the midwife collects small drops of blood from a newborn baby, she puts them on a special piece of filter paper, which is often referred to as a Guthrie Card . This special card, named after Dr Robert Guthrie who developed a simple accurate test for PKU in 1963, allows the blood to be stored as blood spots on the card while it is sent to the laboratory for testing.
haemoglobinopathies	Haemoglobinopathies are disorders of haemoglobin. Haemoglobin is the part of our blood that carries oxygen. There a large number of different haemoglobinopathies, some are more serious than others. Sickle cell disease is a haemoglobinopathy as is thalassaemia . Haemoglobinopathies are more common in some populations within the UK than others. Haemoglobinopathies can be tested for in pregnant mothers, and unborn babies, as well as newborn babies. In the UK some areas have been screening newborn babies for sickle cell disorders since the 1980s. In many areas only those babies thought to be at high risk are tested. This is changing and over the next few years all newborn babies in England will be screened for sickle cell

disorders.

heterozygote	A heterozygote is another term for a carrier . A carrier has a single copy of a change or mutation in a particular gene pair when, for that gene pair, disease only results when both copies have a mutation. The carrier can pass on the changed gene to their offspring. A distinction is made between a simple heterozygote, a person who carries only one copy of a genetic mutation and is therefore unaffected by a disease (e.g. sickle cell trait) and a compound heterozygote, who carries two different genetic mutations and is affected by the disorder (sickle cell disease).
informatics	Health <i>informatics</i> is an evolving scientific discipline that deals with the responsible collection, storage, retrieval, communication and optimal use of health related data, information and knowledge. to improve patient care, medical education, and health sciences research.
negative screening result	A negative screening result suggests that it is unlikely that the child has a disease. Under these circumstances further diagnostic tests are not needed to show whether or not the child is in fact affected by the condition screened for. A negative result is sometimes referred to as a normal result . This is a misleading term, however, as screening is not 100% certain and a child with a negative screening result may later turn out to have the disease, while conversely, a child with a positive result may turn out not to have the condition.
neonatal screening	Neonatal screening can also be called newborn screening. All screening on a newborn baby is called neonatal (or newborn) screening. There are different newborn screening tests, for example neonatal screening includes hearing screening, hips screening and bloodspot screening.
normal (result)	Sometimes when the result of the test shows that the child does not have (or is unlikely to have) the condition tested for, people say the result is normal . In general it is best to avoid using this term, as it is not always clear what normal is meant to be. It is a misleading term to refer to in screening results as screening is not 100% certain and a child with a negative screening result may later turn out to have the disease, while conversely, a child with a positive screening result may turn out not to have the condition
notification	When talking about screening people often refer to <i>notification</i> of results. This can mean a number of slightly different things. Sometimes notification means the reporting of a screening result to a register, or a health monitoring group, such as the child health record. Sometimes notification refers to telling parents or patients the results of their tests. When talking about notification, it is important to be clear about what information is being notified, and who is being notified about this information.
Personal Child Health Record (PCHR)	The Personal Child Health Record is the child health record held by the parent. It is a book normally issued by the health visitor at 10 days of age, or sometimes sooner. The PCHR contains information about the child's health, such as dates of vaccinations

positive result	A positive screening result suggests that it is more likely that the child has a disease. A positive screening result indicates that further diagnostic tests are needed to show whether or not the child is in fact affected by the condition screened for. A child cannot be said to be affected until further diagnostic tests are undertaken and a confirmed result is obtained
presumptive screening results	Presumptive screening results are results which are not yet confirmed, but which are considered highly likely. A presumptive positive for CHT means that it is very likely, or assumed that the child has CHT. This result is then confirmed using a diagnostic test. Screening results are described as presumptive. A presumptive positive result can also be described as a positive screening result or screen positive . A presumptive negative result can also be described as a negative screening result, or screen negative .
process standards	Agreed standards that should be achieved at each stage of the <i>screening</i> process: informing parents, taking the sample, laboratory testing, informing clinicians and parents of the results, and starting treatment of affected newborns as soon as it is beneficial.
quality assurance	Quality assurance is a system for monitoring and maintaining high standards in every aspect of the screening programme.
registers / disease registers	There are a number of disease registers in the UK, which keep a record of the number of people with particular conditions. These registers can serve different purposes: ensuring people with the condition are followed up, and treated and helping us to understand more about these conditions and how they affect people etc. There are different registers which collect slightly different information about different conditions. One of the tasks of the Programme Centre is to develop and maintain national registers for conditions which are screened for in newborn blood spot screening.
screen negative (results)	Screening results are not 100% conclusive. Instead they provide presumptive results , which are then confirmed using diagnostic tests . A screen negative result for CHT , means that it is highly likely that the child does NOT have CHT . This screen negative result is NOT usually confirmed using further tests, but it is assumed the child is not <i>affected</i> .
screen positive (results)	Screening results are not 100% conclusive. Instead they provide presumptive results , which are then confirmed using diagnostic tests . A screen positive result for CHT , means that it is highly likely that the child has CHT , but that this must still be confirmed by further tests.
screening	Screening is when healthy children and adults are tested to see if they are likely to develop a condition. Screening tests don't generally confirm that a person has a disease. Usually they will not feel ill from these conditions in any way at the time when they're screened. Screening allows diseases to be identified early, before any signs of illness. This means people can be treated quickly, and hopefully avoid getting seriously ill. Screening can happen at different stages, and for different conditions. Newborn screening in this country includes tests for

phenylketonuria (PKU), congenital hypothyroidism (CHT), cystic fibrosis (CF), and sickle cell disorders.

sensitivity	The sensitivity of a screening test refers to the proportion of individuals who have the condition who are correctly identified by the screening test. A highly sensitive test may identify nearly all those affected, i.e. it has a sensitivity approaching 100%. The consequence of a test that lacks sensitivity is that individuals with a negative screening result may believe they do not have the disease when in fact they do. They have a false-negative screening result and are falsely reassured by the screen.
setting standards	Setting standards is the process by which minimum acceptable standards, achievable standards and optimal standards are agreed. This is usually through discussion and review of current practice and research by clinicians, managers, researchers and, increasingly, people using the services
sickle cell disorders	Sickle Cell disorders (also known as sickle cell disease) are a group of conditions that affect the way that our blood carries oxygen. The part of the blood that carries oxygen is called haemoglobin, which is found inside our red blood cells. Sickle Cell disorders can also be called haemoglobin-opathies . Types of sickle cell disorders include sickle cell anaemia, haemoglobin SC disease and sickle beta thalassaemia. A person who has a sickle cell disorder has red blood cells which, under certain conditions, change their shape to that resembling a sickle or half-moon. These cells cannot carry oxygen properly. The condition can be very painful and can cause various health problems including anaemia, episodes of pain, strokes, and life-threatening infections. Treatment with penicillin may prevent serious infections in early childhood.
specificity	The specificity of a screening test refers to the proportion of individuals who do not have the disease who are correctly identified by the screen. A highly specific test means that all those who are not affected are correctly identified, i.e. the specificity approaches 100%. The consequence of a test that lacks specificity is that an individual may receive a false-positive result, indicating that they may have a condition when in fact they do not.
true-negative	A true-negative result is one which is thought to be negative and is. A person with a true negative result for CHT , is someone who does not have CHT .
true-positive	A true-positive result is one which is thought to be positive and is. A person with a true positive result for a condition, is someone who has that condition.
UK	The UK includes England, Wales, Scotland and Northern Ireland.
UK Newborn Screening Programme Centre	The UK Newborn Screening Programme Centre has been funded by the Department of Health to develop national standards for newborn bloodspot screening. The Programme Centre is made up of a team of people from Great Ormond Street Hospital NHS Trust, the Institute of Child Health and the Institute of Education.

Working Group

A **working group** is a group of people who meet on several occasions to complete a particular task. For example, a working group may be convened to decide the standards for newborn screening services (Process Standards Working Group), or to prepare parent information on newborn screening (Information and Communication Working Group).

First produced in 2005 by:

Social Science Research Unit
Institute of Education, University of London
18 Woburn Square
London WC1H 0NR
Tel: 020 7612 6397

<http://www.ioe.ac.uk/ssru/>

Founded in 1990, the Social Science Research Unit (SSRU) is based at the Institute of Education, University of London. Our mission is to engage in and otherwise promote rigorous, ethical and participative social research as well as to support evidence-informed public policy and practice across a range of domains including education, health and welfare, guided by a concern for human rights, social justice and the development of human potential.

The views expressed in this work are those of the authors and do not necessarily reflect the views of the funder. All errors and omissions remain those of the authors.

This report is available to download from:

<http://www.ioe.ac.uk/ssru/reports/surveyleafletsnewbornbloodspots.pdf>

ISBN: 0-9550487-2-9

This document is available in a range of alternative formats.
Please contact the Marketing and Development Office for assistance.

Telephone 020 7947 9556 Email info@ioe.ac.uk