Cleft Care UK 2010 - 2012

Care for children with unilateral cleft lip and/or palate in the UK

Research Protocol

Version 3.3
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BACKGROUND

Cleft lip and palate (CLP) is one of the most common congenital abnormalities in the UK with an estimated prevalence of 1 in 700 live births (1, 2). Prevalence varies by ethnicity, geographic origin and gender with males more likely than females to have a unilateral cleft lip and palate (UCLP)(3). In addition a left-sided cleft is more common than a right sided cleft (4). Care of a person with cleft lip and palate is complex. It usually begins with antenatal diagnosis and continues into adulthood. Treatment of cleft lip and palate is resource intensive requiring evaluation and care from a range of specialists including Audiologists, Cleft Nurse Specialists, Clinical Geneticists, Clinical Psychologists, Dieticians, Orthodontists, Medical Photographers, Paediatricians, Paediatric Dentists, Restorative Dentists, Speech and Language Therapists, and Surgeons (from ENT, Plastic Surgery, and Maxillofacial Surgery) to achieve the optimum outcome.

In the 1990s concerns were expressed over the standard of care and outcome in children with cleft lip and palate in the United Kingdom (UK) and the training of those health care professionals providing it (5-8). These concerns led the Clinical Standards Advisory Group (CSAG), to commission a national survey of the care and outcome in children with CLP – referred to in this document as the CSAG survey (9, 10). This survey began in 1996 and ran over a 15 month period. The survey aimed to include all non-syndromic children in the U.K., born with a complete UCLP aged either 5 or 12-years of age. The outcomes assessed included the quality of speech, hearing, naso-labial appearance, dental arch relationships, oral health, the success of alveolar bone grafts (in the 12 year old cohort only) and child/parent satisfaction (11-14).

In 1998 the group published their main report (1). Fifty seven centres were identified as providing a cleft service in the U.K. with the number of cases being treated per year varying considerably. They found that the outcomes of cleft care in the U.K. were generally poor. One of the key findings was the poor outcome with respect to dental arch relationships and profile, when compared with three European centres (Oslo, Bergen, and Nantes). It was reported that 36% of 5 year olds and 39% of 12 year olds with UCLP had a poor dental arch relationship. Other centres in Europe have shown outcomes with less than 10% of UCLP cases having poor dental arch relations (1).

On the basis of this survey it was concluded that some children with CLP were not receiving optimal care in the UK. The group made nine recommendations to the government all of which were accepted (Appendix 1). The acceptance of these recommendations led to a major reorganization of cleft services across the UK, with the concentration of resources and expertise in fewer units. Current arrangements have configured eleven cleft centres carrying out operations at seventeen cleft sites in the UK working within multi-disciplinary teams.

The overall aim of the current study is to evaluate the impact of this service reorganization on the standard of care currently provided for, and the outcomes achieved in, children with cleft lip and palate in the UK. Centralisation of care in a smaller number of larger centres has important resource and logistic implications for both users and service providers and this study will attempt to estimate the cost and cost effectiveness of the changes in cleft care. The work is funded by a NIHR grant “Evidence based health care for major congenital and acquired problems of the head and neck”, Reference Number: RP-PG-0707-10034.

OBJECTIVES

1. To describe the care currently provided for five year old children with UCLP and to compare this to that care originally described in the CSAG survey.
2. To describe the outcomes in five year old children with UCLP and compare this to the outcomes described in the original CSAG survey.
3. To estimate the economic impact of the centralisation of cleft care services.
4. To estimate the personal costs to parent/guardian and families of children with UCLP.
5. To quantify preferences for different attributes of centralised cleft care from the perspective of parents/guardians of children born with UCLP.

RESEARCH MANAGEMENT

DETAILS OF SPONSOR

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SAFETY REPORTING

As this is an observational study we are only collecting adverse events directly related to study related procedures according to U H Bristol Research Adverse Event Policy.

MONITORING AND AUDIT

The study will be monitored and audited in accordance with Trust policy. All study related documents will be made available on request for monitoring and audit by UH Bristol and the relevant Research Ethics Committee.

DATA PROTECTION

Data will be collected and retained in accordance with the Data Protection Act 1998.

STORAGE OF RECORDS

Study documents (paper and electronic) will be retained in a secure location during & after the study has finished. At the end of the study, all paper study documents will be scanned and then electronically stored on a NHS secure encrypted hard disk. All source documents will be retained for at least 25 years following the end of the study.

INDEMNITY

This is an NHS-sponsored research study. For NHS sponsored research HSG(96)48 reference no. 2 refers. If there is negligent harm during the study when the NHS body owes a duty of care to the person harmed, NHS Indemnity covers NHS staff, medical academic staff with honorary contracts, and those conducting the study. NHS Indemnity does not offer no-fault compensation and is unable to agree in advance to pay compensation for non-negligent harm. Ex-gratia payments may be considered in the case of a claim.

ETHICS & R&D APPROVALS

The study will be performed subject to Research Ethics Committee (REC) approval, including any provisions of Site Specific Assessment (SSA), and local Research and Development (R&D) approval.

RESEARCH GOVERNANCE STATEMENT

This study will be conducted in accordance with the Research Governance Framework for Health and Social Care and Good Clinical Practice.

CONSULTATIONS

COMPLETED

Cleft Research Workshop	26 – 27February, 2009
Craniofacial Society of Great Britain and Ireland Conference	25 -27 April, 2009
Frenchay Cleft team Audit day	2 July, 2009
Speech and Language meeting with Liz Albery, Debbie Sell and Sue Mildinhall.	7 August, 2009
Frenchay Meeting Liz A and Richard W	18 August, 2009
Frenchay Meeting Psychology Tina Owen and Julia Cadogan	25 August, 2009
Cleft Genetics Sarah Smithson. St Michaels Hospital	7 September, 2009
Frenchay Meeting Psychology Tina Owen	17 September, 2009
Psychology SIG meeting, Newcastle	22 September, 2009
Cleft Clinical Directors and Cleft Service Managers SIG meeting, Birmingham	2 October, 2009
ENT Meeting GOS Tony Sirimanna and Debbie Sell	6 October, 2009
Cleft Nurse Meeting Cathy Marsh, Frenchay	19 October, 2009
Cleft Net East conference call	3 November, 2009
Deborah Franklin, Paediatric dentistry	5 November, 2009
Paediatric dentistry SIG meeting (Deborah Franklin)	10 November, 2009
North West/N Wales conference call	11 November, 2009
Medical Photography Frenchay	12 November, 2009
Trent Cleft centre conference call	16 November, 2009
Northern and Yorks conference call	18 November, 2009
West Midlands Cleft Centre conference call	24 November, 2009
Leeds Cleft Centre conference call	25 November, 2009
Spires Cleft Centre conference call	27 November, 2009
Belfast Cleft Centre conference call	27 November, 2009
Lead Cleft nurse meeting Manchester	2 December, 2009
Orthodontist SIG meeting	4 December, 2009
Visit Belfast Cleft Centre	17 December, 2009
Visit Cleft Net East	5 January, 2010
Psychology SIG meeting	12 January, 2010
Swansea Cleft service conference call	13 January, 2010
Visit Leeds Cleft Centre	21 January, 2010
Visit Midlands Cleft Centre	29 January, 2010
Visit North Thames Cleft Service – Chelmsford	2 February, 2010
Visit North Thames Cleft Service – GOS	17 February, 2010

Visit South Thames Cleft Service	1 March, 2010
Visit Edinburgh Cleft Service	2 March, 2010
Cleft workshop 2010	4-5 March , 2010
Visit Swansea Cleft Service	12 April, 2010

PLANNED

Visit Oxford Cleft Service	23 April, 2010
Visit Northwest and North Wales Cleft Centre	27-28 April, 2010
Visit Salisbury Cleft service	7 May, 2010
Visit Trent Cleft Service	24May, 2010

We plan to visit all Cleft centres before the CSAG II commences on the 1st of July 2010.

TIME TABLE

Draft protocol for circulation	December 2009
Identification of eligible cases	November 2009- January 2010
Visits to cleft centres	December 2009 – June 2010
Submit ethics application	April 2010
Meet with R&D contacts (support costs)	January - March 2010
Staff training days for research clinic	January -June 2010
Cleft workshop	4-5 March 2010
5 year old survey	July 2010 – December 2012

ASSESSMENT OF PREFERENCES FOR DIFFERENT ATTRIBUTES OF CENTRALISED CLEFT SERVICES

RATIONALE

In the past few years, the NHS has been increasing its efforts in raising patient and public involvement in the delivery of health and social care services. As the process of centralisation of cleft services throughout the U.K. has developed at different rates and different models of centralised care have emerged, one of the ways to evaluate the re-organisation of this service is to quantify the relative value of different attributes of this service from the perspective of parents/guardians of children with unilateral cleft lip and palate (UCLP). The data collected would provide useful information in the delivery of cleft services to optimise outcomes within a given budget.

A discrete choice experiment (DCE) using best-worst scaling (15) will be used to estimate the relative value of different attributes of centralised cleft care services. This method is made up of five stages (16). The first stage involves the identification of attributes and this will be done through a review of the literature, and semi-structured qualitative interviews with parents/guardians of children born with UCLP. The second stage entails the assignment of levels to the characteristics identified which should be plausible and actionable, and this will be explored with parents/guardians during the semi-structured interviews. Where applicable, local and national standards, e.g. with reference to waiting times, will be incorporated into the development of levels for each attribute. The third stage involves the choice of scenarios that would describe the possible service (or outcome) configurations based on the attributes and

levels chosen. An experimental design will be used to establish a manageable number of scenarios. The last two stages cover the collection of data and data analysis.

STUDY SAMPLE

Preliminary semi-structured qualitative interviews

Potential study sample will be parents/guardians of children with UCLP. These parents/guardians will be purposively selected with the aim of sampling for maximum variation in terms of their geographical location within the UK, and travelling distances between their homes and the cleft service administrative sites where their children receive care. Sample size for this part of the study will be determined by data saturation.

Discrete Choice Experiment (DCE)

Parents/guardians of children with UCLP will be asked to take part in the DCE. It is likely that there will be at least two potential subgroups of parents/guardians who may have different preferences: those who live within a short distance from their local cleft centre (i.e. within 30 miles), and those who live a long way from their local cleft centre (i.e. exceeding 30 miles). This will be confirmed through the interviews with parents/guardians. Other possible subgroups will be explored through the same semi-structured interviews. The sample size for each subgroup in a DCE is usually set between 30 and 100 individuals (16). The aim will be to achieve, on average, a sample of 30 individuals per subgroup (i.e. at least 60 participants in total or more if more subgroups were identified). Recruitment of parents/guardians will continue until at least 60 (or more if necessary) completed responses have been achieved.

SETTING

Preliminary semi-structured qualitative interviews

Given that participants will be drawn from across the UK, it will be most practical to conduct these interviews by telephone.

Discrete Choice Experiment

The DCE questionnaire will be distributed in two ways:

a) For parents/guardians of children already recruited into the study and who have consented to be contacted again, the questionnaire will be posted to them.

b) For parents/guardians of children who will be prospectively recruited into the study, they will be asked at the end of the clinic audit day if they would like to take home and complete the questionnaire (together with the Health and Lifestyle questionnaire).

IDENTIFICATION OF ELIGIBLE PARTICIPANTS

Preliminary semi-structured qualitative interviews

A request will be sent to each clinical director for their help in providing a potential list of four parents/guardians: two who live near to the cleft service administrative site, and two who live a long distance away from the site. These would be parents/guardians of 5 year old children who have already attended the audit clinics at their local cleft centre for the time period 01/09/09 – 31/05/2010 and consequently is not part of the CSAG II project.

Discrete Choice Experiment

Both parents/guardians of children already recruited into the study and who have consented to be contacted again as well as those who will be prospectively recruited into the study will be invited to take part.

INCLUSION CRITERIA

For both parts of the research, informed consent to participate is essential. In addition,

a) For the preliminary semi-structured qualitative interviews, parents/guardians of children with UCLP who have already attended the audit clinics at their local cleft centre when they were 5 years old will be included into the potential list of participants.

b) For the DCE, parents/guardians of children with UCLP who have or are attending the audit clinic at their local cleft centre when they are aged 5 years old will be invited to participate.

EXCLUSION CRITERIA

The aim is to obtain a wide-ranging perspective from parents/guardians and hence no exclusion criteria have been set. Individual cases will be checked for suitability to participate based on the inclusion criteria.

PROCESS

Preliminary semi-structured qualitative interviews

An email will be sent to all clinical directors requesting each of them to provide a potential list of four parents/guardians: two who live near to the cleft service administrative site, and another two who live a long distance away from the site.

Initially, ten parents/guardians from each of the two subgroups will be purposively selected and contacted by letter (together with an information leaflet, a consent form and a reply slip) (Appendices 2 to 5) asking for their consent to participate in the research. These letters will be sent via the hub centre. Upon receipt of signed consent forms, a member of the research team

will contact each individual by telephone to arrange for a convenient time to conduct the semi-structured interview by telephone.

If, after contacting all the parents/guardians on the initial list, no parents/guardians agree to participate, the cleft centres will be approached again for help to provide further names of parents/guardians.

Once all interviews, which will be taped, have been conducted, the qualitative data will be transcribed and analysed in order to develop the DCE questionnaire.

Discrete Choice Experiment

i) Parents/guardians of children already recruited into the study and who have consented to be contacted again:

A copy of the DCE questionnaire together with a cover letter and an information leaflet will be posted to them. They will be asked to complete the questionnaire and mail it back to the researchers within a week. If the questionnaire is not returned within 7-14 days, the research team will mail out another questionnaire together with a FREE POST envelope asking them to complete the questionnaire. Half of the group will receive version 1.5a of the DCE questionnaire and the other half will receive version 1.5b of the questionnaire.

ii) Parents/guardians of children who will be prospectively recruited into the study

Currently, this group of parents/guardians are being asked at the end of the research audit clinic day if they would like to take home and complete a health and lifestyle questionnaire. The DCE questionnaire will be added to this to form a questionnaire pack. If they agree to complete the questionnaire pack, they will be given the pack together with a information leave and FREE

POST envelope. They will be asked to return the completed questionnaires to the researcher within one week. If the questionnaires are not returned within 7-14 days, the research team will mail out another questionnaire pack together with a FREE POST envelope asking them to complete the questionnaires. Half of the group will receive version 1.5c of the DCE questionnaire and the other half will receive version 1.5d of the questionnaire.

The complete version of the DCE questionnaire contains 17 choice questions of which 16 are derived from a fractional factorial experimental design and one is a test for internal consistency. In order to minimise participant's fatigue due to having to complete 17 choice questions, the questionnaire has been divided into 2 versions (see DCE questionnaire version 1.5a and version 1.5b). Each version is made up of 9 choice questions, i.e. half of the 16 derived from the experimental design and the test for internal consistency question. Version 1.5a will be abridged to exclude some background questions that have already been included in Health and lifestyle questionnaire and become version 1.5c. Likewise, version 1.5b will be abridged and become version 1.5d.

OUTCOME MEASURES

Data from the DCE based on best-worst scaling will be analysed using weighted least squares. The results will demonstrate the relative value of the different attributes that parents/guardians of children born with CLP place on cleft services.

METHODS FOR 5 YEAR OLD SURVEY

RATIONALE

This survey follows the protocol used in the original CSAG survey which attempted to locate and study all children born with non-syndromic UCLP in defined periods. In addition, the survey will also examine if children with cleft have the same growth development as children without a cleft and examine the psychosocial predictors of outcome such as social and family circumstances, parenting and parental characteristics and child characteristics.

To ensure that we are comparing a similar group of children in the UK, the current study is an observational cross-sectional study of 5-year-olds with UCLP. It is not necessary to survey all children in the UK born with some form of clefting as several multi-centre comparisons have provided evidence that care for and outcomes in, UCLP cases are representative of cleft care across the UK. All of the 5-year-olds born with UCLP in the current study will have been treated within a centralised or centralising service. By the age of 5, children in the UK will have started their formal education and will be speaking to people they are unfamiliar with. Centralized multidisciplinary cleft care teams throughout the U.K. have developed at different rates and have differing models of centralized care. This will allow us to compare differences in the timing of centralisation and a number of different models of care.

STUDY SAMPLE

We aim to study all 5-year-old children from cleft centres in the UK born in the two year period between **01/04/2005 to 31/03/2007.** At the time of the audit the child should be aged between 5 years 3 months and 5 years 9 months. The original CSAG survey identified 326 five-year-olds born with UCLP in the UK over a two year period and outcome records were collected

for 239 five-year-olds (1). This represents a recruitment rate of 73%. As care is now centralised and better documented we anticipate that we will be able to obtain information from at least 250 patients born with with UCLP.

SETTING

The majority of children will be seen in research audit clinics run by the cleft centres. Families who would usually attend outreach clinics will be encouraged to come to the central research audit clinic (travel expenses will be offered to cover the cost of attendance). Where this is not possible the children will be assessed in outreach clinics.

IDENTIFICATION OF ELIGIBLE PARTICIPANTS

The cleft centres in the UK will be asked to identify eligible participants by using the inclusion and exclusion criteria. As a cross check, eligible participants will also be identified through the CRANE database. The data manager from the CRANE database will send each cleft centre a list of eligible participants based upon the inclusion criteria, the cleft centre will use this list to validate their own records.

INCLUSION CRITERIA

5 year old children with complete unilateral clefts of the lip and palate, including any with soft tissue bands of less than 5mm, born during the two year period **01/04/2005 to 31/03/2007.** The children will be seen between ages 5.3 to 5.9 years. If a child fails to attend the initial scheduled research audit clinic, they will be invited to attend the subsequent research audit clinic up until the age of 6.5 years. The cleft centres will provide a list of eligible participants with the following indicators: a) year of birth b) sex

EXCLUSION CRITERIA

Children whose developmental delay or syndrome is sufficient to prevent them from co-operating with procedures for data collection, such as;

- No speech
- No (or very little) comprehension
- Other criteria checked on a case by case basis agreed with research team

It is thought important that we do not exclude cases that might have been included in the original CSAG survey. Therefore, we should aim to be as inclusive as possible with full documentation on each case; thus any case that is questionable should be discussed between the cleft centre and the research team and if deemed unsuitable for inclusion, the case will be omitted from the data analyses. Cleft centres will be asked to make available clinical photographs of children who are excluded on the grounds of having soft tissue bands of more than 5 mm.

The cleft centres will also provide a list of the excluded children for the research team for verification that will contain the following details:

Year of birth	**Sex**	**Reason for exclusion**

PROCESS OF RECRUITMENT

PREPARATION FOR THE RESEARCH AUDIT CLINICS

1. A clinic coordinator will be identified from each cleft centre to liaise with the research team regarding the arrangements for jointly agreed data collection days.
2. Before the clinic visit, at each cleft centre, the clinic coordinator together with the clinical director and the research team will identify the assistance required by the cleft team in order to achieve intended outcomes from the research audit clinic:
    a. Key personnel will be identified for each of the stations in the research audit clinic.
    b. Facilities will be arranged for the research audit day.
3. Each cleft centre will identify eligible participants at their centre using the inclusion and exclusion criteria. They will then agree dates for clinics and numbers likely to attend each of these clinics. This will be verified by comparison with a list extracted from the CRANE database.
4. The Cleft centre will send out an invitation to eligible participants to attend a research audit clinic. All invited parents/guardians and children will receive an information letter and information sheet about the research explaining the purpose of the study and what will happen during the research audit clinic day. Participation in the study will be completely voluntary .
    a. Information letter to the parent/guardian (Appendix 9)
    b. Information sheet to the parent/guardian (Appendix 10)

5. Each child's parent/guardian should also be contacted by telephone by the clinic coordinator during the week before the data collection day to confirm that they will be able to attend and ask if they have any questions about the research. Children who are unable to attend on the date offered will be offered an alternative date.
6. The cleft centre will provide the research team with a list without personal identifiers of the eligible participants that have been scheduled for the research audit clinic.
7. The Research Team will prepare the following for each participant;
    a. Route map to the child, which explains in pictures what will happen during the day. (Appendix 11)
    b. A "goody bag" for each child containing a small colouring book, colour pencils, a biro for the parent, a small teddy bear.
    c. A thank you letter to parent/guardian (Appendix 12)
    d. A consent form (Appendix 13)
    e. An individual envelope containing:
        i. Labelled data capture forms
        ii. A "satisfaction with service" questionnaire for completion by parent/guardian.
        iii. A "health and lifestyle questionnaire" for completion by parent/guardian.
        iv. A labelled plastic bag and lab-card for impressions or box for duplicated models and wax bite.
        v. A labelled free post envelope for its return.

PROCEDURE ON THE RESEARCH AUDIT CLINIC DAY

1. A member of the research team will meet with the cleft team before the research audit clinic starts to review the arrangements for the day and deal with any last minute issues or concerns.
2. The research team, when possible, will display signs for the different “stations” where the child will be examined. The signs will match the figures in the child’s folder: “Listening test”, “Speaking test”, “Moulds”, “Dental Check-up”, “Height and weight” and “Photos”.
3. The research team will prepare the the following for the research clinic audit.
    a. Assembly of the measuring station.
    b. Ensuring that all adminstrative material is in order.
    c. Providing a box for parent/guardian for the anonymous return of the questionnaire on satisfaction of service .
4. On the day of the scheduled audit, the parent(s) and their child will be asked if they are willing to participate in the study and whether they have any questions. The questions will be answered by a member of the research team. If the parent refuses to participate in the study, they will be informed that data obtained during the clinical examinations will only be stored in their medical file and that researchers will have no access to them. They will also be informed that their decision will have no impact on their current or future clinical care. If it is not possible for the research team to attend, the clinical team will seek and obtain consent.

5. Eligible parent/guardian will be able to sign the consent form and the child will be asked for verbal consent.
6. The research team will prepare the identification labels for the forms used at the different stations.
7. Each child will be given their own personal envelope folder together with a “goody bag” by a member of the research team or clinical team. The folder will contain the “route map” (Appendix 11) showing the different stations they will visit during the day. The route map will be explained to the parent and the child by a member of the research team or clinical team.
8. Children will visit each station with their parent/guardian. Once the child has had records taken the appropriate member of staff should sign the “route map” provided in the folder before the child proceeds to the next station. The parent or child can withdraw from any section/part of the clinic at any time.
9. When all the measurements have been taken, the research staff will collect the envelope folder from the child and check that all sections of the "route map" have been signed. They will also check that all records have been collected and that the relevant data capture forms are stored in the child's individual folder.
10. The parent will be asked to complete the satisfaction of service questionnaire, they can complete the questionnaire at the centre by dropping it in the questionnaire box provided or they can mail it back in the free post envelope provided to them. A member of the research team or clinical team will also provide each set of parent/guardian a labelled copy of the health and lifestyle questionnaire together with a free post

envelope. Parent/guardian will be asked to complete the questionnaire in their home environment and send it back to the researchers in the prepaid envelope.

11. The child will then be given a “small teddy bear” and their parent/guardian will be given a letter thanking them for attending.
12. The Research Team will collect all participants’ folders at the end of each research audit clinic together with the research questionnaire; audio and video tapes or digital recordings; photographs, impressions for study models or study models and data capture forms. The research team will be responsible for the correct labelling of all data forms and items.

DATA COLLECTED FROM THE CLINICAL RECORDS AND RESEARCH AUDIT CLINIC

SPEECH ASSESSMENT

Method

Each centre will nominate one speech and language therapist (SLT) to be the designated contact person for the study at each surgical site

1. In advance, this SLT will identify the locations and therapists who have reviewed or provided treatment for the child. The SLT will complete the therapy background information form using information in the medical records and the team based speech and language therapy notes, and by liaison with local services when appropriate.
2. At the clinic, parent/guardian will be asked to confirm details at the time of the audit on the speech treatment history which has been collected on the therapy background information form.
3. For the assessment, speech audio video recordings will be collected on all subjects using the equipment, procedures and speech sample as described by Sell et al., 2009 (17) (or its equivalent) from the 5-year-old study cohort. All the recordings will be made by one of the centre based SLTs, who has been Cleft Audit Protocol for Speech-Augmented (CAPS-A) trained. Recordings are to be made in a quiet room with the subject facing natural light if possible. Each child will sit in front of a pale grey background, supplied by the research team. The microphone will be placed on a stand, 23 -30 centimeters away from the child, at the level of their mouth and placed to one side. The face and upper neck will be framed in the picture. The speech sample material will be presented beside the camera at the subject's eye level. Following data collection the SLT will check, using

the headphones, that a high quality sample has been recorded. The SLT will document the tape number on the Therapy Background Information Form.

The speech sample will include:

1) A sample of 2 minutes of conversation: according to Sell et al 2009 (17). Encouraging open-ended questions through a progression of questions on a particular topic is advised.
2) Counting from one to twenty and from 60 to 70.
3) Saying (not singing) a nursery rhyme e.g. Jack and Jill.
4) Repetition of each of the 16 sentences after the therapist using the Great Ormond Street Speech Assessment (GOS.SP.ASS.) picture book.

Equipment

- Therapy Information Form (Appendix 14)
- The Great Ormond Street Speech Assessment (GOS.SP.ASS.) picture book.
- Video cameras with microphone, microphone stand, headphone, spare batteries, tripod, pale grey background and means to hang, DV tapes, sticker for reward, sticker to mark completion of station to be inserted in child's route map
- The research team will copy the recording in accordance to the prior arrangements established with each cleft team. The master copies will remain at the Centre and the copies will be passed to the research team.

Personnel

The speech and language therapist at each cleft centre. The Speech Steering Group (Liz Albery, Sue Mildinhall, Debbie Sell) together with the research team will advise on data collection and through their networks ensure all the centres are clear with regards to the protocol. Speech

outcome data at the time of the assessment will be collected by the local team according to their local protocol. For example some centres use the Great Ormond Street Speech Assessment (GOS.SP.ASS.) clinical tool at this assessment for the purposes of collecting comprehensive clinical data (18, 19). Others use CAPS-A as the speech audit tool (20).

For this outcome study, the process of analysis will be conducted once all the data from all the centres is collected. All the samples will be placed in a randomised order onto DVDs. Analysis will be undertaken by independent listeners. 10% of the cases will be analysed for inter- and intra-rater reliability.

OUTCOME MEASURES FROM CLINICAL RECORDS AND RESEARCH AUDIT CLINIC

- Assessment of, history of and current speech and language therapy intervention in 5-year-olds, including: patterns of SLT delivery, availability of speech therapy treatment when required, analysis of amount of SLT contact time, methods of SLT delivery, foci of therapy and assessment of residual needs for speech and language therapy (numbers that require SLT intervention, but have not received it or have received insufficient therapy).
- Nasality (hypernasality, hyponasality), Nasal airflow (nasal emission, nasal turbulence), Speech Cleft Characteristics, Developmental Immaturities.

AUDIOLOGY ASSESSMENT

Method

An Audiologist or Audiological Physician (including a Paediatrician with a special interest in Audiology) / ENT surgeon will conduct the data-collection at each research audit clinic.

1. The audiologist and/or member of the research team will examine the clinical records (prior to the audit clinic) and record information on the data capture form.
2. Parent/guardian will be asked to confirm details of previous ENT assessments and episodes of treatment, including the insertion of grommets, undertaken for their child.
3. Standardised Pure Tone Audiometry thresholds (BSA guidelines) using an audiometer and a tympanometer will be undertaken for each participant by the local Audiologist.

Equipment

- Each Audiologist will be asked to provide a standard calibrated audiometer and a tympanometer.
- Stage A calibration checks will be carried out on testing equipment as per standard national practice
- Data capture form (Appendix 15)
- Sticker to mark completion of station, to be inserted in child's route map

Personnel

The local Audiologist, Audiological Physician or ENT surgeon.

OUTCOME MEASURES FROM RESEARCH AUDIT CLINIC AND CLINICAL RECORDS

- Record Audiological/otological interventions carried out
    - Grommets
    - Hearing aid use past/current

- Management of middle ear effusion to include watchful waiting
- Other medical/Otological procedures

- Audiological/ontological assessment in 5-year-olds
  - Full audiogram testing AC and BC as appropriate to assess hearing thresholds rather than using screening audiometry
  - Type of hearing loss
  - Degree of hearing loss
  - Management of hearing loss as recorded on the audit assessment day
  - Middle ear function assessed by tympanometry

ASSESSMENT OF DENTAL ARCH RELATIONSHIP

Method

1. The cleft team orthodontist (or appropriately trained dentist/dental care professional) will complete the presurgical orthopaedic treatment form and will take an alginate impression together with a wax squash-bite in centric occlusion. The overjet will also be recorded. If impressions cannot be obtained then intra oral photographs of the teeth in occlusion will be obtained.

    a. Intra-oral photographs should include a frontal view, right and left lateral views and, if possible, a palatal view.

    b. The orthodontist (or appropriately trained dentist/dental care professional) should ensure that the patient is in the correct occlusion when taking the photographs.

    c. The orthodontist (or appropriately trained dentist/dental care professional) should check that the photographs are adequate to assess crossbites and incisor inclination prior to allowing the patient to leave

    d. A clinically recorded overjet measurement for each anterior tooth should be taken at that same appointment and should be provided with the photographs'

The research team will either

    a. receive a copy of the trimmed models made from the impressions

    b. take the impressions with them to make a trimmed model for the research team and a copy model for the cleft centre

c. The research team will need to receive a copy of the photographs when these have been taken in place of the models.

2. The model will be placed in model box for transportation or the impressions will be placed in labelled plastic bags with lab cards and transported by the research team in a "cool bag" to the laboratory.

3. At the laboratory, the impressions will be cast and models will be prepared by an orthodontic technician in the laboratory according to an established protocol and the bases will be labelled with the participants ID number. For those cleft centres that provided an impression a duplicate set of trimmed models will be sent to them. If models are provided by the cleft centres then these will be duplicated so that all casts and models will be in a standard format when evaluated as an outcome measure.

Equipment

- Presurgical orthopaedic treatment form (Appendix 16)
- Sets of small trays suitable for use in five-year olds
- Wax for squash bites
- Cool box to transport the impressions, plastic bags, identification labels
- Alginate impression material, fixative & disinfectant
- Mixing bowls and spatulas
- Sticker to mark completion of station, to be inserted in child's route map
- If appropriate an alginate impression of the child's finger may be taken and cast in plaster as a small memento of the child's visit.

Personnel

The impressions will be taken by the orthodontist (or appropriately trained dentist/dental care professional)

A local dental surgery assistant to mix the alginate

Orthodontic technician (will be located in the North Bristol laboratory)

OUTCOME MEASURES FROM RESEARCH AUDIT CLINIC AND CLINICAL RECORDS

- Inter-arch relationships measured with the 5 year old index and potentially the 4 scale index developed in Manchester.

ASSESSMENT OF DENTAL HEALTH

Methods

1. The paediatric dentist and/or therapist will examine the clinical records and record information on the data capture form.
2. The paediatric dentist and/or therapist will ask the parent about the following;
    a. Who provides their regular dental care
    b. Have they been assessed by a Consultant in Paediatric Dentistry?
    c. Has their child ever had a neonatal appliance? (show picture as example)
    d. Has their child ever had nasoalveolar moulding? (show picture as example)
    e. Details of functional problems with fistula if present
3. Using standard protocols and using data collection forms we will record the following:
    a) dmfs for five year olds
    b) Oral hygiene using the Index of Oral Cleanliness (21)
    c) As part of the dental health assessment, the buccal occlusion will be recorded.

Equipment

- Data capture form (Appendix 17)
- Dental mirrors
- Dental chair
- Dental light
- Forms to record buccal occlusion, dmfs and oral cleanliness
- Sticker to mark completion of station, to be inserted in child's route map

Personnel

The Paediatric dentist or therapist will have received a standardised training and calibration for the CSAG II project.

OUTCOME MEASURES FROM RESEARCH AUDIT CLINIC AND CLINICAL RECORDS

- Dentist providing usual care – general dental practitioner or specialist paediatric dentist
- Past dental treatment
- History of neonatal appliance
- Buccal occlusion, dmfs and oral cleanliness
- Presence/absence of a residual fistula

ASSESSMENT OF SURGICAL DETAILS

Method

The Cleft surgeon will examine the child and their clinical record and record information on the data capture forms. The examination will identify if any oral fistula is present and if there are any functional problems involved. The clinical record will provide information about phenotype and treatment.

Equipment

- Data capture form (Appendix 18)

Personnel

The Cleft surgeon.

OUTCOME MEASURES FROM RESEARCH AUDIT CLINIC AND CLINICAL RECORDS

- Type of primary lip repair
- Surgical complications
- Type of palate repair
- Presence of oral fistula

2-DIMENSIONAL ASSESSMENT OF FACIAL AESTHETICS

Method

The assessment of facial aesthetics will follow the recommended guidelines for photography of cleft audit patients (22, 23). The child will be asked to sit on a chair placed 0.5 metres in front of a standardised black or white non-reflective background which has been mounted on a wall, or a plain wall can be used as background. The hair will be arranged to show the entire ear.

The following views will be taken for each child with the lens length fixed to give standardised magnification.

1. Left lateral face (1:8)
2. Right lateral face (1:8)
3. ¾ left lateral face (1:8)
4. ¾ right lateral face (1:8)
5. Facial (1:8)
6. Facial smiling (1:4)
7. Whistling (1:4)
8. Worm's eye view (1:4)
9. Lip & Nose (1:3)

Each photograph will be catalogued and recorded for each patient

Equipment

- Camera Nikon D3s or other appropriate camera
- 105mm macro lens
- Lighting equipment for camera

- Black or white background paper/sheet or plain wall
- Black tack
- Sticker to mark completion of station, to be inserted in child's route map
- Capture form (Appendix 19)

Personnel

The photographs will be taken by the cleft centre medical photography personnel.

OUTCOME MEASURES FROM RESEARCH AUDIT CLINIC AND CLINICAL RECORDS

- Profile and frontal photographs to determine the appearance of the lip, nose and profile of 5-year-olds using the Five Point Scale (24)

ASSESSMENT OF HEIGHT, WEIGHT AND HEAD CIRCUMFERENCE

Method

Height

Position - Remove shoes, stand the young person with feet flat, so that underside of heels are in contact with the ground. The heels are placed together, so that the medial malleoli are touching, (unless the child has knock knees). The child stands straight so that heels, calves, buttocks and shoulders are in contact with the vertical backboard of the stadiometer. Shoulders should be relaxed and sloping forward in a natural position, hands and arms are loose and relaxed with palms facing medially. The head is positioned in the Frankfurt plane*.[*Frankfurt plane - the lower orbit of the eye in the same plane as the upper margin of the external auditory meatus.]

Measuring - Slide the headboard down the backboard until it touches the young person's head. To ensure that the head stays in contact with the headboard and to minimise the effect of hair thickness, place a 1kg weight on the headboard. Ask the young person to "relax the shoulders and stretch up but keep the heels on the ground". Assist this stretching by applying gentle upwards pressure beneath the mastoid processes. Check that the heels are touching the ground. Read the height from the counter to the last completed millimetre and record on the data sheet

Weight

Ask the young person if they have passed urine before coming to the measuring session. Record on the data sheet. Enter either MALE or FEMALE STANDARD for the young person depending on the gender, then enter the young person's height in centimetres to the nearest centimeter (up to point 4 is rounded down and point 5 and above is rounded up). The young

person, undressed to underwear / lightweight sports clothes, then steps on to the measuring platform, which has been wiped with disinfecting alcohol. Position young person in such a manner, that both feet are located in parallel with the toe and heel in contact with their respective electrodes. Encourage the young person to stand still, front facing without bending the knees.

Measuring - Measurement has been completed when the weight and fat ratio reading are fixed and the buzzer beeps. Ask the young person to step off the measuring platform. Remove the printout from the printer, and clip it to the data sheet. Record the young person's weight, impedance, fat percentage and TBW on the data sheet.

Head Circumference

Position with the young person sat comfortably and relaxed with head in the Frankfurt plane.

Measuring - The head is measured at the widest horizontal circumference above the eyebrows and ears; the tape is kept taut but not tight.

Equipment

- Harpenden Stadiometer
- Harpenden anthropometric tape
- Tanita scales
- Height, Weight and Head circumferences report form (Appendix 20)

Personnel

The research staff together with cleft nurse/clinic nurse

OUTCOME MEASURES FROM RESEARCH AUDIT CLINIC AND CLINICAL RECORDS

- Height
- Weight
- Head Circumference

ASSESSMENT OF PSYCHOSOCIAL STATUS

Method

The parents/guardians of all children will be able to complete the psychology questionnaire that addresses psychosocial status. This will be achieved in one of the following ways;

1. A cleft team psychologist or a member of the research team will administer the questionnaire to the parent.
    a. Self completion
    b. Psychologist assists with the questionnaire
2. The psychology questionnaire will be sent home beforehand, at the clinic the psychologist will collect the questionnaire and address any aspects if needed.

Equipment

- Copies of the psychology questionnaire (Appendix 21)
- Biros
- Envelopes

Personnel

Local cleft team psychologist or research team member experienced in counselling

OUTCOME MEASURES FROM RESEARCH AUDIT CLINIC AND CLINICAL RECORDS

- Assessment of parental answers to the psychology questionnaire
    - Parent/guardian' perception of the impact of the cleft for their child.
    - Parent/guardian' perception about their child's appearance.

ASSESSMENT OF SATISFACTION OF SERVICE

Method

The parents/guardians of all children will be given the questionnaire "satisfaction of service" together with a free post labelled envelope at the end of the research audit clinic day. They will be asked to complete the questionnaire, either to return it to the questionnaire box or mail it back to the researchers within a week. If it is not returned within 7-14 days, the research team will mail out questionnaire together with a free post labelled envelope asking them to complete the questionnaire.

Equipment

- Copies of the satisfaction of service questionnaire (Appendix 22)
- Biros
- Free post labelled envelopes

Personnel

Research team member

OUTCOME MEASURES FROM RESEARCH AUDIT CLINIC AND CLINICAL RECORDS

- Assessment of parental answers to the satisfaction of service questionnaire.

HEALTH & LIFESTYLE QUESTIONNAIRE

Method

The parents/guardians of all children will be asked at the end of the research audit clinic day if they would like to take home and complete a health and lifestyle questionnaire. If they agree, they will be given the "health and lifestyle questionnaire" together with an information sheet and a free post labelled envelope. They will be asked to complete the questionnaire and mail it back to the researchers within a week. If it is not returned within 7-14 days, the research team will mail out questionnaire together with a free post labelled envelope asking them to complete the questionnaire. The questionnaire focuses on a variety of psychosocial aspects (25-28) and personal costs concerned with caring for a child with a cleft lip and / or palate.

Equipment

- Health and Lifestyle questionnaire (Appendix 23).
- Separate versions of the questionnaire will be provided for completion by the child's mother and father and will be identified accordingly.
- Each questionnaire will be labelled with the child's identification number.

Personnel

The study team member who is running the data collection aspect of the audit clinic will be responsible for administering this questionnaire.

OUTCOME MEASURES FROM RESEARCH AUDIT CLINIC AND CLINICAL RECORDS

The questionnaire comprises several parts measuring different aspects of family life and caring for a child, and personal costs incurred by the parent/guardian as result of caring for their child with UCLP. These are as follows:

- A basic demographic section collecting data about:
    - Ethnic group
    - Parent/guardian' age when the child was born
    - Parent/guardian' highest level of education
    - Parent/guardian' current / most recent occupation
    - Family set-up: who lives at home and their relationship to the child
    - Details of other family members with cleft lip / palate ad relationship to the child
- The parent-child relationship
    - Warmth
    - Support
    - Control
- A stress inventory (measuring stress over the previous week)
- A measure of parental protectiveness towards the child
- The General Health Questionnaire
    - Physical and mental health
    - General well-being
- A measure of child temperament
- A measure of child vulnerability
- An estimation of personal direct costs and indirect costs

STATISTICAL ANALYSIS

We will use multiple linear regression, logistic regression and ordered logistic regression where appropriate to compare the outcomes with those observed in 1998 and to control for confounding factors; this will enable evaluation of the impact of centralisation on outcome of treatment for CLP. We will use multi-level models to explore variations in current practice (including areas such as skin closure techniques, antibiotic use and speech and language therapy) and outcomes across centres.

POWER CALCULATIONS

In the baseline survey of outcomes in five year old children with CLP nearly 40% had poor dental arch relations; 40% had untreated dental caries and 19% were judged to be impossible to understand or were just intelligible to strangers. With a sample size of 250 children and setting the (2-sided) value of alpha at 0.05 we will have over 90% power to detect an absolute difference in dental arch relations and caries of 20% and over 80% power to detect an absolute difference in intelligible speech of 10%.

REFERENCES

1. Clinical Standards Advisory Group. Cleft lip and or palate, Report of a CSAG Committee. London1998.
2. Gorlin RJ, Cervenka J, Pruzansky S. Facial clefting and its syndromes. Birth Defects Orig Artic Ser1971 Jun;7(7):3-49.
3. Gregg T, Boyd D, Richardson A. The incidence of cleft lip and palate in Northern Ireland from 1980-1990. Br J Orthod1994 Nov;21(4):387-92.
4. Coupland MA, Coupland AI. Seasonality, incidence, and sex distribution of cleft lip and palate births in Trent Region, 1973-1982. Cleft Palate J1988 Jan;25(1):33-7.
5. Asher-McDade C, Brattstrom V, Dahl E, McWilliam J, Molsted K, Plint DA, et al. A six-center international study of treatment outcome in patients with clefts of the lip and palate: Part 4. Assessment of nasolabial appearance. Cleft Palate Craniofacial Journal1992 September;29(5):409-12.
6. Mars M, Asher-McDade C, Brattstrom V, Dahl E, McWilliam J, Molsted K, et al. A six-center international study of treatment outcome in patients with clefts of the lip and palate: Part 3. Dental arch relationships. The Cleft Palate-Craniofacial Journal1992 Sep;29(5):405-8.
7. Mølsted K A-MC, Brattström V, Dahl E, Mars M, McWilliam J, Plint DA, Prahl-Andersen B, Semb G, Shaw WC, The RPS. A Six-Center International Study of Treatment Outcome in Patients with Clefts of the Lip and Palate: Part 2. Craniofacial Form and Soft Tissue Profile. The Cleft Palate-Craniofacial Journal1992 September;29(5):398-404.
8. Shaw WC, Dahl E, Asher-McDade C, Brattstrom V, Mars M, McWilliam J, et al. A six-center international study of treatment outcome in patients with clefts of the lip and palate. Parts 5. Cleft Palate Craniofacial Journal1992 Sep;29(5):413-8.
9. Murray JJ. Cleft lip and palate services. A review of developments five years after the CSAG report. Int J Paediatr Dent2003 Nov;13(6):395-402; discussion 3.
10. Sandy J, Williams A, Mildinhall S, Murphy T, Bearn D, Shaw B, et al. The Clinical Standards Advisory Group (CSAG) Cleft Lip and Palate Study. British journal of orthodontics1998 Feb;25(1):21-30.
11. Bearn D, Mildinhall S, Murphy T, Murray JJ, Sell D, Shaw WC, et al. Cleft lip and palate care in the United Kingdom--the Clinical Standards Advisory Group (CSAG) Study. Part 4: outcome comparisons, training, and conclusions. Cleft Palate Craniofacial Journal2001 January;38(1):38-43.
12. Sandy JR, Williams AC, Bearn D, Mildinhall S, Murphy T, Sell D, et al. Cleft lip and palate care in the United Kingdom--the Clinical Standards Advisory Group (CSAG) Study. Part 1: background and methodology. Cleft Palate Craniofacial Journal2001 January;38(1):20-3.
13. Sell D, Grunwell P, Mildinhall S, Murphy T, Cornish TA, Bearn D, et al. Cleft lip and palate care in the United Kingdom--the Clinical Standards Advisory Group (CSAG) Study. Part 3: speech outcomes. Cleft Palate Craniofacial Journal2001 Jan;38(1):30-7.
14. Williams AC, Bearn D, Mildinhall S, Murphy T, Sell D, Shaw WC, et al. Cleft lip and palate care in the United Kingdom--the Clinical Standards Advisory Group (CSAG) Study. Part 2:

dentofacial outcomes and patient satisfaction. Cleft Palate Craniofacial Journal2001 Jan;38(1):24-9.
15. Flynn TN, Louviere JJ, Peters TJ, Coast J. Best-worst scaling: What it can do for health care research and how to do it. . *Journal of Health Economics*2007;26(1):171-89.
16. Lancsar E, Louviere J. Conducting discrete choice experiments to inform healthcare decision making: a user's guide. Pharmacoeconomics2008;26(8):661-77.
17. Sell D, John A, Harding-Bell A, Sweeney T, Hegarty F, Freeman J. Cleft Audit Protocol for Speech (CAPS-A): a comprehensive training package for speech analysis. Int J Lang Commun Disord2009;44(4):529-48.
18. Sell D, Harding A, Grunwell P. A screening assessment of cleft palate speech (Great Ormond Street Speech Assessment). Eur J Disord Commun1994;29(1):1-15.
19. Sell D, Harding A, Grunwell P. GOS.SP.ASS.'98: an assessment for speech disorders associated with cleft palate and/or velopharyngeal dysfunction (revised). Int J Lang Commun Disord1999 Jan-Mar;34(1):17-33.
20. John A, Sell D, Sweeney T, Harding-Bell A, Williams A. The cleft audit protocol for speech-augmented: A validated and reliable measure for auditing cleft speech. Cleft Palate Craniofac J2006 May;43(3):272-88.
21. Bearn DR, Aird JC, Jenkins WM, Kinane DF. Index of Oral Cleanliness (I.O.C.). A new oral hygiene index for use in clinical audit. Br J Orthod1996 May;23(2):145-51.
22. Jones M. IMI National Guidelines:Photography of Cleft Audit Patients Institute of Medical Illustrators2004.
23. Jones M, Cadier M. Implementation of standardized medical photography for cleft lip and palate audit. J Audiov Media Med2004 Dec;27(4):154-60.
24. Asher-McDade C, Roberts C, Shaw WC, Gallager C. Development of a method for rating nasolabial appearance in patients with clefts of the lip and palate. Cleft Palate Craniofac J1991 Oct;28(4):385-90; discussion 90-1.
25. Buss A, Plomin R. A temperament theory of personality development. New York: Wiley; 1975.
26. Goldberg D, Hillier V. A scaled version of the General Health Questionnaire. Psychological Medicine1979;9:139-45.
27. Thomasgard M. Parental perceptions of child vulnerability, overprotection, and parental psychological characteristics. Child Psychiatry & Human Development1998;28(4):223-40.
28. Waylen A, Stewart-Brown S. Factors influencing parenting in early childhood: a prospective longitudinal study focusing on change. Child: care, health and development 2009;In press.