British Association of Paediatric Surgeons Congenital Anomalies Surveillance System (BAPS-CASS)

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Guidance for Submitting an Application for Inclusion of a Study

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Author (s)	**Marian Knight**
Approved by	**Simon Eaton**

Version; date	Amendment details	Reason for amendment
1.0 06.03.2013		
2.0 01.08.2013	Alteration to topic proposal form	Inclusion of incidence assessment
3.0 17.10.2013	Alteration to topic proposal form	Addition of contact address

1. Submitting applications to BAPS-CASS

Applications for including a study in the BAPS-CASS programme are considered in two stages. Initial calls for topic proposals are considered and prioritised by the BAPS Research Committee. **Note that individuals submitting topic proposals are not necessarily expected to lead the study if it is prioritised by the Research Committee.** After prioritisation of a topic, the BAPS-CASS Management Group will seek applications from individuals or teams wishing to lead the proposed study. BAPS-CASS staff will provide advice and support for potential lead applicants to complete a full project proposal. Applications from lead applicants are considered by the BAPS-CASS Steering Group which meets every four months. Applicants whose studies are accepted into the BAPS-CASS programme will then be expected to obtain funding and approval for the study from a research ethics committee (REC) before the study can be included on the report cards, and a copy of the REC study approval should be sent to the BAPS-CASS office.

*Note: Application to the BAPS-CASS programme is not a guarantee that the study will be accepted; applicants should **not,** therefore, **seek funding** for the study before their study has been accepted onto the programme.*

2. External Investigators' responsibilities

Investigators whose studies are included on the report cards are expected to fulfil certain undertakings. This reporting system is dependent entirely on the time and effort of reporting clinicians.

2.1.1 Dissemination strategy

It is extremely important that information obtained from BAPS-CASS studies is fed back to clinical staff in a timely manner in order that it may be used to make practical improvements in prevention and treatment of these uncommon conditions and allow for more effective service planning. Each study must therefore nominate a researcher to act as study guarantor who will undertake to make certain that the study results are submitted for publication within two years of completion of data collection, and that reporting clinicians are fully participating in the study. Outside of this time limit, BAPS-CASS reserves the right to analyse and publish the data itself.

2.1.2 Reports to RECs and other relevant bodies:

It is the External Investigator's responsibility to submit relevant reports, data or information to RECs and other bodies, if applicable.

If studies are included into research portfolios, the BAPS-CASS office should be notified. Provision and uploading of accrual data, as required by the portfolio remain the External Investigator's responsibility.

3. BAPS-CASS' responsibilities

In return for the funding, BAPS-CASS will undertake the following:

- Peer review of study proposal by BAPS-CASS Steering Group and assistance with

developing a data collection instrument (Data collection form)

- Assistance with information for submission of application to REC
- Data collection form formatting by NPEU webmaster/ designer
- Study publicity through BAPS-CASS newsletters and mailings to individual reporters
- Collection of case notifications
- Mailing of data collection forms
- Following up of missing forms
- Data gathering, validating and cleaning
- Checking back all data and double-entry into a customised database
- Querying of missing or invalid information
- Provision of an interim dataset as per letter of agreement to enable testing of analysis procedures and abstract preparation
- Provision of a complete, clean dataset to investigators four months after completion of the study
- Provision of mentorship from Steering Group members, if required
- Study results publicity through BAPS-CASS newsletters and other routes as relevant

*Note: BAPS-CASS will function only as a data gathering and cleaning service and under usual circumstances **will not** undertake any data analysis*.

4. Study costs

The full economic costs of each study will be determined using the University of Oxford TRAC methodology before an application is made for funding. The amount needed will vary according to the number of studies in progress at one time, the duration of the study, salary and consumable costs, but as an indication, the full economic costs of study administration are approximately £27,000 at 2012 costs.

5. Criteria for inclusion of studies in the BAPS-CASS programme

Studies are expected to fulfil three or more of the following criteria:

5.1 The condition is an important cause of infant surgical morbidity and/or mortality.

5.2 The condition is an uncommon infant surgical disorder of pregnancy, thus inclusion within the study programme of BAPS-CASS will not impose too great a burden on reporting clinicians [usually no more than approximately 300 cases per year in total].

5.3 The research questions posed by the study can be suitably addressed using the BAPS-CASS methodology (prospective descriptive or cohort studies).

5.4 Other sources of information exist to enhance and/ or assess completeness of data collection.

In addition, the following points will be taken into consideration when applications from potential lead investigators are considered by the Steering Group:

5.5 Evidence of the engagement of the lead investigator/study guarantor with the BAPS-CASS programme

5.6 Evidence of previous multi-centre collaborative work by the lead investigator

5.7 The location of the investigator, in that BAPS-CASS is a multi-centre collaboration and equitable distribution of study leads across centres is an important principle of the collaboration

5.8 The inclusion of appropriate collaborators within the study investigator group. The Steering Committee may recommend conditional inclusion of studies into the BAPS-CASS programme subject to involvement of specific additional co-investigators

5.9 Whether and when previous BAPS-CASS studies have been undertaken into similar or related conditions

5.10 The academic reputation of the investigator in the proposed area of study

6. Study acceptance and rejection

If a study lead investigator is recommended by the BAPS-CASS Steering Group, the recommendation will then be submitted for consideration by the BAPS Executive. Only after endorsement by the BAPS Executive will the study and proposed lead investigator formally be accepted into the BAPS-CASS programme.

Following final acceptance of the study and before commencement the investigator and study guarantor will be asked to sign a letter indicating agreed responsibilities in relation to the project (Appendix 1). **Please note** that final acceptance of the study by the committee does not necessarily mean that surveillance can commence immediately. The timing of study inclusion in the BAPS-CASS programme will depend on relative disease burden and existing conditions under surveillance and commencement may be delayed because of programme balance considerations. The BAPS-CASS Programme Manager will be able to give an indication of a provisional commencement date at the time of acceptance.

Unsuccessful applicants will be sent an initial letter detailing the Steering Group's reasons for their decision. If they wish to appeal against the decision they are requested to write a letter to the Chair of the Steering Group explaining the reasons why they disagree with the grounds for the rejection stated in the steering committee letter. If the Chair feels that these reasons are valid, the project may be brought before the Steering Group for further review.

7. Information and contact details

Preliminary enquiries are welcomed. Please contact:

- Prof Marian Knight (BAPS-CASS Principal Investigator)
  Tel: 01865 289727 Email: marian.knight@npeu.ox.ac.uk
- Melanie Workman (BAPS-CASS Programme Manager)
  Tel: 01865 617774 Email: baps-cass@npeu.ox.ac.uk

8. Completion of Topic Proposal Form

1. Condition to be studied
   Give the full name with any recognised abbreviation.

2. Name of proposer
   Please give the name of the proposer. Please provide a full postal address, telephone number and email address in case of queries.

3. Clinical research questions
   State the aims of the study and list clearly the clinical questions which could be answered by the proposed study.
   For example:
   - What is the current incidence of disease X in the UK and Ireland?
   - What are the prognostic factors for disease X?
   - How does it present?
   - How is disease X managed in the UK and Ireland?
   - What are the outcomes for infants?

4. Incidence estimate
   Please give an estimate of incidence with any potential regional differences, stating the source of the incidence estimate.

5. Background information
   This should include a brief assessment of the state of current knowledge, including any estimates of incidence with any potential regional differences and indicate the need for the study including scientific and clinical importance.

8.1 Submission of Topic Proposal Form

The application form should be completed using no smaller than 10 point type and covering no more than two sides of A4 paper and sent to (please send both electronically and a signed version by post):

BAPS-CASS Administrator
National Perinatal Epidemiology Unit
University of Oxford
Old Road Campus
Oxford OX3 7LF
baps-cass@npeu.ox.ac.uk

BAPS-CASS

TOPIC PROPOSAL FORM

Proposed condition to be studied
Name of Proposer (Include name, address, telephone number and email address)
Research Questions (explain the aims of the study and the questions you think this study might answer)
Estimate of current incidence (please include reference to relevant literature and/or other source of incidence estimate information)
Any other background information (e.g. state of current knowledge, clinical and scientific importance. Please include an estimate of current incidence)

9. Completion of Application to Lead a BAPS-CASS study

1. Title of research
   Give the full title and indicate in brackets a short title for the research of no more than 70 characters.

2. Condition to be studied
   Give the full name with any recognised abbreviation.

3. Investigators
   Please list all investigators and indicate the Principal Investigator, principal contact and Study Guarantor. Please provide a full postal address, telephone number and email address for the principal contact.

4. Background information
   This should include an assessment of the state of current knowledge and indicate the need for the study including scientific and public health importance. This should be in language comprehensible to a lay person.

5. Research questions
   State the aims of the study and list clearly the questions which will be answered.
   For example:
   - What is the current incidence of disease X in the UK and Ireland?
   - What are the prognostic factors for disease X?
   - How does it present?
   - How is disease X managed in the UK and Ireland?
   - What are the outcomes for infants?

6. Case definition
   Please give a clear case definition for the condition of interest, with reference if available, preferably one that is internationally recognised. This definition will be given to BAPS-CASS reporters to enable them to identify the appropriate cases and should be clear and unambiguous with any unfamiliar terms explained fully.

7. Expected numbers
   Please give an estimate of the expected number of cases per year and describe the information on which this estimate was based. Any possible regional differences in incidence should be included.

8. Proposed duration of study
   The normal study duration is 12 months. Give justification if a longer study duration is proposed.

9. Planned methodology
   Indicate whether the planned study is of descriptive or cohort design. Provide a simple power calculation if possible.

10. Alternative sources of information
    Describe any other sources of information (e.g. UKOSS, Binocar) about the disease in question. If other groups of clinicians, e.g. radiologists, pathologists, obstetricians are likely to see cases, it is essential that plans are made to seek cases through them as this improves ascertainment and reduces bias.

11. Funding
    Outline the potential funding arrangements for the project, naming the body(ies) to which applications will be submitted and giving the date by which the outcome of such applications will be known.

12. Additional documents to be submitted
    Attach the CV of the proposed study lead (maximum 2 pages).

BAPS-CASS

FULL APPLICATION FOR INCLUSION OF A STUDY

Title of research
Condition to be studied
Research Team (Indicate Proposed Lead Investigator, study guarantor* and principal contact with name, address, telephone number and email address)
Background (explain the need to study the condition and include state of current knowledge, public health and scientific importance)
Principal research questions

Case definition
Expected numbers (please supply an estimate of the expected number of cases per year)
Proposed duration of study
Planned methodology (descriptive or cohort. Provide a simple power calculation if possible)
Alternative sources of information (Identify any other sources from which information about this condition may be obtained and indicate any plans to use these sources)
Funding (Outline possible funding arrangements and name the bodies to which a grant application will be submitted)

Additional documents to be submitted: Attach the CV of the Study Guarantor (maximum 2 pages).

*The Study Guarantor undertakes to ensure that results of studies are submitted for publication within two years of completion of data collection.

APPENDIX 1: BAPS-CASS Letter of Understanding

The BAPS-CASS Steering Committee has approved the XXX* study and we are looking forward to placing it on the blue card for YY* months. There are some particular points that need to be agreed between the investigator and BAPS-CASS.

Study costs

This BAPS-CASS research collaboration will incur costs to the National Perinatal Epidemiology Unit, University of Oxford as well as to the investigator's home institution. The investigator will be required to obtain funding to cover these costs before the commencement of the study. An NPEU investigator should be named as a collaborator on the application (not necessarily an applicant) in order to allow these costs to be transferred between institutions as appropriate. The BAPS-CASS Principal Investigator will provide letters of collaboration/ support in respect of all applications. In the event of the extension of the study beyond its original agreed period, appropriate additional funding may be required after data collection costs have been reviewed. The study finances will be agreed subject to a formal agreement between the University of Oxford and the investigator's host institution.

Conduct of the Study

Once your study has been approved by the BAPS-CASS Steering Group and endorsed by the BAPS Executive, you must provide documentary evidence of REC approval before the study can commence. It is the responsibility of the investigators to obtain REC approval, but guidance may be obtained from the BAPS-CASS Programme Manager or Administrator regarding elements of the application which relate to the data collection and handling undertaken by BAPS-CASS.

Please note that the timing of study inclusion in the BAPS-CASS programme following REC approval will depend on relative disease burden and existing conditions under surveillance and commencement may be delayed because of programme balance considerations.

The draft data collection form, once approved by the BAPS-CASS Steering Group, will be reformatted to the BAPS-CASS standard design, and should not exceed 4 sides of A4 at this stage.

Reporting Study Findings

The study has been approved by BAPS-CASS because important clinical or public health issues are being addressed and so publication of the findings is expected. All results will be disseminated in accordance with the BAPS-CASS Dissemination Strategy (see section 11 of this document, Appendix 2).

All investigators are expected to provide a short summary of the project progress and outcomes for BAPS-CASS Newsletters. Where appropriate, and with agreement of the chapter authors, summary results will be included in the appropriate chapter of other relevant reports or publications.

Pending submission of the final results for publication, investigators are expected to present the results at appropriate conferences, noting particularly the annual meeting of the British Association of Paediatric Surgeons. In order to facilitate this, investigators will be provided with one interim dataset before study completion. These data will usually be provided four months after the mid-point of the study in order to allow sufficient time for completion of data collection forms but may be provided at a different time if requested to comply with conference

abstract deadlines. To allow for data collection and entry the earliest time at which the interim dataset can be provided is six months after study commencement. Two weeks' notice is required to provide the interim data. Where abstracts are presented, the BAPS-CASS Steering Group should be supplied with a copy of the abstract prior to submission.

Study investigators are expected to submit full results for publication in a peer-reviewed journal within two years of the completion of data collection. The BAPS-CASS Steering Committee should be supplied with a draft copy of any report before submission for publication, and similarly a copy of the final version of the paper upon acceptance. A description of the BAPS-CASS methodology has been prepared and should be used as the basis for the description of the BAPS-CASS methodology in any publication based on the BAPS-CASS system, available via the BAPS-CASS office.

References to all completed studies will be listed on the BAPS-CASS website, and requests for reprints will be addressed to the study investigators where these are received by BAPS-CASS.

Content and title of papers and presentations are entirely at the discretion of the researcher. BAPS-CASS requires its acknowledgement collectively in the list of authors, and all BAPS-CASS reporters should be listed in the acknowledgements. The BAPS-CASS office will provide the relevant list. Researchers will be required to include a disclaimer in any publication stating that the views expressed do not necessarily represent those of the BAPS-CASS Steering Group. The Steering Group retains a right of veto over any publications that it regards to be scientifically unsound. In order to minimise the risk of this happening, the Steering Group will undertake to provide or locate appropriate mentorship for less experienced researchers on request.

When a member of the Steering Group contributes to a project to such an extent that the study or resulting paper could not be completed without the contribution, consideration should be given to the inclusion of that individual as an author on any publication in accordance with existing recommendations regarding authorship of scientific papers.

Each study must nominate a researcher to act as study guarantor who will undertake to make certain that the study results are submitted for publication within two years of completion of data collection. Outside of this time limit, BAPS-CASS reserves the right to analyse and publish the data itself.

Please let me know of any problems that arise and of any other advice or assistance you may require.

Yours sincerely,

Prof Marian Knight
BAPS-CASS Principal Investigator

*To be completed by BAPS-CASS staff upon acceptance of study

BAPS-CASS Letter of Understanding

I/ we* have read and agreed to the conditions outlined in the BAPS-CASS letter of understanding.

*** Please delete as appropriate**

Signed ……

Date ……..

10. APPENDIX 2: BAPS-CASS Dissemination Strategy

1. Background

BAPS-CASS has been developed to address important clinical and public health issues concerning different rare surgical disorders infancy. One of the main aims of BAPS-CASS is to provide information which can be used to improve prevention and treatment of these rare conditions. In order to achieve this aim, appropriate dissemination of study results is key[1]. Additionally, BAPS-CASS relies on clinicians to report cases and complete data collection. It is vital to maintain the enthusiasm for reporting to BAPS-CASS by providing prompt and timely feedback of study results to these clinicians in order that they may use the information gained to help in day to day clinical practice.

2. Dissemination methods

2.1 Studies in progress

Before studies commence, a short article describing the background and aims of the study will be included in the BAPS-CASS newsletter. This newsletter will be distributed to all reporting clinicians, steering group members, BAPS Executive members and will be freely available on the BAPS-CASS website. During data collection, study updates in terms of cases collected will be included in the newsletter.

In order to facilitate testing of analysis strategies and presentation at appropriate scientific conferences, investigators will be provided with an interim dataset to include information on all cases reported before the mid-point of the study. These data will be provided four months after the mid-point of the study in order to allow sufficient time for completion of data collection forms.

2.2 Completed studies

All investigators will be asked to provide a short summary of the outcomes of the project for the BAPS-CASS newsletter in the year following completion of data collection.

Pending submission of the final results for publication, investigators will be encouraged to present the results at appropriate conferences, noting particularly the annual meeting of BAPS. Where such abstracts are presented, the BAPS-CASS Steering Committee should be supplied with a copy of the abstract before submission.

[1] Canadian Health Services Research Foundation. Developing a dissemination plan. Ottawa: CHSRF. Available at: http://www.chsrf.ca/knowledge_transfer/communication_notes/comm_note_dissemination_plan_e.php

Study investigators will be asked to give an undertaking to submit full results for publication in a peer-reviewed journal within two years of the completion of data collection. The BAPS-CASS Steering Group should be supplied with a draft copy of any report before submission for publication, and similarly a copy of the final version of the paper upon acceptance. References to all completed studies will be listed on the BAPS-CASS Website, and requests for reprints will be addressed to the study investigators where these are received by BAPS-CASS.

Where the Steering Group judges it appropriate, in consultation with the investigators and appropriate journal, a news release may be made to the media upon publication of particularly notable results.

3. Adoption

Study investigators will be asked to agree to abide by this dissemination strategy as part of the Letter of Agreement on acceptance of their study.

Marian Knight

6 March 2013