



The BPSU Study Application Handbook - Guide to BPSU Phase 2 study application process

Updated 07.01.19

BPSU parent bodies:



with support from:



Overview

This document provides a step-by-step guide to getting approval for your study from the BPSU Scientific Committee (BSC), the multi-centre research ethics committee (REC), the Confidentiality Advisory Group (CAG) of the Health Research Authority (HRA), the Scottish Public Benefit and Privacy Panel for Health (PBPP) and Social Care and your local NHS Trust Research and Development (R&D) Department.

This guide includes a list of key contacts, abbreviations and a flowchart of the application process. Additional helpful documents which may be found on the BPSU website www.rcpch.ac.uk/bpsu are also referenced in this document.

The Scientific Coordinator and Research Facilitator are the first points of contact for general enquiries, including operational and process matters, meeting dates and press releases. Initial enquiries about undertaking a BPSU study should be directed to the BPSU office.

For advice on the development of an application, such as details of surveillance methodology, ethics or questionnaires, contact should be made with the relevant medical adviser (for communicable or non-communicable diseases) or the scientific coordinator. Contact details for medical advisers can be provided by the BPSU office. Medical advisers correspond with applicants and convey the views of the committee regarding research proposals.

The Chair of the BSC may be contacted directly; however this would not usually be necessary during the course of an application.

Key contacts

BPSU Chair

Dr Shamez Ladhani, Chair of the BPSU Scientific Committee
Email: shamez.ladhani@phe.gov.uk

BPSU Medical Advisers

Dr James Lopez Bernal, Medical Adviser
Email: james.lopezbernal@gmail.com

Dr Jonathan Fok, Medical Adviser (communicable disease)
Email: jonathan.fok@phe.gov.uk

Dr Lamiya Samad, Medical Advisor (non-communicable disease)
Email: l.samad@ucl.ac.uk

BPSU Office

Richard Lynn, Scientific Coordinator
Tel: 0207 092 6173/4 Email: richard.lynn@rcpch.ac.uk / bpsu@rcpch.ac.uk

Abbreviations

BSC	British Paediatric Surveillance Unit Scientific Committee
CAG	Confidentiality Advisory Group
HRA	Health Research Authority
IG Toolkit	Information Governance Toolkit
(M)REC	(Multi-centre) Research Ethics Committee
NRES	National Research Ethics Service
MRC	Medical Research Council
Section 60	Health and Social Care Act 2001 provision for unconsented data use
Section 251	NHS Act 2006 provision for unconsented data use (superseding Section 60)
R&D	Research and Development (Department within NHS Trusts)
IRAS	Integrated Research Application System
PAC	Privacy Advisory Committee (Northern Ireland)
PBPP	Public Benefit and Privacy Panel for Health and Social Care (Scotland)

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1: Introduction - Making an enquiry to the BPSU

Applications for inclusion of a study on the reporting cards are considered by the BPSU scientific committee (BSC), which meets every two months. As the success of the BPSU methodology relies entirely on the willingness of consultant paediatricians to complete and return the monthly Orange Card and study questionnaires - referred to as questionnaires, it is essential that BPSU studies are scientifically robust, adequately resourced and contribute to clinical and public health practice without putting too great a burden on reporting doctors. The application process has been developed to reflect these responsibilities.

This **Phase 2 (P2)** document is to be used once the Phase 1 application has been approved.

The P2 applications should be completed and accompanied by any letters and questionnaires that are to be used in the study. An applicant may be invited to attend the BSC meeting to discuss their proposal and any queries that have arisen.

Unfortunately some applications will be unsuccessful however good the research idea may be. Applications are most often turned down because the BSC considers that the study is not suited to BPSU surveillance methodology.

Important considerations before submitting your Phase 2 application

- In passing to P2 the application would have met the BPSU eligibility criteria.
- Study aims must be appropriate for national surveillance methodology, for example, to establish incidence of a rare disorder or to investigate variations in clinical management.
- Undertaking research without patients' knowledge or consent and in children with rare diseases raises ethical concerns. PPI enables researchers to invite contributions from patient groups and consult on the acceptability of their approach. We would expect that you have identified PPI groups, discussed the study with them and you should, provide a letter of support for your application.
- Applications should reach the BPSU office at least four weeks prior to the BSC meeting to allow the scientific coordinator / medical advisers to comment on the application and revisions to be made prior to committee papers being sent out. Deadlines for forthcoming meetings are available from the BPSU office or on the BPSU website (www.rcpch.ac.uk/bpsu/apply).
- It can take several months to complete the application process as revisions to the methodology and questionnaires are often required. Please make an enquiry directly to the Scientific Coordinator please note our fast-track process; is usually reserved for those conditions considered public health emergencies.
- The study surveillance period is usually 13 months though this can be extended if it is felt that additional case ascertainment is required to address the study objectives.
- There is a contribution charge for undertaking a study through the BPSU. The charge being **£15,000** in the first year (for 13 months of surveillance) and **£10,000** a year for subsequent years, these amounts will be invoiced for at the start of each year of surveillance. Any additional printing required will be charged to the applicant following communication. **Please note the full economic cost of a study is £25,000 and this should be sought when approaching commercial funders.**

2: The BPSU Phase 2 Application

The Phase 2 application

This section gives detailed guidance on how to complete P2 application process. The BPSU Scientific Committee (BSC) will give fair and impartial consideration to the applications. If appropriate, advice from independent referees may be sought. Please note that though your application has moved from a P1, this in no way implies that the study is likely to be accepted at P2. Principal investigators are often invited to attend a meeting of the BPSU SC to discuss their P2 proposal more fully.

When planning your application submission investigators are asked to take into account the following:

- The criteria for study application to the BPSU.
- The process from submission of the P2 to acceptance may take several months. This process can be accelerated for conditions of public health importance which require immediate evaluation.
- The scientific coordinator / research facilitator must receive applications four weeks before the BSC meeting date in order to gather comment on the application.
- Following feedback the BPSU office must receive finalised applications which are ready for submission two weeks prior to the BSC meeting, to allow time to circulate documents for review.
- The BSC meets every two months. Dates are available from the BPSU Office or on the website at www.rcpch.ac.uk/bpsu/apply
- Please read and follow the guidance for completing the application form as failure to do so can delay or even lead to rejection of the application.
- Timing of inclusion of new studies onto the BPSU card depends on the number and the nature of other studies being surveyed.

Outcomes from a Phase 2 Application

The BSC meets five - six times per year to consider applications. The following outcomes are possible:

- 1) P2 may be accepted without revisions or clarifications - unlikely
- 2) P2 accepted but with several minor points needing to be addressed or clarified and usually subject to final approval by chairs action
- 3) Further review, or specialist advice, may be sought and following a subsequent revised P2 to the BSC a final decision is made
- 4) P2 methodology approved but questionnaire or other supporting information such as the public information leaflet needs amending
- 5) The study is rejected

Rejection of an application indicates simply that it is not a suitable application for the BPSU scheme. The BSC will give reasons for its decision and offer suggestions on how the study could be undertaken outside of the BPSU scheme.

Following acceptance of the study proposal and questionnaire at P2, research ethics, CAG, HRA and PBPP approval will be required. Please refer to the flowchart below for details of these processes. If you have any further queries relating to the BPSU application procedure please do not hesitate to contact the medical advisers.

NB: Once a P2 application has been accepted, the BSC expects the study to commence surveillance within 12 months of Chair's approval. If surveillance has not commenced in 12 months of Chair's approval the BPSU Office will be in contact with you to discuss your application and the reasons for the delay. If a satisfactory response is not elucidated the BPSU reserves the right to revoke its P2 approval and the study team will be invited to 're-submit' a P1 application.

The review processes required to undertake a BPSU study

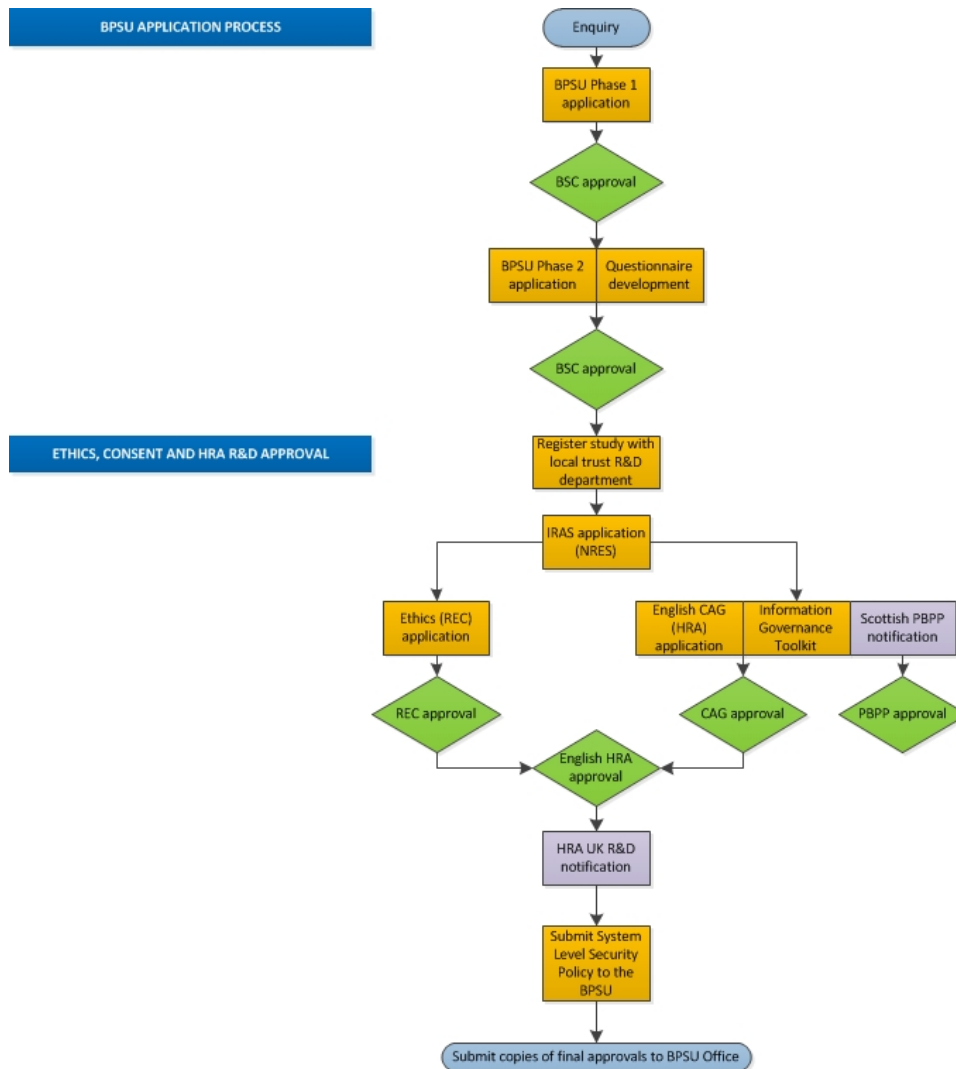


Figure 1: The full BPSU study review process

Guidance on specific questions in the Phase 2 application

1. Title of the study

Please provide the full title of the study. You may wish to provide an alternate brief title if the full title is long or complex, but please avoid using abbreviations or acronyms.

2. Title to appear on orange card

The character limit is 65 for the orange card study title.

3. Investigators

Please list all investigators involved in the study, their contribution to this study, job title, AND affiliation. Please also indicate clearly the principal contact for correspondence on this application, giving a full contact address, e-mail address and telephone number. Please indicate also the individual who is the designated Principal Investigator – this person will be responsible for research governance. At least one of the study investigators should be a paediatrician receiving the orange card.

You should have a named contact in Ireland who can support and promote the study, and provide advice on the suitability of your study methods and questionnaire for Irish paediatricians. The BPSU or Irish Paediatric Surveillance Unit can help you find a suitable contact that has a specialist interest in the condition that you are studying.

4. Lay summary

This should be a short, clear summary of the condition and study in terms that can be understood by a lay person. This will be the publicly available summary that is put on the BPSU website if the study is accepted. The lay summary should be no more than 250 words. Advice on writing a lay summary is available from INVOLVE at www.invo.org.uk/resource-centre/plain-english-summaries/ or contact the BPSU for current examples.

5. Describe the study

This should explain a) the condition to be studied, b) a review of the background to the study proposal, including current knowledge about incidence and prevalence, c) the public health and scientific importance of the study, d) the study methodology, and e) the expected benefits of the study. This explanation should be easily understood by a lay person as the BSC includes lay and medical reviewers.

6. Proposed duration of study

Study's normally run for 13 months; however there will be cases where two or more years of surveillance of a very rare condition may be required to provide adequate cases for the study. Applicants must therefore specify in their P2 application how long they wish to undertake surveillance and subsequent follow-up. Justification for the proposed study duration should be included in the supporting statement. Continuation of surveillance beyond one year is subject to receipt of a yearly progress report.

If the applicants wish to follow-up cases, the follow-up period would normally be for one or two years. If a longer period of follow-up is required then this may be permitted if sufficient

justification is provided. Please note that formal justification for the length of follow-up should be presented to CAG, or equivalent institutions within the devolved nations, whose approval will also be required.

Each investigator must also contribute a short report on their study each year to form part of the BPSU Annual Report. Please note that the BSC has the option to limit initial surveillance duration to 13 months.

7. Case definition and reporting instructions

The *surveillance case definition* defines the cases that you would like clinicians to report. This may be wider than the *analytic case definition* in order to ensure cases are not missed. For example, the surveillance case definition will often include suspected cases where confirmation is awaited. The *analytic case definition* describes very carefully those children who will be included in the study, i.e. will become your 'confirmed cases' for further analysis.

In most studies, the age range for cases will include ages from birth up to but not including 16 years. Please consider if children in the upper age range will be seen by paediatricians for this condition. A lower age cut-off may be used if older children are likely to be seen by adult specialists, for example, as case ascertainment through BPSU paediatricians in older age-groups would therefore be incomplete.

Finally, you should have a set of *reporting instructions* telling clinicians which children should be reported to you. The reporting instructions will reflect your surveillance case definition but are likely to be a shortened or simplified version of these. Examples of case definitions used in previous studies are provided in Appendix 2.

8. Expected numbers

Please supply an estimate of the number of cases expected each year, i.e. yearly incidence rate, indicate the sources that you have used to estimate this. More than 360 cases per year (or 30 per month) would normally be considered too high for the BPSU due to the monthly volume of notifications and the fact that regional studies may be sufficient. Please note that there are often duplicate reports so that the number of cases reported might be considerably higher than the number of true cases included in the analysis.

Indicate the source of denominator data for calculating incidence. This is often a routine data source, such as the following:

- England and Wales - Office for National Statistics mid-year population estimates or birth statistics (www.statistics.gov.uk).
- Northern Ireland - Northern Ireland Statistics and Research Agency (www.nisra.gov.uk)
- Republic of Ireland - Central Statistics Office (www.cso.ie)
- Scotland - ISD Scotland (<http://www.isdscotland.org>)

9. Research questions/surveillance objectives

Give a clear statement of the specific research questions that will be investigated by this study. These usually fall into the categories of 1) estimating incidence/birth prevalence; 2) describing the clinical features at presentation; 3) describing management and outcomes.

It must be possible to address these questions:

- a) Without direct contact with patients
- b) Without seeking investigations that would normally not have been undertaken by the paediatrician
- c) Without a separate comparison (control) group.

There does not need to be a long list of objectives. Consider how you will ask suitable questions in the questionnaires to gather information to answer your research objectives. Consider if you will have a sufficiently large sample size to address your objectives, for example regional variations in incidence could not usually be addressed by a BPSU study as the sample size would be too small. **Please note** that the BPSU surveillance methodology is not suitable for identifying causal relationships, as the frequency of 'risk' factors identified amongst notified cases cannot be compared with the frequency of these factors in unaffected 'control' children.

10. Methods

Please provide clear details of the study methodology that you intend to employ to answer your research objectives. If you plan to request clinical specimens or vary your methods from conventional BPSU studies, then please provide details.

If you wish to collect data via an online portal, please discuss this with the BPSU scientific coordinator. This will require you to have a secure database into which the data entered by clinicians automatically flows. The system will need to be registered on the IG toolkit and judged as 'satisfactory': www.igt.hscic.gov.uk The REDCAP online system is acceptable to BPSU.

11. Alternative sources of reporting

If it is likely clinicians other than paediatricians are likely to see cases it is essential to consider whether to involve these clinical specialists in case reporting as this will improve case ascertainment and reduces bias. This is particularly effective if the specialists have an established network, for example a specialist interest group or a laboratory network. Please list any additional sources of case reporting that you intend to use (and provide letters of support as appropriate).

Describe also the purpose of each additional source, how you will collect data and match between sources, and your proposed plan for analysis. Consideration of what identifiers are required to allow data linkage for de-duplication.

12. Proposed level and nature of public involvement

Please describe how you have involved, or intend to involve, the public in your study and whether this is consultation, collaboration or user-led (see below). You should supply further details of this activity, including the organisations that you have approached and how they have been and will be involved in your study. In seeking a PPI letter of support please inform them of the BPSU methodology – the BPSU office can advise you on wording. Submission of a letter of support which includes an acknowledgement that the BPSU methodology involves data collection without patient consent is advisable.

Using the approved template (available to download at www.rcpch.ac.uk/bpsu/resources) you should submit a public information leaflet which should include information about the condition and the study. This can be distributed to relevant groups / organisations and posted on the BPSU website. This is not a leaflet aimed solely at patients or parents; it must be suitable for the

general public. It will be available on the BPSU website and clinicians are asked to display the leaflet within their hospitals, e.g. on ward noticeboards.

The template includes standard descriptions of the BPSU methodology and also draws attention to the fact that any NHS patient can tell their clinician or health service provider if they do not want any of their health data held by the NHS to be used in anonymised research or audit. This is a legal requirement and demonstrates fair processing under the Data Protection Act 1998 and to ensure all individuals can dissent from research. You will be expected to include a description of your study, its purpose and potential benefits of the research to society, health services or affected individuals.

Please provide us with examples of any additional information materials (e.g. posters) that you will produce for the study.

Please attach any letters of support.

Definitions for the terms you are being asked to assess are included here:

Consultation Researchers consult members of the public about the research e.g. through individual contacts, one-off meetings.

Collaboration This includes active, on-going partnership between researchers and the members of the public e.g. involvement of members of the public on the project steering group, or as a research partners on a project.

User led/controlled Members of the public lead the research and are in control of the research. This is often through a community or voluntary organisation led by the service users.

For further information on public involvement in research visit: <http://www.invo.org.uk> or visit the BPSU website: www.rcpch.ac.uk/bpsu/patientsandpublic.

13. Questionnaire and letters to notifying paediatricians

Copies of questionnaire and covering letters to respondents must be attached even if they are only in draft form. The BSC will request final versions of your questionnaires and letters before final acceptance. It is essential to pilot your questionnaire with general paediatricians before submitting it to the BPSU for consideration even if this is only a small number. Ideally you should ask colleagues to test the questionnaire against one or more sets of notes to assess whether all the information requested is available from routine case notes.

Please describe any pilots and changes made to questionnaires subsequent to piloting. It is advisable that you also consult any lay/public involvement representatives involved in the study about the questionnaire, in order to determine whether the questions are appropriate or if there are any missed opportunities to collect information that is particularly relevant to families and patients.

Please note that the BPSU provides instructions for the design of questionnaires (see below and Appendix 3) within this guide. The BPSU has also devised a template questionnaire with additional guidance, which you are strongly advised to use. It is strongly advised that you liaise with the designated Medical Adviser before submitting your questionnaire as failure to do so may lead to delays in processing your application or its rejection.

Provide details of the identifiable data that you will be collecting and justify why each identifier is required, e.g. for de-duplication or clinical data analysis.

14. Ethical approval

In this section please provide details of the current progress of your IRAS application. For more information about ethics approval, please see section **Error! Reference source not found..**

15. Funding arrangements

Outline the funding arrangements for the project. The BPSU requests a contribution charge of £15,000 for an initial 13 month study and £10,000 for subsequent years. Funding arrangements should not only cover BPSU costs but also administrative costs including research assistance/secretarial salaries.

Please name the body(ies) to which grant application(s) have been submitted or from whom funds will be available. Give the date by which arrangements are expected to be agreed. State whether funding is from a commercial source or whether you are personally in receipt of funds to undertake the research.

Note the Full Economic Cost (FEC) for the BPSU is £25,000 for a 13 month study. If your funding is via a commercial source the FEC figure should be stated in any application.

Also, if funding is from a commercial source, you may be expected to demonstrate, for example through a contract with the funders, that this will not influence the reporting of results, and you may wish to discuss this with the scientific coordinator. Where the study is funded by a third party (commercial or non-commercial), it is unlikely to be acceptable for them to have access to identifiable data.

16. Organisational Arrangements

Provide details for managing the project, such as administrative, scientific and information system support. Particular attention will be paid to whether the resources are sufficient to run a successful project, processing reports in a timely manner, information technology support etc. We strongly advise that a research administrator or officer be employed if the expected number of case reports is greater than 100 per year. You should be aware of the requirements for security and confidentiality in handling patient identifiable data described on p.19.

17. Attached documents checklist

Please include with your application form the following additional documents:

Covering letter Please attach a signed covering letter from the main contact/principal investigator for the study.

Supporting letters Please attach any letters of support that you consider relevant for the SC to consider, for example award letters from funding bodies or letters confirming support by collaborating partners such as paediatric specialty groups.

Questionnaire and covering letters

Please attach all questionnaires and letters that will be used within the study. Please provide a version number and date for each.

Data analysis table Please supply a breakdown of how the questions are to be analysed and how they will address the objectives of the study. Examples are available on request.

Public information leaflet/poster

Please attach any public information material.

Supporting letters Please attach any letters or statements of support, if appropriate.

Letter from funding body

Please attach confirmation of funding, if/when available.

Signature

An electronic version of the application can be submitted directly to the BPSU Office at bpsu@rcpch.ac.uk at least 3 weeks before the BSC meeting. A signed paper copy must also be sent to the BPSU office.

Questionnaire design

Listed below are some key issues to keep in mind when designing your questionnaire. A questionnaire template can be found on the BPSU website at www.rcpch.ac.uk/bpsu/resources and general advice on questionnaire design is also provided in Appendix 3.

Investigators are welcome to discuss questionnaire design with the medical advisers and/or BSC and copies of questionnaire used by existing studies are available on the BPSU website or on request from the BPSU Office. A letter of introduction should be sent with the questionnaire and a thank you letter should be sent on return of the questionnaire (Appendix 4). This is vital in keeping the continued support of the clinicians.

Several studies have collected data via an online portal, please discuss this with the BPSU scientific coordinator. Please note this will require you to have a secure database into which the data entered by clinicians automatically flows. The system will need to be registered on the IG toolkit as 'satisfactory': www.igt.hscic.gov.uk RECAP is the favoured online system used.

Key points

- Questionnaires should be as brief and simple as possible, so as not to impose an excessive burden on the paediatrician. Two A4 pages are usually adequate for the questionnaire. Reasons for requiring a longer questionnaire must be outlined in the application. However, a well-laid out four-page questionnaire is preferable to one of two pages that are cramped and difficult to complete. As a guide, the questionnaire should take no longer than 15 minutes to complete.
- 'The British Paediatric Surveillance Unit' should be included in the heading of questionnaires and covering letters with the BPSU logo and those of the parent bodies. The logo can be provided by the BPSU office. The public information leaflet should also include appropriate logos and the BPSU office will advise on these.
- Information sought should be easily accessible to the reporting clinician from medical case notes. Anonymised copies of discharge letters cannot be sought.
- A 'tick box' format should be used wherever possible, remember to include a 'don't know' or 'not tested' box where appropriate.
- The cover page of the questionnaire should contain the hospital and minimal identifiable data; this can then be separated from the clinical details and stored separately to protect confidentiality. Names and addresses should not be sought although a unique identifier (e.g. NHS or CHI number) is usually essential. Minimal patient personal information to allow identification of duplicate reports and collection of follow-up data (e.g., date of birth, sex, partial postcode or NHS number) is accepted by the BPSU, but you will also need to justify this to REC, CAG and PBPP.
- Specialist terms or abbreviations that may not be familiar to paediatricians should be explained in full. In general please try to avoid abbreviations and acronyms
- Standard accepted classifications should be used where possible. Ethnic group should be requested using the 2011 Census Classifications.
- Respondents should be asked to return the questionnaire even if they are unable to complete all items.
- A reply paid envelope for return of the data collection sheet is **essential**.

Appendix 1: Abbreviations and useful web addresses

Abbreviation	Organisation	Web links
CAG	Confidentiality Advisory Group (of the HRA)	www.hra.nhs.uk/resources/confidentiality-advisory-group/
GROS	General Register Office for Scotland	http://www.nrscotland.gov.uk/
HES	Hospital Episode Statistics	www.hesonline.nhs.uk
HRA	Health Research Authority	www.hra.nhs.uk http://www.hra.nhs.uk/resources/hra-approval-nhs-organisation-guidance/
HRA APT	HRA Approval Programme Team	hra.approvalprogramme@nhs.net
IC	NHS Information Centre	www.hscic.gov.uk
IGT	NHS Information Governance Toolkit	www.igt.hscic.gov.uk
IRAS	Integrated Research Applications System	www.myresearchproject.org.uk
	NHS Research Ethics Scotland	nhsg.nrspcc@nhs.net
ISD	Information and Statistics Division (Scotland's ONS)	www.isdscotland.org
MRIS	Medical Research Information Service (NHS Information Centre)	https://www.england.nhs.uk/2013/07/consultation-hosp-data/
ONS	Office for National Statistics	www.ons.gov.uk
PBPP	Public Benefit and Privacy Panel for Health and Social Care (Scotland only)	www.informationgovernance.scot.nhs.uk
PAC	Privacy Advisory Council (Northern Ireland)	http://www.privacyadvisorycommittee.hscni.net/
SLSP	System Level Security Policy	www.rcpch.ac.uk/bpsu/resources
UKCRC	UK Clinical Research Collaboration	www.ukcrc.org/regulation-governance
Other useful web links		
	Research database forms (and other example forms from IRAS) and e-learning module	www.ukcrc.org/regulation-governance/integrated-research-application-system
	MRC Data and Tissue Toolkit	www.dt-toolkit.ac.uk/home.cfm
	MRC Personal Information for Medical Research Guidance	www.mrc.ac.uk/pdf-pimr.pdf

Appendix 2: Case definition – development and examples

Developing a case definition

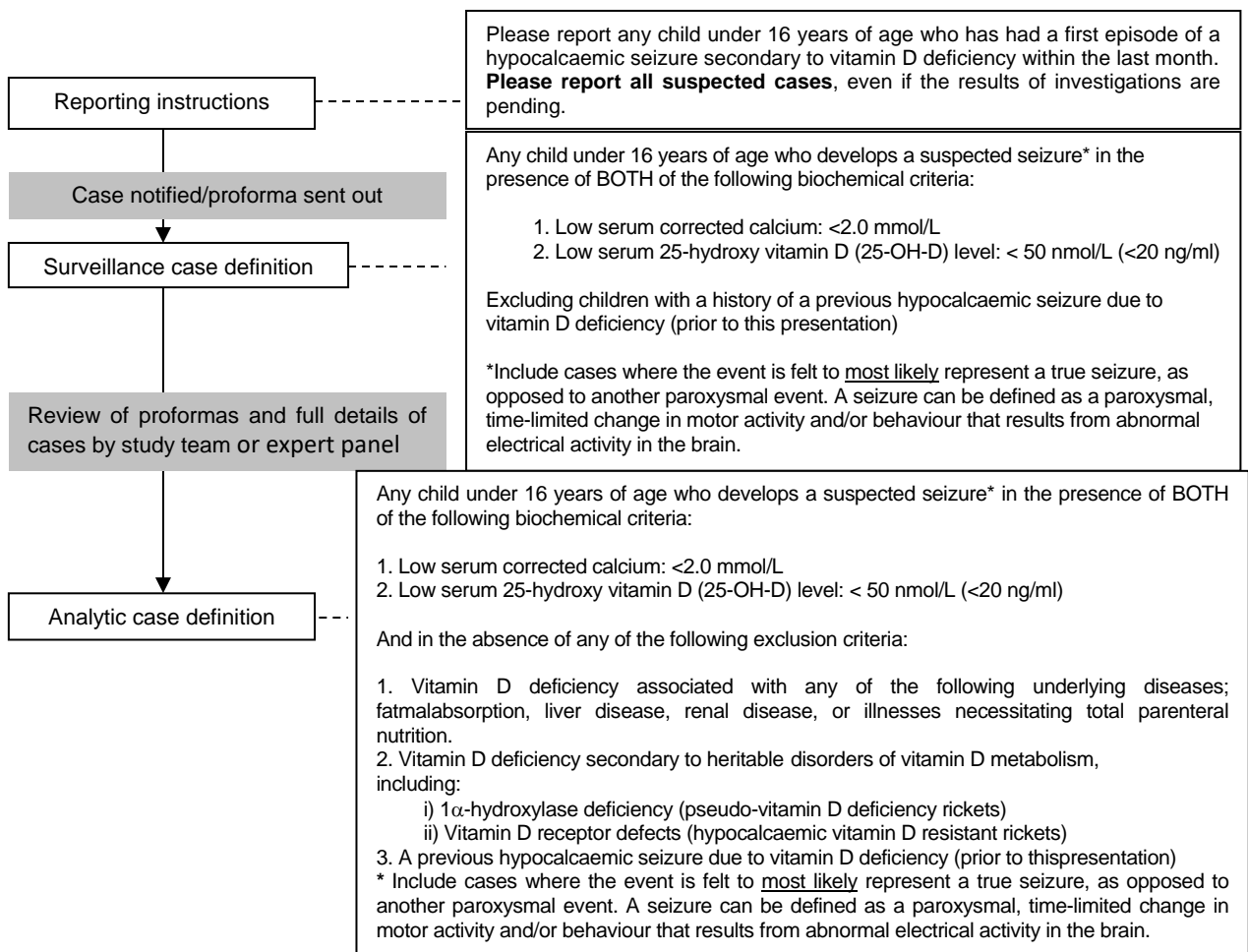
The case definition along with the research objectives are often the most important factor in the success or failure of a surveillance study and may be the main reason for the BSC to require revisions to the application. Failure to be able to apply a clear unambiguous case definition will result in the application being rejected. Please give careful thought to the case definition and if necessary seek advice from the BPSU office.

If you are developing a case definition, consider which symptoms, signs and tests you use to make the diagnosis. Symptoms and signs, such as fatigue or fever, which are common to many conditions are unlikely to be useful elements of a case definition on their own, however they may be clearly diagnostic of a disorder when found in association with other specific symptoms or signs.

The **surveillance case definition** defines clinically the cases that investigators are aiming to identify. It should state the age range, clinical symptoms and signs and results of investigations which would indicate a child is definitely or is likely to be a case. The surveillance case definition may be broader (less specific) than the **analytic case definition** applied using information from the questionnaires. For example, the surveillance case definition may include suspected but unconfirmed cases, whilst the analytic case definition for incidence estimates should include confirmed cases only. The **reporting instructions** are based on the surveillance case definition and state simply which cases should be notified to the study by clinicians.

Example reporting instructions & case definitions

Vitamin D Deficiency



Appendix 3: Guidance on developing a questionnaire

(We are grateful to Dr Helen Bedford for her help with this guidance).

This is an introduction to some of the considerations involved in questionnaire design including practical suggestions.

Self-completion questionnaires - by clinicians

The advantages of these are that they are:

- Less costly than interviews, require less time and energy to administer
- Can include a national, geographically spread sample using a mailed questionnaire

The disadvantages are:

- Possible response bias, i.e. non-responders differs in some ways from responders, giving an unrepresentative picture.
- Questionnaire design is crucial; it must be absolutely clear as there is no opportunity to explain questions.
- Responses are final - no opportunity to probe.
- Responses are limited to what is available in clinical notes.
- No control over who actually completes questionnaire, this may be the intended consultant or a junior staff member.

Questionnaire design

You will require a well-designed questionnaire. In practice designing a questionnaire is a skilled job and there are many pitfalls.

Key points are:

- Avoid ambiguity, bias and confusion.
- Don't underestimate the time it will take to construct the finished product. The more intelligible it seems, the greater the expertise that has gone into it. Always seek views of intended audience during drafting, i.e. discuss with colleagues
- Using questionnaires or parts of questionnaires that have been previously tried and tested is acceptable and also provides you with the additional advantage of being able to compare your findings with that of others.
- Pilot questions or the whole questionnaire with a local sample of clinicians, preferably against real sets of case notes.

Letters of introduction

You need to introduce the study by letter.

Letters should include:

- Aims of study and what you hope to get out of it e.g. 'we hope that the findings will help to improve services for children with disabilities in Brighton'.
- Assurance that information will be treated as confidential - it is best to state CONFIDENTIAL on the front sheet.
- Who you are, and your credentials
- Recognition of the effort required by the respondent
- Thanks
- Instructions re returning questionnaire
- Who to contact for more details
- Use plain English

The Questionnaire

- Length - 2-3 sides of A4 is considered to be maximum
- Layout is important, it must look attractive and not too formidable, and subjects must feel able to answer it.
- Using sections and boxes to separate different parts of the questionnaire can make it clearer and more inviting.
- Put instructions for completing at the top e.g. 'Tick the box next to the answer that applies to you'.
- If you are asking for more detail make sure there is enough space for people to write in.
- Skips are useful but must be clearly indicated e.g. ' GO TO QUESTION 3b'
- Language used is very important, should be appropriate to the sample e.g. language used for general public would differ from that used for health professionals.
- Don't be tempted to ask too many questions, stick to the minimum only, keep your research questions in mind all the time.
- It's occasionally useful to invite people to tell you anything else they want to at the end of the questionnaire but consider how this will be analysed
- Sensitive questions should be placed towards the end, then if the respondent does not wish to answer these, they may still have answered the others.

Types of question

1. 'Closed' or pre-coded questions, e.g. 'Was the child born preterm (before 37 weeks gestation)?'
 - Yes
 - No
 - Don't know

Advantages:

- This is useful if the range of answers to a question is limited and well established but always remember to include a 'don't know' option where relevant.
- Means people have to write very little, maybe useful for busy people.
- Makes analysis more straightforward.
- Make group comparisons easier
- Useful for testing specific hypotheses.
-

Disadvantages:

- May be a problem if all options are not included.
- Spontaneous responses lost
- May be bias in answer categories, e.g. if respondents prefer to opt for 'socially acceptable', 'don't know' or 'middle' option.
- Can be too crude

2. Open-ended questions

Allows respondent to answer in their own words, and highlight the particular issues that are important to them. e.g. *'How would you describe your relationship with your doctor?'*

Advantages

- Useful if you can't determine in advance what the main categories will be, useful in pilot surveys, means rich data is collected but dealing with the information in analysis is more difficult because if you have 50 responders you may get 50 completely different answers.
- Can place a burden as more time/thought required by respondents.
- More difficult to analyse against specific objectives.

In practice most questionnaires use a combination of closed and open-ended questions e.g.

'Does the child have any problems with his/her eyesight?'

- Yes
- No
- Don't know

If YES, please describe

It is often useful to begin with a closed 'yes/no' questions, then follow-up with an open-ended question that asks for detail.

Summary of question types

Open-ended: These allow a respondent freedom to write detail and express opinions, e.g. please describe..., tell me about...

Closed: These require a selection from a fixed set of answers, e.g. yes/no/don't know or male/female. A rating scale may be used to offer a wider range of answers.

Leading: These suggest a 'correct' answer to the respondent and are poor questions, e.g. *Do you think seatbelts should be compulsory in cars?*

Double-barrelled: These have two different questions rolled into one so it is not clear which is being answered, e.g. *Do you agree or disagree with ..., or Was the child unwell or in a state of collapse at the time of diagnosis?*

Hypothetical: These cannot be confirmed so are about opinion only, e.g. *Would the patient have been better without treatment?*

Measurement scales

These can be very useful for questions about health which tend to be on a continuum. Whether you choose a question or a scale depends on the nature of the variable you are measuring, e.g. whether it is categorical or continuous.

Examples of measurement scales:

Likert scale: used to indicate various degrees of strength of agreement or disagreement; commonly used to measure attitude, e.g.

'I would like my child to have his/her vaccinations in one injection rather than two'
strongly agree/ agree/ undecided/ disagree/ strongly disagree.

There are usually five or seven points on the scale, as an odd number allows respondent to express a neutral response to the statement. Responses can be allocated a score.

Guttman scale: This is also used to measure attitude and consists of a set of items with which people are asked to agree or disagree. The number of items usually small and a number of statements relate to a single concept. One score is allocated to each of the statements with which the person agrees, and they are allowed to agree with one or more statement.

e.g. *Statements relating to social isolation:*

- 1. I feel lonely*
- 2. I'm finding it hard to make contact with people*
- 3. I feel there is nobody I am close to*
- 4. I feel I am a burden to people*

Semantic differential scales: These are based on the importance of language reflecting a person's feelings. Respondents asked to make judgments about certain concepts and bipolar adjectives are stated at either end of a 7 point scale. These are only useful when responses to questions or statements can be categorised into conflicting adjectives.

e.g. *The session on questionnaire design was:-*

- | | | | | | | | | | |
|----|----------------------|---|---|---|---|---|---|---|--------------------|
| 1. | <i>Unhelpful</i> | 1 | 2 | 3 | 4 | 5 | 6 | 7 | <i>Helpful</i> |
| 2. | <i>Bad</i> | 1 | 2 | 3 | 4 | 5 | 6 | 7 | <i>Good</i> |
| 3. | <i>Uninformative</i> | 1 | 2 | 3 | 4 | 5 | 6 | 7 | <i>Informative</i> |

Visual analogue scales: These are frequently used in the clinical setting, e.g. measurement of pain:

No pain at all _____ X _____ *Worst pain imaginable*

Traditionally the line is 100mm in length and subjects are asked to mark a point on the scale which represents the amount of sensation they are experiencing. The mark on the line can be measured and a score allocated between 0 and 100mm.

Rating scales: These are used to evaluate performance or for the prediction of risk, and are necessary when objective measures of some skills are not available or are too complicated for general use. The points on the scale are derived from expert ratings and the methods of rating may be complex or simple. Examples are the Glasgow coma scale and Edinburgh postnatal depression score. Assessors must be well practiced in the use of the scale to ensure high degree of inter-rater reliability.

Question construction

- Use plain English and the most simple language you can
- Avoid double barrelled questions, e.g. 'Do you agree or disagree with the following statement: Examinations are a poor method of assessing ability and should be banned'.
- Avoid ambiguity - e.g. as in this question taken from a survey questionnaire sent to all female staff irrespective of whether they were pregnant or not: 'Is your work made more difficult because you are expecting a baby?'
- Avoid leading questions, e.g. 'You don't think.....do you?'
- Be specific, e.g. if you want opinions on how an outpatients department organised their appointments system, it is no good asking 'Are you satisfied with the outpatients department at X hospital?'
- Avoid vague words like regularly, frequently, occasionally, which might be interpreted differently. Define things like 'collapse' or 'crisis', which may mean different things to different people.
- The wording of the question is crucial, for example when the General Household Survey was collecting information on chronic illness they asked 'Do you suffer from any disability?' and the response was far lower than they expected. Next time they asked 'Do you have any disability?' and got a more accurate response.

Obtaining a good response rate with a mailed questionnaire

- Follow-up non-responders twice with reminders - can expect about 1/2 final response rate with first letter, another 1/3 with second and a few more with third.
- Need to be able to identify those who have not responded, so note consultant names/patient codes at the top of the questionnaire.
- Send reminders when replies stop coming back, usually after about 2-3 weeks with second class post. E-mail and telephone reminders are also acceptable.
- Always include another copy of the questionnaire in case it has been mislaid.
- FREEPOST or reply paid envelopes are essential.
- Printing questionnaire on coloured paper means it stands out from other correspondence.
- White envelopes differentiate from business mail.

Assessing the quality and adequacy of a questionnaire

Reliability of a questionnaire is a main criterion - this is the extent to which a questionnaire produces similar results under the same conditions on all occasions.

Validity - many different types of validity exist but it broadly refers to the extent to which a questionnaire measures what it is supposed to measure. Establishing validity can be difficult but piloting is a very important exercise in achieving validity and reliability.

Pilot Studies

It is sensible to pilot your questionnaire, so difficulties can be ironed out before the main study starts. It is best to ask several clinicians to test the questionnaire against a set of real notes and provide feedback on questions which are difficult to understand or data which is hard to find in the notes.

Analysis

Always think about your method of analysis when you are designing the questionnaire, as this may affect the design. For example, specific questions may relate to specific fields in an electronic database. Coding of responses, transforming them into numerical data to enable analysis, may be carried out during the planning stage of questionnaire design in which case you will need a coding frame either printed on the actual questionnaire or separately, or after the data is collected. The majority of closed questions can be pre-coded.

Examples of BPSU questionnaires

Current questionnaires are available on the BPSU website at www.rcpch.ac.uk/bpsu/resources.

References and further reading

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Appendix 4: Template letters to accompany questionnaires

On headed paper from the investigator and including a logo from the British Paediatric Surveillance Unit of the Royal College of Paediatrics and Child Health

EXAMPLE OF A LETTER TO THE REPORTING PAEDIATRICIAN

PRIVATE AND CONFIDENTIAL

[Name]
[Address]
[Date]



Dear Dr [Name],

Re: [Study name]

Thank you for notifying a case(s) for this study, via the British Paediatric Surveillance Unit (BPSU) 'orange' card system.

We are writing to gather further information about this case as described in the attached data collection form. This should take no more than 20 minutes and is really appreciated.

There are three ways in which you can return the data: by REDCap, by email or by post. By far the easiest method, preferred by most clinicians, is the REDCap system. This will require you to be issued with a password which then allows you to enter data directly into a completely secure web-based application.

1. If collecting data through REDCap:

Please click the link here which will send you to a secure online data collection form. You will be able to complete the form in your own time as and when the data becomes available, and then submit once complete.

2. If you would prefer to send the form via nhs.net email:

We would be very grateful if you could complete the attached password-protected data collection form (in MSWord format) and then return it to us, even if there are some sections that you have been unable to complete. Please ensure the file is password-protected on return, and sent from an nhs.net account to the [study] nhs.net email account. A password will be sent to you in a separate email.

3. If you prefer to post the form back to us:

Please let us know that you would like to post the form back to us by emailing XXXXX. We will post you a form to complete and return by prepaid secure post.

We will not be contacting your patient or his/her family at any time. Some patient identifiable data are needed to avoid duplication and to allow an estimation of the completeness of reporting. These will be removed once we have confirmed that the case has not already been reported to us. All of the information that you provide will be treated in strict confidence.

The study is funded by the XXXXXXXXXXXX. It has been approved by the Health Research Authority, the XXXXX Region MREC and the Confidentiality Advisory Board (Ref XXXXX). It has also been approved by the Public Benefits and Privacy Panel, for cases being reported in Scotland.

Please do not hesitate to contact us for any information should you have any queries about the questionnaire, eligibility of a particular case for inclusion in the study or any other aspects of the study.

We are extremely grateful to you for reporting to the BPSU and for taking the time to provide further information about your patient.

Finally, we will also ensure that you are sent a copy of the final report of the study.

Many thanks for your help,

Kind Regards

EXAMPLE OF A THANK YOU LETTER FOLLOWING COMPLETION OF THE QUESTIONNAIRE

On headed paper from the investigator and including a logo from the British Paediatric Surveillance Unit

[Name]
[Address]
BPSU reference:
Study ID:



[Date]

Dear Dr [Name],

Re: [Study name]

Thank you for returning a completed questionnaire to our BPSU surveillance study. We are very grateful to you for supporting this study.

Thank you very much for taking the time to provide us with this information. We will provide reports about the study on the BPSU website (www.rcpch.ac.uk/bpsu/xxxx) including information about any publications.

We propose to send a follow-up questionnaire to you one year after the date of the child's initial diagnosis of XXXXX to find out about clinical outcomes.

Please do not hesitate to contact us by phone or email (XXXXX@nhs.net) at the address below if you have any queries regarding the questionnaire or the study.

Yours sincerely,

EXAMPLE OF A REMINDER LETTER FOR NOTIFICATION/FOLLOW-UP QUESTIONNAIRE

On headed paper from the investigator and including a logo from the British Paediatric Surveillance Unit

PRIVATE AND CONFIDENTIAL

[Name]

[Address]

BPSU reference:

Study ID:

[Date]



REMINDER LETTER

Dear Dr [Name],

Re: [Study name]

We would like to thank you for your notification of a child with XXXXX to the British Paediatric Surveillance Unit (BPSU) monthly orange card scheme.

We recently sent you a <notification/one year follow-up> questionnaire to complete. We enclose a further copy of the questionnaire for your reference and would be very grateful if you could return the information to us as soon as possible *even if some diagnostic test results are pending*.

We recommend that you respond via the REDCap system by following this [link](#), or by sending it via nhs.net email to XXXX@nhs.net. Please ensure that this is password-protected and in MS Word format. A password will be sent in a separate email. If neither of these options is possible, please contact the study team at the address below. We will arrange for a paper copy of the questionnaire to be sent to you, which can be returned by prepaid secure post.

We will contact you by email or telephone in 3 weeks if we have not heard from you to ascertain that you have received the questionnaires and are not experiencing any difficulties in completing them. We are very happy to offer advice and support to you so please do not hesitate to contact us by phone or email (XXXXXX@nhs.net) if you have any queries regarding the questionnaire or the study.

Thank you very much for taking the time to provide us with this information. We will provide reports about the study on the BPSU website including information about any publications.

Yours sincerely,

EXAMPLE OF FOLLOW-UP LETTER TEMPALTES

On headed paper from the investigator and including a logo from the British Paediatric Surveillance Unit

Name]
[Address]
BPSU reference:
Study ID:



Dear Dr [Name],

Re: [Study name]

We would like to thank you for your notification of a child with XXXXX to the British Paediatric Surveillance Unit (BPSU) monthly orange card scheme, and for completing the initial form we sent you. We are now contacting you to establish clinical outcomes at one year of age.

There are three ways in which you can return the data: by REDCap, by email or by post. By far the easiest method, preferred by most clinicians, is the REDCap system.

4. If submitting data through REDCap:

Please click the link here which will send you to a secure online data collection form. You will be able to complete the form in your own time as and when the data becomes available, and then submit once complete.

5. If you would prefer to send the form via nhs.net email:

We would be very grateful if you could complete the attached password-protected data collection form (in MSWord format) and then return it to us, even if there are some sections that you have been unable to complete. Please ensure the file is password-protected on return, and sent from an nhs.net account to the XXXXX@nhs.net email account. A password will be sent to you in a separate email.

6. If you prefer to post the form back to us:

Please let us know that you would like to post the form back to us by emailing XXXXX@nhs.net. We will post you a form to complete and return by prepaid secure post.

If you are no longer caring for this child, we would be very grateful if you could provide us with details of the child's new paediatrician to whom we can write to obtain follow-up information.

Thank you very much for taking the time to provide us with this information. We will provide reports about the study on the BPSU website including information about any publications.

Please do not hesitate to contact us by phone or email (XXXXX@nhs.net) if you have any queries regarding the questionnaire or the study.

Yours sincerely,

LETTER TO GO TO PATIENT GROUP TO SEEK THEIR SUPPORT

[Name]

[Address]

[Date]

Dear [Name],

Re: Research into XXXXX condition

We would like to inform your group about a research project we are developing to look at XXXXX disease. Would there be interest in collaborating with us over the research? We are particularly looking to receive comments on the protocol, the public information leaflet and dissemination of the project and its subsequent findings. For our part we would be happy to come to present our work to your members. This is a surveillance project to examine disease epidemiology; that is the number of new cases of the condition being seen by clinicians, also how the disease is presenting, management and initial outcomes. In order to undertake this project we wish to involve the British Paediatric Surveillance Unit (BPSU - www.rcpch.ac.uk/bpsu) the foremost authority on facilitating such studies.

The BPSU facilitates epidemiological studies of rare childhood conditions across the four UK countries (Scotland, Northern Ireland, Wales and England) and the Republic of Ireland. In order to get a fuller picture as possible about the condition and achieve reliable and meaningful results, it is vital to try and collect as many cases as possible. Every paediatrician is contacted every month to ask them to recount any cases they have seen. They will notify us about who have reported a case, and we will then contact the paediatrician directly to get relevant clinical information.

Two important aspects of the BPSU methods that you should be aware, limited identification information is collected and patients or families consent is not sought. Both of these requirements are tightly governed by the relevant research regulatory bodies. BPSU justification is that the research does not involve any intervention and patient care is not affected. If consent was required, some paediatricians may not ask for that and some patients may decline, this would result in us underestimating the numbers of patients in the five countries. Some limited identification data (NHS number, DoB) is needed to avoid us double counting the same patient if reported by more than one paediatrician. This information is removed as soon as the data collection is completed. We are sure you will appreciate that with rare disease each case is crucial, so underestimating or the inability to remove duplicate reports can have a major impact on the conclusions we can draw from the study. The regulatory bodies have accepted these arguments and recognise the public benefit of our studies justifies the methods.

However, as you are a body which in some way represents the views and interests of patients and their families, we feel it is important to be open and honest about the scientific methods and ensure you are aware of these when you offer support or endorsement to our studies. BPSU would like to receive supporting letters for the study so if you are envisaging writing a supporting letter, it would be helpful if you include a short sentence that states you recognise that the BPSU methods require unconsented collection of data which includes some limited partial identifiers.

We are happy to discuss this further directly please do contact us.