

**An Evaluation of the
Surveillance System
of the British Paediatric Surveillance Unit
2008-09**



Health
Protection
Scotland



UCL

Institute of Child Health



Department
of Health

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1 Executive Summary

The British Paediatric Surveillance Unit (BPSU) was established in 1986 to undertake active surveillance of rare conditions affecting children. BPSU is a joint undertaking between the Royal College of Paediatrics & Child Health, The Institute of Child Health, London and the Health Protection Agency.

A monthly Orange Card is mailed to around 3000 consultant paediatricians in the United Kingdom and Republic of Ireland containing a list of up to thirteen conditions of interest. Paediatricians are asked to return the card indicating whether they have seen a case of any of the conditions in the past month or if not, to indicate nothing to report. Researchers wishing to study a condition via the BPSU submit an application to the Executive Committee which considers the proposal on scientific merit to determine whether it is suitable for BPSU surveillance. Notifications from Orange Card returns are passed to investigators who then contact the respondent directly to request further details and confirm case ascertainment.

A formal evaluation has recently been undertaken to assess the effectiveness of the BPSU system in carrying out its surveillance role and to explore its acceptability among researchers and notifying paediatricians, seeking to identify areas of both strengths and weaknesses to inform the continuing development and improvement of the BPSU. The evaluation was carried out in two phases – an internal review of the BPSU system applying CDC (Atlanta) guidelines for evaluating public health surveillance systems and a postal questionnaire survey of a sample of responding paediatricians and of researchers who had carried out a study via the BPSU. The findings form the basis of this report.

The overall response rate for Orange Cards is consistently over 93% and a similar response rate is seen for requests for additional information about notified cases. The Orange Card has been sent out without fail every month since the BPSU was established. The majority of respondents to the questionnaire survey indicated the paper Orange Card remained their preferred option for contact and notification to the BPSU.

The majority of studies remain on the Orange Card for duration of thirteen months but the system has been flexible to allow more sustained surveillance of conditions of major public health importance, including paediatric HIV infection, new variant Creutzfeldt Jakob disease and congenital Rubella infection. Joint studies with other bodies have also been undertaken to facilitate studies of conditions involving both paediatricians and other disciplines outside of the usual BPSU reporting base. Responses to the questionnaire survey have confirmed the willingness of paediatricians to respond to urgent surveillance questions in response to a public health emergency and email contact was the preferred format by the majority in this situation.

Since its inception, a total of 60 studies have been completed via the BPSU of which 51 have published their findings in a peer reviewed journal. To date, BPSU studies have given rise to a total of 169 peer reviewed publications. Responses to the questionnaire survey indicated that over 80% of respondents considered the surveillance of rare conditions in children to be important and 40% indicated that the outcomes of BPSU studies had changed their clinical practice.

BPSU currently retains data on case reporting to individual studies only for the duration of the study and there has been no archiving of data up to this point. The establishment of a database allowing audit of the study application process from the time of initial enquiry to study completion and which retains a minimum dataset for all completed studies has been identified as a priority.

Dr Simon Mitchell

Chair of the BPSU working party on the BPSU Evaluation

2 Background

The British Paediatric Surveillance Unit (BPSU) was jointly set up by Royal College of Paediatrics and Child Health (RCPCH), Health Protection Agency (HPA) and the Institute of Child Health London. In 1986 it began to undertake active surveillance of rare conditions affecting children. It also provides a mechanism for gathering data at national level in response to specific public health questions, including the capacity to undertake active surveillance in an urgent situation. The BPSU methodology has been successful in carrying out active surveillance for specific conditions over time-limited periods with consistently high coverage at a national level. This model has since been replicated by other specialities within the UK and in several other countries.

The surveillance of rare paediatric disorders has an important public health function: collecting reliable and timely information about the distribution and determinants of disease in the population and facilitating effective healthcare responses to reduce morbidity and mortality and improve health ¹.

Currently, around 3000 paediatricians in the UK and Ireland participate in the BPSU monthly active reporting scheme (the Orange Card).

After 22 years of surveillance, a critical evaluation of the BPSU's performance has been undertaken to assess the evidence that the BPSU is achieving its objectives in the surveillance of rare childhood conditions.

In addition, assessment of measures determining the effectiveness and perceived usefulness of surveillance activities were undertaken in order to identify areas of strength and opportunities for improvement.

The evaluation was broken down into 4 key areas

- 1. The BPSU operational model**
- 2. BPSU's contribution to knowledge of uncommon childhood infections and disorders among paediatricians and impact on public health policy.**
- 3. BPSU's engagement with the public and its contribution to public awareness of uncommon childhood diseases and disorders.**
- 4. Acceptability of the BPSU and its contribution to the training and development of its reporting base**

This report concentrates on key areas 1, 2 and 4. A further evaluation addressing key area 3 will commence in late 2008.

2.1 The aims of this evaluation are

1. To assess the efficiency, reliability, flexibility and responsiveness of surveillance operational practices employed by the BPSU (considering attributes suggested by the Communicable Disease Centre (CDC) Atlanta guidelines for evaluating surveillance systems²) in order to build on strengths and identify opportunities for improvement.
2. To estimate the BPSU's contribution to the advancement of knowledge of the prevention, diagnosis and management of uncommon childhood conditions and infections among paediatricians and its impact on informing public health policy.
3. To assess the acceptability of the BPSU's surveillance operations among paediatricians and researchers in order to ensure that surveillance activities are carried out as efficiently as possible, without causing an excessive burden to notifying paediatricians.

2.2 Specific objectives

The evaluation had the following specific objectives in assessing the BPSU's surveillance practices and outputs, based on the CDC guidelines for evaluating surveillance systems. Generally, the evaluation of public health surveillance systems involves an assessment of **usefulness** and the **system attributes**.

For our evaluation of the BPSU these included to:

1. assess the usefulness of the BPSU in contributing to the knowledge, prevention and management of rare childhood conditions.
2. assess the utility and timeliness of processes throughout all points in the surveillance loop and to assess the views of paediatricians regarding the timeliness of BPSU's processes.
3. assess the flexibility of BPSU methodologies to respond to new demands (e.g. emerging infections, questions of health policy, pharmaco- and immunovigilance needs) as the situation arises.
4. assess the quality of the surveillance data collected by the BPSU on which the individual surveys are based.
5. assess the acceptability of BPSU methodology and the value and credibility of the outcomes among the reporting base.
6. assess the simplicity of the BPSU's methods in collecting, analysing and reporting surveillance data.
7. assess the stability and resilience of the BPSU's systems
8. assess the representativeness of the BPSU's surveillance data over time and place to indicate whether all geographical regions and clinical specialties are adequately and proportionately represented in the reporting base.

Two other specific outcome measures, sensitivity and positive predictive value have been previously assessed using capture-recapture methods⁶.

2.3 Description of the BPSU surveillance system

The underlying principle for surveillance studies undertaken through the BPSU is the complete ascertainment of cases of specific rare childhood conditions in the UK and Ireland within a limited time period. The methodology is simple and designed to keep to a minimum the burden on paediatricians and clinicians reporting cases.

Access to the BPSU system is open to any clinician or research group and is assessed for inclusion on the basis of the scientific merits of the study proposal and its suitability to BPSU surveillance methodology. Generally, only rare childhood disorders (or rare complications of a more common disease), which are of such low incidence or prevalence as to require cases to be ascertained nationally in order to generate sufficient numbers for study, are accepted into the system. The number of conditions under surveillance is usually limited to 12 at any given time.

Accepted studies are required to conform to high standards of scientific rigour and practicality which are assessed through peer review during the application process. The application process consists of two phases: a screening phase at which an outline of the proposed study is considered by the BPSU Executive Committee (EC), and a subsequent detailed consideration of the full application form and study proposal. A decision to accept a condition into the surveillance system is based on its scientific merits, rarity of the condition and its importance to public health. Once approved by the BPSU Executive Committee, studies require Research Ethics Committee approval and Patient Information and Advisory Group (PIAG) approval for support under Section 251 of the NHS Act 2006 (formerly Section 60 of the Health and Social Care Act 2001), before they commence.

Consultant paediatricians who are either members of the RCPCH or the Faculty of Paediatrics of the Royal College of Physicians of Ireland, and a small number of paediatricians who are not members of the RCPCH but undertaking consultant level clinical practice, receive the monthly “Orange Card”. This notification card lists the conditions currently under surveillance. Clinicians complete the number of cases of any condition seen in the preceding month and return the card to the BPSU office. Importantly participants are requested to return the card even if they have no cases to report from the preceding month and to complete the ‘nothing to report’ box before returning the card. Surveillance is thus ‘active’ with the stimulus for reporting coming from the BPSU office. Clinicians who do not return cards for two consecutive months receive e-mail reminders (Figure 1).

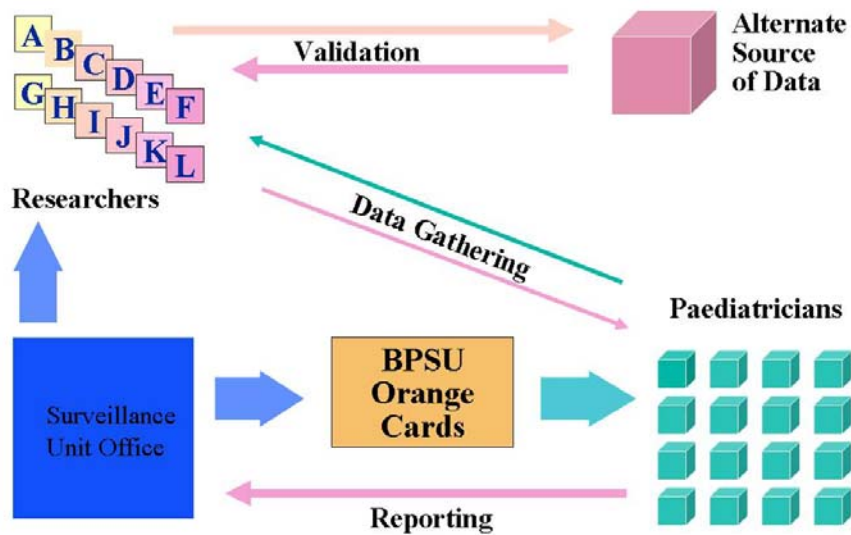
On receiving a case report the BPSU informs, via e-mail, the relevant study team. The study team then contacts the reporting clinician for further information about the case, usually through a questionnaire. The study team reports back to the BPSU, indicating when cases have been confirmed or identified as duplicate case reports.

The BPSU also actively encourages investigators to use alternative data sources such as the microbiology laboratories, and routine data, such as from the Office for National Statistics (ONS) or Hospital Episode Statistics (HES), to validate data. To improve case ascertainment for specific studies where a child may see specialist clinicians, consultants working in a number of other specialties relevant to the study have been invited to participate in the scheme, including child psychiatrists,

pathologists and microbiologists. In addition to improving case ascertainment, these complementary data sources help to validate the surveillance system.

Surveillance outputs are fed back via the BPSU's annual report as well as publications in peer-reviewed journals^{3,4}.

Figure 1 The BPSU surveillance loop



2.4 Structure of the BPSU

As previously described the BPSU is a tripartite organisation set up in 1985 by the British Paediatric Association (predecessor of the RCPCCH), HPA and the University College London, Institute of Child Health, (ICH). Health Protection Scotland and the Faculty of Paediatrics of the Royal College of Physicians Ireland are also closely involved with the BPSU. Each of these organisations has representatives on the BPSU EC. There are two full time staff, the scientific coordinator and the research facilitator.

In addition to assessing surveillance study proposals, the BPSU EC is responsible for setting yearly objectives and outlining longer-term strategy. The committee includes paediatricians, representatives of the partner organisations, lay representatives and two medical advisers. The medical advisers, one from the ICH (non-communicable disease) and the other from the HPA (communicable disease), assist the scientific coordinator in processing project applications. Along with the BPSU scientific coordinator they advise applicants on methodology and application processes; manage communication between the EC and applicant during the application process and support the development of study proposals. These posts are funded by the HPA and ICH. Administratively the BPSU sits within the Research Division of the RCPCCH.

The BPSU is currently funded by the Department of Health and additional funds are raised from researchers using the BPSU system for surveillance. The funds are managed by the Finance department of the RCPCCH.

3 Methods

The Updated CDC Guidelines for Evaluating Public Health Surveillance Systems² were applied as the main tool for the evaluation methodology. This is the most frequently used tool in evaluating public health surveillance systems and it guides the measurement of attributes such as timeliness, completeness, simplicity and accuracy, as well as the degree to which the data is used to stimulate public health action. Some attributes such as the costs of the systems were however excluded from this evaluation.

Data from the following sources were used:

1. BPSU databases, electronic and paper records
2. Published reports related to the BPSU and studies undertaken through the BPSU
3. A survey of a representative sample of clinicians who receive the Orange Card on a monthly basis
4. A survey of lead investigators from all 25 studies which were on the Orange Card at any time during a five-year period between October 2002 and September 2007.

BPSU database, electronic and paper records

An algorithm was devised to allow for appropriate data to be extracted and analysed from the BPSU database – see Appendix 1. This involved accessing information from the BPSU epidemiology programme, in particular the card responses from recent years. Regional analysis was undertaken using accumulated data previously published in BPSU annual reports.

The effectiveness of the application process was measured by looking at the time period before a study was placed on the card. This data was collected from written information presented at BPSU EC meetings and minutes from these meetings.

Published papers and reports

Basic data relating to all the papers published from BPSU studies was extracted into a database. This data was then analysed for topic, journal of publication and date of publication.

Survey of clinicians

To determine the perceptions of the reporting base to the various attributes of the surveillance systems, as well as its usefulness, a structured questionnaire survey of a representative sample of reporting paediatricians was conducted. A random sample of 600 clinicians on the database as of September 2007 was taken. Clinicians who had been receiving cards for less than nine months (added to the database after August 2007) were not included. Of this sample of 600 clinicians, 50% were sent the survey through the post, with reply paid envelopes and the remaining 50% clinicians were sent the same survey which they could access by clicking on link sent in an email. The web based survey programme *SurveyMonkey* was used to facilitate this.

An e-mail reminder was sent to all those who had not responded after two weeks. This email offered those who had initially been sent a postal survey the opportunity to

respond using the electronic survey or asked them to return the paper survey which had been previously sent to them. A final electronic reminder was sent to all remaining non-responders after a further month. After two months the survey was closed.

The answers from the structured questions were downloaded from *SurveyMonkey* to be analysed in MS Excel for descriptive frequency analysis, while the answers to open-ended questions were reported verbatim and subsequently reviewed and organised into specific themes.

Survey of investigators

In order to assess the effectiveness of the BPSU study application process and project management a separate survey was sent to all lead investigators undertaking surveillance during the period 1st October 2002 – to 30th September 2007, a total of 25. This was undertaken using *SurveyMonkey* with the data being downloaded for analysis

4 Results and Recommendations

Summary of general points

This evaluation considers the operation, outputs of the BPSU as a surveillance system and makes recommendations for change that will require further monitoring or audit after a suitable period. Overall, the BPSU system for surveillance of rare childhood conditions appeared to be effective in achieving its aims and is well-supported by reporting clinicians and investigators responding to surveys.

The tripartite nature of the organisation and collaboration of the three partner organisations in establishing and managing the BPSU is an important feature of the BPSU but not always clear in BPSU-related publications; this could be addressed in future BPSU communications strategy.

The involvement of patients and the public in the BPSU is not addressed within this report and a separate evaluation of this key area is envisaged to commence in late 2008.

General recommendations

1. The BPSU system works effectively and should continue to undertake surveillance of rare childhood conditions in the UK and Ireland as well as to continue to develop methodology, operational processes and communication strategies.
2. The ongoing activities of the BPSU should continue to be monitored and evaluated in the following ways:
 - a. Undertaking evaluation of BPSU engagement with patients, the general public and the research community
 - b. Auditing individual BPSU processes as changes are implemented
 - c. Repeating the evaluation of operations and outputs of the BPSU in 5-10 years.
3. Highlighting the work of the BPSU and its nature as a tripartite organisation, particularly through the websites and Press Offices of the three partner organisations, would enhance the profile of the BPSU and increase the impact of published outputs.

4.1 Usefulness

4.1.1 Number and classification (peer review publication or not) of surveillance and research outputs from BPSU surveys

Performance indicator	Assessment method	Expected outcome
<ul style="list-style-type: none"> Number and classification (peer review publication or not) of surveillance and research outputs from BPSU surveys 	<ul style="list-style-type: none"> Review and classify BPSU's surveillance outputs 	<ul style="list-style-type: none"> A database of all surveillance outputs (papers, presentations, posters, reports, as well as relevant newspaper articles) from the BPSU that is accessible whenever required.

All known surveillance and research outputs (all study publications and methodology papers) and International Network of Paediatric Surveillance Units (INOPSU) contributions (meeting details and publications) were collated. The BPSU keeps an MS Access 'Publication Database' of all known publications and presentations related to surveillance studies undertaken through the BPSU. Basic information about published papers are included but there is no formal system for obtaining copies of papers, though investigators are requested to notify the BPSU office of new publications. The database is currently being audited by BPSU Research Facilitator to assess the accuracy of data held within the system.

Press reports not currently maintained on this database but in a separate listing held at the BPSU office and the RCPCH Press Office. Press Offices of partner organisations do not maintain lists of BPSU publications.

Since the inception of the BPSU, studies using the BPSU system have been published in peer review journals and presented at national and international conferences. The known surveillance outputs can be grouped into the following categories:

Table 1 Research output of BPSU studies

Type of Research Output	Number
Publication	166
Conference oral presentations	108
RCPCH Meeting presentations	98
Conference Posters	16
Research Letters	3
Book Chapter /Reports	8

Further analyses of the 169 publications/letters in peer review journals are provided in Table 2. The journal "Archives of Disease in Childhood" was the most popular peer review journal for studies undertaken through the BPSU, publishing 52 papers, 31% of all BPSU related publications. Appendix 2.

Table 2 Journals in which BPSU studies have been published

Journal	Number of publications	% of 169 publications	Impact Factor
Arch Dis Child	49	29	2.8
BMJ	18	11	9.7
Communicable Disease Report	15	9	-
Lancet	12	7	28.6
Commun Dis Rep CDR Wkly	6	3	-
Arch. Dis. Child. Fetal Neonatal Ed.	3	2	2.3
AIDS	5	3	5.6
Others (less than 5 publications each)	61	36	-

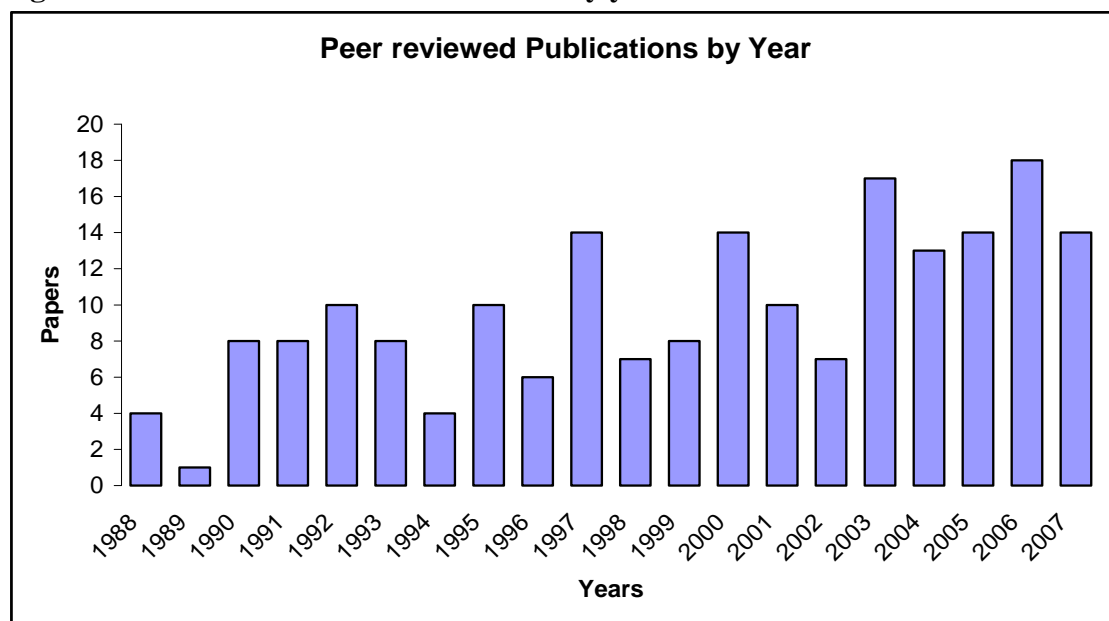
BPSU studies that have led to the most publications are listed below. Not surprisingly, the studies that have been on the orange card for prolonged periods have also produced the most publications in peer review journals.

Table 3 Publications resulting from BPSU studies

Study	Number	Percentage of 169 publications	Duration of study
AIDS/HIV	25	15	21 years
Congenital Rubella	12	7	17 years
Haemophilus influenzae type b (Hib)	10	6	8 years
Human Herpes Viruses-6 and 7	6	4	8 years (2 studies)
Medium Chain Acyl CoA Dehydrogenase Deficiency (MCADD)	6	4	6 years(2 studies)
Congenital Cataract	5	3	1 year
Inflammatory Bowel Disease	5	3	1 year
Reye's Syndrome	5	3	5 years
Progressive Intellectual and Neurological Deterioration (PIND)	4	2	11 years
Vitamin K deficiency bleeding	3	1	6 years (3 studies)

There has been a steady increase in peer reviewed publications from BPSU studies since the first publication in 1988 (Figure 2). Of 60 studies completed to date, only nine have not published in a peer review journal within two years of completion of data collection. Studies which have not published and the reasons for these are described in more detail in Section 3.5.3.

Figure 2 BPSU Peer review Publications by year



4.1.2 Quantification of BPSU's contribution to international collaborations i.e. other surveillance units, INOPSU

Performance indicator	Assessment method	Expected outcome
<ul style="list-style-type: none"> Quantification of BPSU's contribution to international collaborations i.e. other surveillance units, INOPSU 	<ul style="list-style-type: none"> Review BPSU's contribution to international collaborations i.e. other surveillance units, INOPSU 	<ul style="list-style-type: none"> A report (with a time line) of the BPSU's involvement in INOPSU A list of international collaborative papers.

A review of BPSU's contribution to surveillance units in other countries and its contribution to INOPSU was carried out with the BPSU scientific coordinator.

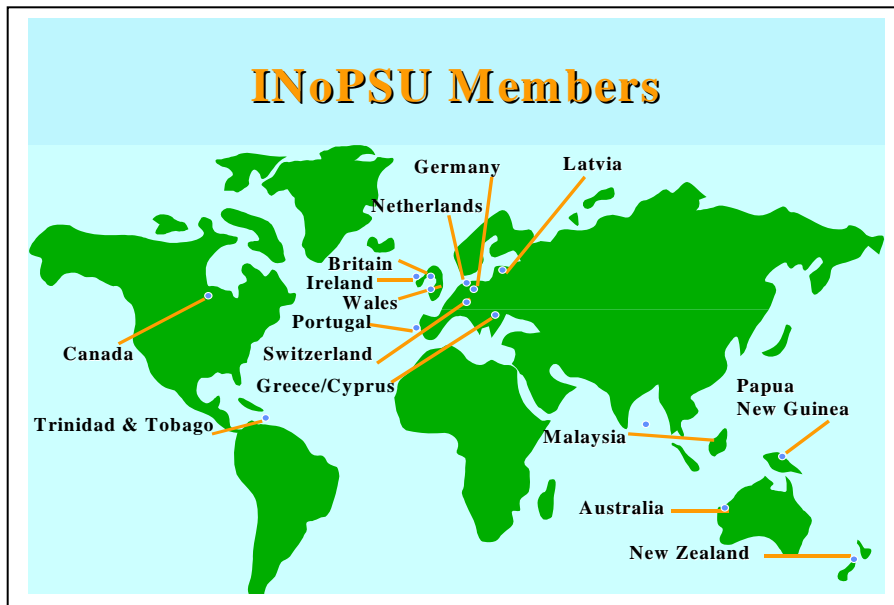
Following successful initial operations of the BPSU, the methodology was adopted in the 1990s by several other countries whose paediatric services were amenable to an active surveillance approach; these were developed with support from the BPSU. Surveillance units were founded in 1992 in the Netherlands and Germany and in 1994 in Switzerland. In total, thirteen countries have developed paediatric surveillance units based on the UK model (www.inopsu.com), although three are no longer active. The BPSU also continues to respond to requests for advice and support from national paediatric societies who are developing surveillance units.

The development of European surveillance units also led in 1994 to the establishment of an informal European network and to the first two European comparative studies of Vitamin K deficiency bleeding⁹ and Haemophilus influenzae type b¹⁰, which have since been followed by several further international comparative studies; L Pereira-da-Silva 2005¹², Grenier D 2006¹³.

In 1996 a proposal by the Australian Paediatric Surveillance Unit to form Units INOPSU was enacted by the paediatric surveillance units existing at that time. INOPSU was formed in August 1998 at a meeting in the Netherlands of ten units and the first INOPSU conference was held in June 2000 in Ottawa, Canada⁵.

A secretariat consisting of a convenor, deputy convenor and liaison officer now exists to take forward the aims of the network but the BPSU continues to play a central role in developing and organising INOPSU. The BPSU scientific coordinator led the initial drafting of the INOPSU mission statement and statement of terms and objectives and as liaison officer has continued to play a leading role in developing the network. At the 2006 INOPSU conference, a contribution to the running costs of administering INOPSU currently carried out by the BPSU office was agreed by those units who could this.

Figure 3 INOPSU members in 2007



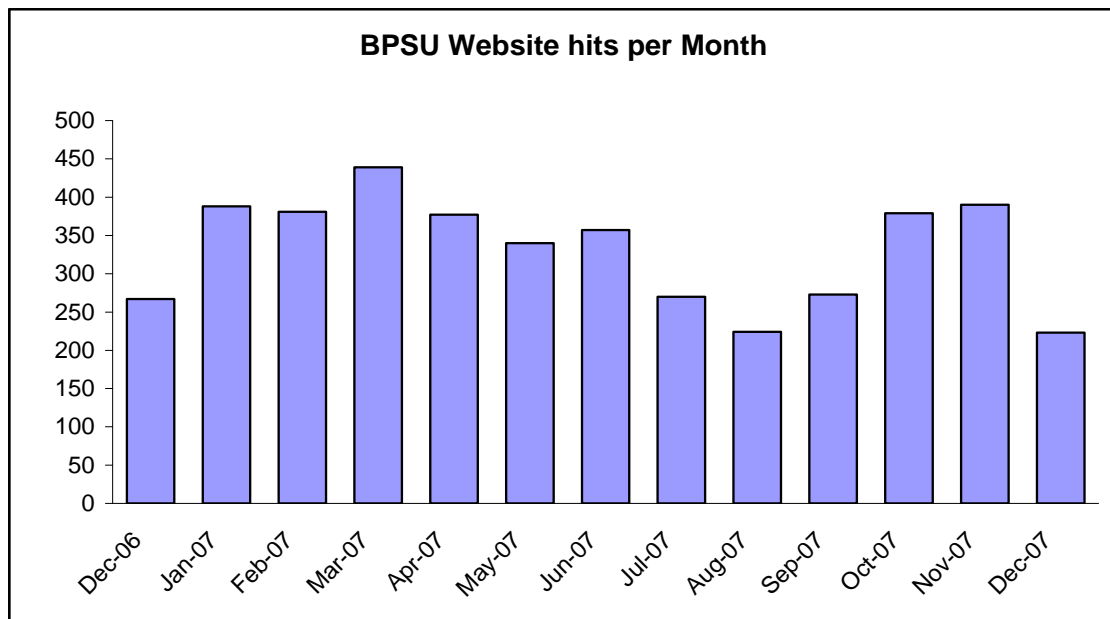
All the INOPSU members share results, protocols and facilitate contact between researchers in different countries. Three INOPSU reports have been published as a shared endeavour by participating countries.

4.1.3 Hits on the BPSU website

Performance indicator	Assessment method	Expected outcome
▪ Hits on the BPSU website	▪ Review website hits	▪ Analysis of website hits over time

The BPSU has been monitoring the hits on its website since December 2006. On average there are 330 site entries per month. This has remained relatively constant except for a dip during the summer and winter holidays.

Figure 4 Hits on BPSU website in 2007



4.1.4 The reporting base's perception on the usefulness of BPSU surveillance

Performance indicator	Assessment method	Expected outcome
<ul style="list-style-type: none"> The reporting base's perception on the usefulness of BPSU surveillance 	<ul style="list-style-type: none"> Closed questionnaire to participating paediatricians 	<ul style="list-style-type: none"> Perception of the reporting base on the usefulness of BPSU surveillance

Paediatricians that form the reporting base were asked five questions relating to their perception of the usefulness of the BPSU surveillance system.

Table 4 Effect of outcomes BPSU studies on clinical practice

Have the outcomes of BPSU studies changed your clinical practice?	Total	%
No	258	57
Yes	195	43
(blank)	1	0
Total	454	
10 most frequently mentioned studies to have changed clinical practice	No (%) of times mentioned	
Vitamin K	27 (15)	
Kawasaki	14 (8)	
Group B Streptococcus	13 (7)	
Diabetic ketoacidosis	12 (7)	
Progressive Intellectual and Neurological Deterioration (PIND)	10 (6)	
HIV	9 (5)	
Neonatal herpes	9 (5)	
Medium Chain Acyl CoA Dehydrogenase Deficiency (MCADD)	7 (4)	
Thyrotoxicosis	6 (3)	
Hyperbilirubinamia	5 (3)	
Cytomegalovirus	5 (3)	

Table 5 Other utility of BPSU studies

Have BPSU surveillance studies been of use to you in any other way	Total	%
No	164	36
Yes	275	61
(blank)	14	3
Grand Total	454	
Other benefits of BPSU studies mentioned (some are mentioned multiple times)		
<ul style="list-style-type: none"> • <i>It is important to feel part of team looking at rare diseases for the benefit of patients</i> • <i>HIV follow up: ensure that children are not being lost in the system</i> • <i>Raises awareness</i> • <i>General Information</i> • <i>Approach to epidemiology of rare conditions, effectiveness of networking of clinicians</i> • <i>CPD and reflection about cases</i> • <i>Improving evidence base for management</i> • <i>Educational role</i> • <i>Useful for teaching</i> • <i>Increased my knowledge about some of the rare conditions</i> • <i>Provides epidemiological data for service development initiatives</i> • <i>Prevalence figures for business case</i> • <i>Teaching junior doctors</i> • <i>I find it reassuring to know that these conditions are being considered</i> • <i>Puts rare conditions in context</i> • <i>Made me aware of changes in clinical practice</i> • <i>Interesting to see data and evidence of disorders and may be useful in future management and development of services</i> • <i>Bench marking practice</i> • <i>Useful epidemiology, reinforces network approach</i> • <i>Protocol cards, information sheets and reports are useful</i> • <i>Establish frequency of rare conditions and underlying causes</i> • <i>For the development of guidelines</i> • <i>Timely reminder of rare conditions or maternal conditions affecting the newborn infant</i> • <i>Understanding trends</i> • <i>Useful for self-directed learning</i> • <i>Good evidence of national statistics on some of the common and rare conditions.</i> • <i>Gives me an idea of what criteria are used to define rare conditions.</i> • <i>Clarification of case definitions</i> • <i>Ideas for my own studies</i> • <i>In getting a feel for how common or uncommon conditions are</i> • <i>Prompts our own internal audit of what is going on.</i> • <i>Helped to make me aware of patterns of rare conditions where I am unlikely to gain much personal experience</i> • <i>Excellent epidemiology</i> • <i>Supporting my existing practice</i> • <i>One was also the basis for my MD thesis</i> • <i>Court</i> • <i>Awareness of specific conditions, eg, MCAD, CMV</i> • <i>When talking to patients and reassuring them about the kind of studies my information is based on</i> 		

Table 6 Perceived importance of the study of rare paediatric conditions

Do you think that the study of rare paediatric conditions is important? Please rate out of 5.	Total	%
1 - <i>very important</i>	276	61
2	102	22
3	34	7
4	33	7
5 - <i>not important</i>	4	1
(blank)	5	1
Total	454	

Table 7 Frequency of accessing information sources AND the usefulness of these information sources

Resource	%		%				%
	Ever Accessed	often	sometimes	never	N/A	Found Useful	
Archives of Disease in Childhood	91	63	28	3	6	87	
BPSU Bulletin	90	47	43	4	6	79	
BPSU Annual Report	79	31	48	10	11	75	
Other peer review journals	62	21	41	16	22	84	
Presentations at the RCPCH Annual Meeting	60	10	50	24	18	80	
Other scientific meetings, conferences and congresses	50	8	42	30	20	20	
HPA Communicable Disease Report (Online)	30	3	27	52	18	67	
BPSU website (http://bpsu.inopsu.com)	15	1	13	66	20	73	

Table 8 Satisfaction with the feedback you receive from the BPSU?

Are you satisfied with the feedback you receive from the BPSU?	Total	%
1 – very satisfied	217	48
2	219	48
3 – not satisfied	15	3
(blank)	3	1
Grand Total	454	

4.1.5 BPSU’s researchers’ perception on the utility of BPSU surveillance

Performance indicator	Assessment method	Expected outcome
▪ BPSU’s researchers’ perception on the usefulness of BPSU surveillance.	▪ Closed questionnaire to BPSU researchers’	▪ Perception of BPSU researchers on the usefulness of BPSU surveillance

Investigators who had run studies through the BPSU in the last 5 years were asked to complete a survey on the utility of the feedback from the BPSU. One response was requested for from each study group. Twenty four (89%) responded to this request and the majority of questionnaires were completed by the lead investigator (80%).

Table 9 Satisfaction with the feedback researchers receive from the BPSU

	Strongly Agree (%)	Agree (%)	Neutral (%)	Disagree (%)	Strongly Disagree (%)
Contribution to the medical literature	65	35	0	0	0
Evaluating current medical management/policy	48	48	0	4	0
Informing future medical management/policy	39	57	4	0	0
Contributing to screening policy	33	14	24	14	14
Contributing to other disease prevention policies	13	26	43	9	9
Contributing to your professional development	30	61	4	0	0

Interpretation of results: usefulness

- The BPSU surveillance system is a simple and effective system for the surveillance of rare childhood disorders in the UK and Ireland.
- There has been a steady increase in publications from BPSU studies in peer review journals since the first publication in 1988 and of the 60 studies completed to date, only nine have not published in a peer review journal within two years of completion.
- Several BPSU studies are reported to have led to changes in clinical practice; most frequently mentioned are the studies on Vitamin K deficiency bleeding, Kawasaki disease and Group B *Streptococcus*.
- BPSU studies are useful in a wide variety of other ways including the improvement of knowledge, epidemiology competence, teaching, and research skills.

- The journal “Archives for Disease in Childhood” was the most popular peer review journal for studies undertaken through the BPSU, publishing 30% of all BPSU related publications.
- Thirteen countries have developed paediatric surveillance units based on the UK model and the BPSU is a founder member and the current administrative centre of INoPSU, a network of paediatric surveillance units formed in August 1998 to facilitate international collaboration and comparative studies.
- Since monitoring began in 2006, the mean number of entries to the BPSU website has been 330 per month suggesting a significant level of interest in the BPSU.
- The BPSU maintains a database of publications and presentations related to the BPSU since 1986, however this has not been audited for accuracy and processes for updating are unclear. Press reports are not recorded on this database nor has a link to the Press Offices of the partner organisations been established to ensure that new publications are appropriately disseminated.
- Investigators who read the information provided by the BPSU found it useful in most cases. Investigators asked for more information to be provided on writing the questionnaires in particular to be provided with examples of other studies questionnaires. Suggestions made to improve the application process included ensuring that guidelines were always up-to-date and that the application process information included details on the possibility of attending EC meetings.

Recommendations:

1. The BPSU surveillance system is a simple and effective system for the surveillance of rare childhood disorders in the UK and Ireland, which should be maintained and developed.
2. The BPSU should continue to encourage and support investigators in publishing their studies in peer review journals but should also aim to enhance future output by targeting higher impact factor journals and developing links with Press Offices in all partner organisations.
3. The database of publications should be audited and a policy for monitoring study outputs, obtaining information on publications from investigators and updating the database on a regular basis should be implemented. Using software specific for maintaining publications data such as "Reference Manager" or "Endnotes" should be considered.
4. The BPSU should maintain an active role in INoPSU and in advising new paediatric surveillance units.

4.2 Data Quality

4.2.1 Availability and accessibility of BPSU surveillance data and output outputs

Performance indicator	Assessment method	Expected outcome
<ul style="list-style-type: none"> ▪ Availability and accessibility of BPSU surveillance data and output outputs 	<ul style="list-style-type: none"> ▪ Review storage systems, organisation and maintenance for surveillance data and surveillance outputs. 	<ul style="list-style-type: none"> ▪ An “End-of-study protocol” that includes all relevant data that should be collected for each study which will be stored in an “End of study directory”.
		<ul style="list-style-type: none"> ▪ An “End of study directory” for each study that will contain: <ul style="list-style-type: none"> ○ A minimum dataset for each study with a line listing of each case reported with final classification. ○ All other relevant information and material on each study.
		<ul style="list-style-type: none"> ▪ A database with summary information of all completed studies ▪ Agreed processes on where to store these and how to manage them

At present the BPSU holds data on case-reporting to individual studies until all follow-up reports have been notified to the BPSU office. The database is primarily designed for administration of the Orange Card reporting system while a condition is under surveillance, although notification data is kept for a short period once the study has been completed before being removed from the administrative database. Printed anonymised copies of the notification reports are kept in the study files.

Paper and electronic copies of study applications forms and protocols are stored but there is currently no formalised system for archiving relevant electronic or paper records once a study has been removed from the orange card.

4.2.2 Reported card return and case-reporting behaviour of paediatricians

Performance indicator	Assessment method	Expected outcome
▪ Reported card return and case-reporting behaviour	▪ Closed questionnaire to BPSU researchers'	▪ Reported card return and case-reporting behaviour of the reporting base

Table 10 Reported card-return behaviour

Of the orange cards you have received, have you returned	Total	%
ALL	385	85%
More than half	57	13%
Less than half	2	0%
Never	1	0%
Only when I see a case	2	0%
Blank	7	2%
Total	454	

Table 11 Reported case-reporting behaviour

How many cases have you reported to the BPSU in the past 2 years	Total	%
0	213	47%
1	104	23%
2	69	15%
>3	37	8%
Unsure	27	6%
Blank	4	1%
	454	

Excluding those who did not respond or were 'unsure', at least 210 of the 454 respondents (46%) had reported at least one case to the BPSU in the past two years. NB Although a list of conditions that had appeared on the cards in the last 2 years was provided to all respondents, this figure is higher than the actual recorded figure of 19% of orange card recipients who normally report a case each year; it is likely therefore that some of the 46% of respondents were referring to case notification prior to 2006.

Although only 213 respondents said that they had not reported a case, 234 gave reasons why they had not. A breakdown of the reason for not reporting is provided in Table 11.

Table 12 Reasons for not reporting cases

Reasons for not reporting	Total	%
You have never seen a case of a condition on the card	188	80%
You have seen a case but thought a colleague had reported	28	12%
Other	18	8%
	234	
Other reasons for not reporting cases		
<i>Forgotten the case details when the card arrives. Expected others on the team to report</i>		
<i>Seen cases thought were outside the surveillance period</i>		

Interpretation of results: data quality

- The BPSU database is designed for the administration of the Orange card. The system holds data on case-reporting to individual studies only for as long as active surveillance is on-going. Processes for monitoring studies from the point of initial enquiry or application and for archiving study data after completion are not sufficient to allow ongoing audit and monitoring of BPSU operational systems.
- Clinicians report high levels of card return but appear to overestimate case-reporting, although this is likely to be subject to recall bias.

Recommendations:

1. A new database should be developed to collect data on all BPSU study enquiries, applications and completed surveillance studies.
 - a. This database should permit audit and monitoring of BPSU operation to be undertaken on a regular basis.
 - b. A minimum dataset for each completed study should exist e.g. an 'End-of-study' directory
2. Systems for archiving individual study information collected by the BPSU should be established.

4.3 Flexibility

4.3.1 BPSU's flexibility in adapting its methodology to undertake studies in response to new public health objectives

Performance indicator	Assessment method	Expected outcome
<ul style="list-style-type: none"> ▪ Number of past studies that were a direct response to new public health objectives and consensus view on success of these responses. 	<ul style="list-style-type: none"> ▪ Review of BPSU studies in terms of its "response to new public health objectives" 	<ul style="list-style-type: none"> ▪ Review of BPSU studies in terms of their "response to new public health objectives"
<ul style="list-style-type: none"> ▪ The BPSU's flexibility in adapting its methodology in order to undertake specific studies for which routine BPSU methodology is not adequate. 	<ul style="list-style-type: none"> ▪ Review BPSU's flexibility in adapting its methodology in order to undertake specific studies for which routine BPSU methodology is not adequate. 	<ul style="list-style-type: none"> ▪ A report of the BPSU's involvement in studies where flexibility in its methods were necessary.

Although the extent to which the BPSU has developed studies in response to specific public health issues cannot easily be quantified, the system does have the ability to rapidly undertake surveillance and provide information to guide public health policy, collect information about emerging infections, to inform screening and wider health policy, and for pharmaco- and immunovigilance.

Studies developed in response to public health concerns: A few studies can be highlighted as being developed, and perhaps even fast-tracked onto the Orange Card, in response to a specific public health concerns, including the studies of HIV/AIDS, variant CJD (PIND study), water birth safety, Reye's syndrome and haemolytic uraemic syndrome. Three studies remain on the Orange Card indefinitely as they are the only method for long-term national monitoring of these important conditions (HIV/AIDS, PIND and congenital rubella).

Studies informing health policy: There are many more studies which have informed public health policy but were less clearly a rapid response to a public health issue of the day. Of 60 completed studies undertaken through the BPSU, six have contributed to national screening policy (congenital toxoplasmosis, neonatal herpes, congenital syphilis and Group B streptococcal infections, congenital hip dysplasia and MCADD), five have contributed to immunovigilance and vaccine policy (MMR meningitis, SSPE, acute flaccid paralysis, haemophilus influenzae infection and Vitamin K deficiency bleeding) and three to pharmacovigilance (Reye's syndrome, Vitamin k deficiency bleeding and suspected adverse fatal reactions).

Studies funded on the grounds of public health importance: A further two studies have been directly funded by the Department of Health in response to issues of perceived public health importance (water births, Vitamin K deficiency bleeding) and one has been funded by the National Screening Committee to evaluate a pilot of screening (MCADD).

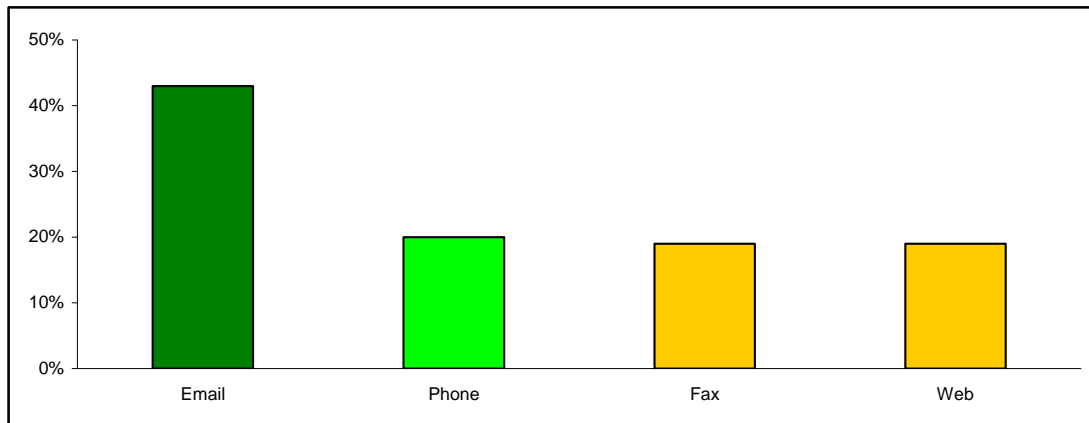
Studies with parallel reporting systems: BPSU studies have included multiple sources of reporting for conditions which might be seen by clinicians within the normal reporting base of the BPSU. In these studies, the BPSU has been flexible in the methodology used, including adding clinicians of other specialties to the orange card system for the study duration (e.g. FMAIT in collaboration with UKOS), setting up and administering a parallel system for investigators (e.g. intussusception), or allowing investigators to manage the system (e.g. MCADD).

Studies with rapid reporting systems: Certain BPSU studies, including haemolytic uraemic syndrome and acute flaccid paralysis, which were considered public health emergencies have required case-reporting by telephone to ensure rapid response.

4.3.2 The reporting base's willingness to report rapidly via accelerated systems in the event of a public health emergency

Performance indicator	Assessment method	Expected outcome
<ul style="list-style-type: none"> The reporting base's willingness to report rapidly via accelerated systems in the event of a public health emergency 	<ul style="list-style-type: none"> Closed questionnaire to participating paediatricians 	<ul style="list-style-type: none"> A report on the perception of the reporting base on its ability to adapt to accelerated reporting systems

Figure 5 Preferences for BPSU related contact in public health emergencies



Respondents to the survey were asked how they would prefer to be contacted if the BPSU were to undertake a study which required rapid reporting due to the public health importance. The majority of respondents favoured e-mail as the method for reporting urgent cases.

Interpretation of results: flexibility

- The BPSU is responsive to public health concerns although this is difficult to quantify. In the past, the BPSU has fast-tracked applications, set up parallel reporting systems and established rapid reporting methods in response to perceived public health emergencies and concerns.
- As the majority of BPSU respondents prefer e-mail as the method for reporting urgent cases, the BPSU should ensure that e-mail addresses are accurately held within the reporting clinicians database to allow a rapid response to a public health emergency if and when this arises.

Recommendations:

- The BPSU should continue to regard flexibility in responding to urgent public health concerns as an important issue and should consider limiting the number of conditions on the Orange Card to ensure that a space is always available for an additional 'rapid-response' study.
- The BPSU should ensure that e-mail addresses are accurately held and regularly updated within the reporting clinicians database to allow a rapid response to a public health emergency if and when this arises.

4.4 Simplicity

4.4.1 Ease of data flow through the system

Performance indicator	Assessment method	Expected outcome
▪ Ease of data flow through the system	▪ Review the BPSU surveillance loop for any obvious bottlenecks	▪ A description of the BPSU surveillance loop for any obvious bottlenecks.
		▪ Documentation of surveillance processes.

At their first contact with the BPSU office, all applicants are given the contact details of the medical advisers and advised that they may request advice about any aspect of the methodology or BPSU application process. Since 2007, the dates for BPSU EC meetings have been made available on the BPSU website.

Applications for inclusion of a study on the orange card are considered by the BPSU EC, which meets every two months. Then a two-stage application procedure follows. An applicant may be invited to attend a BPSU committee meeting to present their proposal and discuss any queries that have arisen.

The BPSU EC, scientific co-ordinator and medical advisers regularly review these processes. In 2004, it was noted that investigators were submitting several revised application forms at each stage of the application process and observed that this was inefficient. A revision of the BPSU application guidance and application forms was undertaken by the medical advisers and scientific coordinator. Prior to this review, there was no upper limit to the number of revisions that might be submitted, however, since this revision, only the following outcomes are possible at P1 stage:

- 1) The P1 may be accepted and a P2 sought without specific modification
- 2) The applicant may be asked to give a presentation to the committee before a decision is made
- 3) Further details may be sought before a decision is made on the P1 acceptability as a potential BPSU study.
- 4) The P1 may be accepted with the provision that several points are addressed in the P2 application.
- 5) The application may be rejected.

A study which is approved at P1 may still be rejected at P2. Since 2006, if an applicant is not invited to discuss their study with the committee at P1, then they will be invited to do so at P2 (except in exceptional circumstances). Guidance and application forms are reviewed and revised on a regular basis and the latest versions (Sept 2007) are available on the BPSU website or from the BPSU office.

A review of the data flow through the surveillance loop was carried out with the scientific adviser (described in the first section) in order to identify present or potential obstacles. It was agreed that the functioning of the BPSU is critically dependent on the efficiency of the BPSU surveillance office.

4.4.2 Ease of application process

Performance indicator	Assessment method	Expected outcome
▪ Ease of application process	▪ Closed questionnaire to BPSU researchers	▪ Perception of BPSU researchers on the ease of the application process

Table 13 Was the ‘Guidelines for Investigators’ document useful for:

	Yes (%)	No (%)	Did Not Read (%)
Application form	65	4	30
Development of the questionnaire	56	13	30
Ethical review procedure	65	8	26

Several of those who ticked ‘did not read’ explained they did so because they were not involved in the initial stages of setting up the study or this information was not available to them when their study started (Congenital rubella, HIV/AIDS).

Contact with the BPSU office while developing the study proposal was found by the majority, to be useful (83%) and responses were regarded as being timely (63%). Table 10 below shows these results in more detail.

71% did not experience any difficulties in obtaining ethical and or PIAG approval. Those that did encounter problems mainly did so with respect to gaining permission to collect (some) patient identifiable data.

Table 14 During the assessment of Phase 1 and 2 applications

	Yes (%)	No (%)	Do not remember (%)
Did you find the comments received from the BPSU useful?	83	0	17
Did you find the time of the responses from the BPSU prompt?	67	8	25
Where you invited to attend a BPSU meeting to discuss your application?	58	21	21
Did you/ Would you find this invitation useful?	71	8	4

4.4.3 Availability of processes and procedures for staff induction and training

Performance indicator	Assessment method	Expected outcome
<ul style="list-style-type: none"> Availability of processes and procedures for staff induction and training to maintain the resilience of the system. 	<ul style="list-style-type: none"> Review availability of processes and procedures for staff induction and training 	<ul style="list-style-type: none"> Induction and training manual for research division staff and the research facilitator and medical advisers

Staff induction processes related to the functioning of the BPSU have been reviewed and are fully documented. Induction processes for new medical advisers and committee members are operated but less well-documented and this is a potential risk in terms of the resilience of the system.

4.4.4 Time spent on maintaining the surveillance system

Performance indicator	Assessment method	Expected outcome
<ul style="list-style-type: none"> Time spent on maintaining the system 	<ul style="list-style-type: none"> Description of the time spent on maintaining the system 	<ul style="list-style-type: none"> A report of the time spent on maintaining the system by the research facilitator

A review of the activities and time required for the various functions of the BPSU Research Facilitator is as below:

Table 15 Activities and time required for the functions of the BPSU Research Facilitator

	Activity	Days spent/month
1	Entering orange negative cards	1.5
2	Entering positive cards and notifying investigators	1
3	Entering case notifications / follow up details	0.5
4	Identifying new doctors, entering on database and preparing packs	2
5	Monthly reminders (preparing email and entering responses)	1
6	General database maintenance	0.5
7	Sir Peter Tizard paper work	0.25
8	Preparing for York conference	0.5
9	Attending BPSU EC meeting	0.5
10	Preparing a new study	1.5
11	Dealing with invoices/accounts etc	0.25
12	Preparing the quarterly bulletin	2
13	Organising monthly BPSU EC papers	2
14	Producing the bi-monthly report	0.5

The medical adviser for non-communicable disease typically spends 1.5 programmed activities (PA) per week and the medical adviser for communicable disease 0.5 PAs per week on BPSU related activity. NB 1 PA=3 hours.

4.4.5 Reported difficulties by paediatricians in reporting cases

Performance indicator	Assessment method	Expected outcome
<ul style="list-style-type: none"> Reported difficulties by paediatricians in reporting cases 	<ul style="list-style-type: none"> Closed questionnaire to BPSU researchers 	<ul style="list-style-type: none"> A report on reported difficulties by paediatricians in reporting cases

Table 16 Difficulty in identifying cases on receipt of questionnaire

How difficult was it to identify patients reported when questionnaire arrived	Total	%
Difficult and I was not able to identify the patient	10	4
Difficult but managed to identify patient	47	20
NO, it has never been difficult (please skip the next question)	161	68
Denominator (respondents who had seen a case in past 2 years)	237	

Although 57 respondents said it was difficult, only 51 volunteered reasons for this as below:

Table 17 Reason for difficulty in identifying cases

If it was difficult to identify cases, why was this?	Total	%
Could not find the orange card with the patient's name	17	30
Did not keep a record of the patient's name on clinicians' section of the orange card	15	26
Other please specify	10	18
You could not obtain the patient's notes	9	16
Blank	6	11
Total	57	
Other reasons for not reporting cases		
<p><i>The principal coordinator failed to help even after several phone calls to give patient details</i></p> <p><i>Time consuming</i></p> <p><i>Tracking patients notes locally</i></p> <p><i>Study could not identify patient other than by initials</i></p> <p><i>Working on 2 sites made identifying details difficult</i></p> <p><i>Getting the notes back is time consuming, finding the information is time consuming, and there is no obvious immediate benefit</i></p> <p><i>Patient was managed in 2 different centres</i></p>		

Interpretation of results: simplicity

- Contact with the BPSU office and medical advisers while developing the study proposal was found by the majority, to be useful and were regarded as being timely.
- Staff induction processes related to the functioning of the BPSU are established and documented for BPSU office staff but are less well-documented for medical advisers and committee members. This is a potential risk to the resilience of the system, particularly at times of changeover.
- Over 50% of investigators found the current documentation useful although one third did not look at it at all suggesting improvements could be made.
- Systems for maintaining a record of notified cases on the Orange card tear-off slip appear robust, as only 4% of clinicians reported difficulty with re-identifying a child whom they had previously notified. Any future electronic systems for reporting should take into account the need for a similar aide-memoire.

Recommendations:

1. Communication with the BPSU office and with the medical advisers is recognised as a useful part of the study initiation process and should be encouraged and developed further. It should also be made clear to applicants that attendance at a meeting of the BPSU EC to discuss their proposal will be expected at some stage.
2. The possibility of explicitly stating within the P1 forms the possibility defending a study proposal at a BPSU executive meeting should be considered by the EC.
3. Induction procedures for medical advisers and new committee members should be reviewed and revised where necessary. Appropriate supervisors for the induction of new members should be identified as part of this process.
4. Current documentation for applicants should be reviewed and the preparation of shorter or more accessible versions considered.

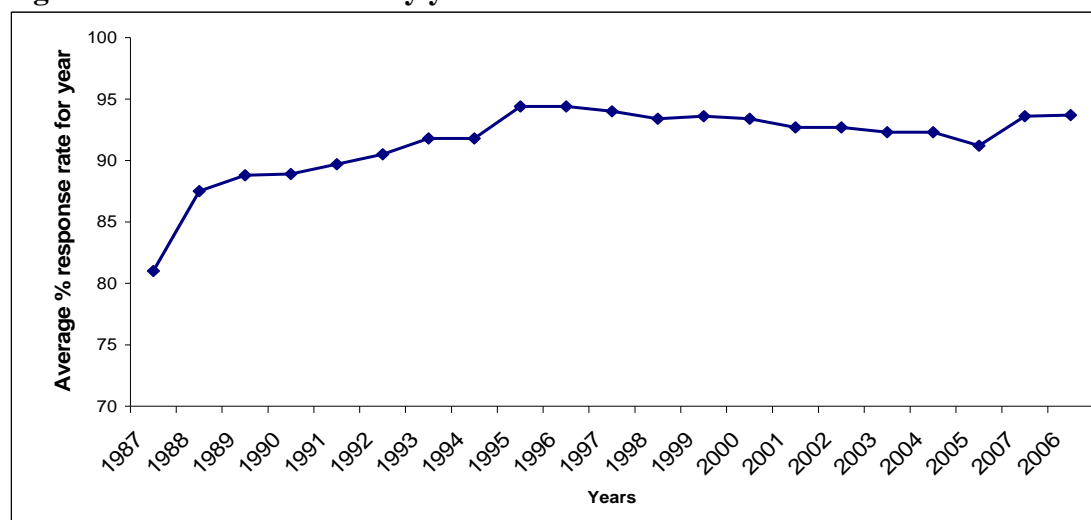
4.5 Stability

Over 93% of consultant paediatricians on the BPSU reporting list respond each month to the card mailing; a similar percentage return questionnaires after reporting a case. These percentages have remained around this level with little variation over recent years of operation of the BPSU.

4.5.1 Card return rates

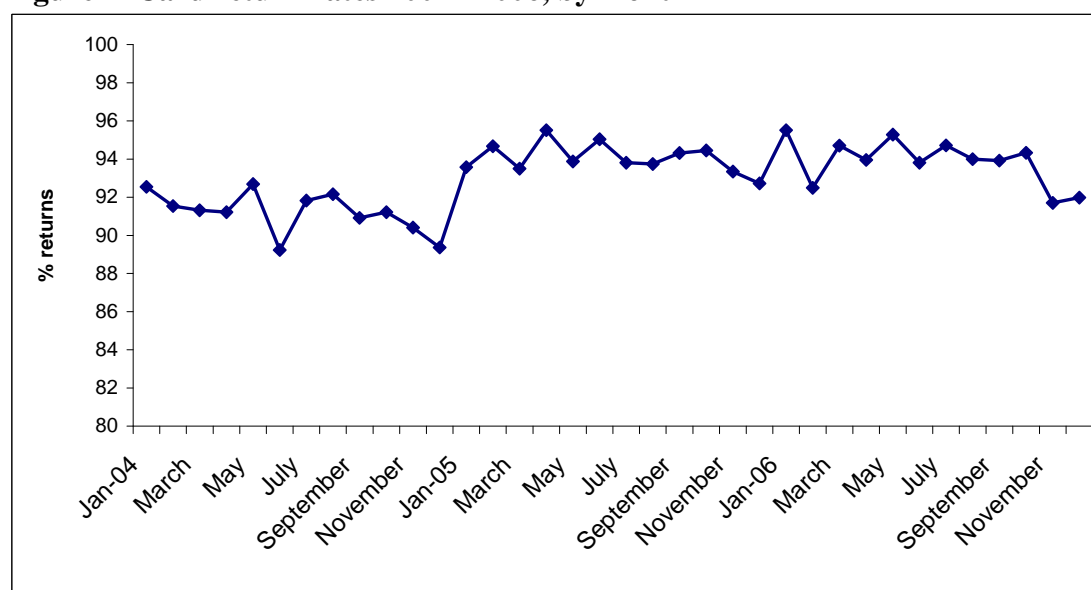
Performance indicator	Assessment method	Expected outcome
<ul style="list-style-type: none"> Card returns by month 2004 – 06 Card return rates by year since 1986 -2006 	<ul style="list-style-type: none"> Review of participation rates over time 	<ul style="list-style-type: none"> A report analysing participation rates over time

Figure 6 Card return rates by year 1987 to 2006



During the 20 years of reporting there was an increase in card return rates over the initial years of surveillance culminating at nearly 95% by the 9th year of surveillance. A slow decline was observed until 2004 after which there was again a steady increase in return rates (see Figure 7 below).

Figure 7 Card return rates 2004 - 2006, by month



Over the past three years there has been an improvement in the response rate most notably in 2005 and 2006 coinciding with the employment of a full time research facilitator.

Until recently, paper reminders have been sent out for those who do not return cards for two subsequent months. These reminders are now sent out by e-mail and further improvements in return rates are expected.

Stability of Printing and Mailing of the cards

Performance indicator	Assessment method	Expected outcome
<ul style="list-style-type: none"> ▪ Periods of full functionality of surveillance system since inception. 	<ul style="list-style-type: none"> ▪ Review the times the system has been fully functional since inception and any interruptions of surveillance and its causes 	<ul style="list-style-type: none"> • A report describing any failures/delays in the sending out of the orange card (e.g. due to workforce absences etc) in the past 5 years.
		<ul style="list-style-type: none"> ▪ Procedures to ensure resilience of the system to these failures.
		<ul style="list-style-type: none"> ▪ Protocols on management of system failures, should they occur.
<ul style="list-style-type: none"> ▪ Times of full functionality of the database supporting surveillance since inception. 	<ul style="list-style-type: none"> ▪ Description of any computer/database failures and procedures to ensure stability, resilience and accuracy of databases in the future 	<ul style="list-style-type: none"> ▪ A report describing any failures of the database
		<ul style="list-style-type: none"> ▪ Procedures to ensure resilience of the system to these failures (data back-up procedures)
		<ul style="list-style-type: none"> ▪ Protocols on what to do to manage such failures, should they occur.

For the past 5 years the orange cards have been produced by a company called CONTINUA – Appendix 2). They are a subsidiary of Rank Xerox who have printed the cards for over 10 years

At the middle of each month 3 files are sent via e-mail to CONTINUA. The files are:

1. A WORD template of the orange card;
2. An MS EXCEL CSV file of mailing list of the current respondents.
3. A set of printing instructions

A pdf template of the orange card is emailed back to the office within a day or two for review. The template is checked to make sure it has the correct month and study details. Once agreed we e-mail back our approval for the card to be printed.

The final printed cards are returned to the office, usually within 2 days. The whole process takes no more than 1 working week. The cards are delivered and subsequently picked up by the mail out house (SOS direct – Appendix 3) for distribution. A few doctors who require a reply paid envelope are identified and one is added to each card. The cards destined for the Republic of Ireland are identified and a self-addressed envelope is included in their posting.

Recorded printing problems in the past five years:

No errors of printing due to the printing house have occurred in the past five years. On one occasion a study that had come to the end of its surveillance was not removed from the next month's template list. The investigators were informed of this, and no adverse consequences occurred.

Recorded mailing problems in the past five years:

In 21 years there has not been a situation where the monthly card has not gone out for a particular month.

Cards are sent out monthly by SOS Direct. No problems have been reported to us from the mail house. The cards are picked up a few days before the end of the month and are then sent out second class with the aim to reach the respondent by the last day of the month. Occasionally the cards have had to go out a day or to early or late e.g. over Christmas. Problems have occasionally been reported in receiving returned cards when there is a problem with the Post Office. In 2007 there were a couple of postal strikes that appear to have affected the return of the cards to some extent. The card response rate remained over 90% despite the strikes so it there was no significant detrimental affect resulting from the postal strikes.

Card returns are monitored and if there is a problem in a specific month. The BPSU is in a position to send e-mails around in place of the orange card to identify whether there are any further cases to report. The need to do this has not arisen. Normally, when cards are not received from a particular respondent for two subsequent months, they are sent an e-mail reminder and a letter if email fails. Various explanations have been offered as to why the cards had not been returned including, leave, sickness, lost in the NHS postal office, secretary not returning card and absence of a secretary to return card.

Managing printing and postal failures:

The current practice as it exists: If for some reason the BPSU research facilitator is not in a position to arrange for the printing and postage of the orange cards e.g. due to leave or sickness the work will be undertaken by the scientific coordinator.

If for some reason the scientific coordinator was unavailable as well, the Principle Research Officer of the college research division will contact the printing house and

1. ask them to use the existing card template but
2. ask them to amend the Month details on the card and then

3. produce a print run using the data supplied the previous month.

The principal research officer would also contact the mail house and

1. arrange a pick up for delivery
2. ask for all the cards to be sent out as is

These instructions for managing the print run and mail out are written in a manual on how to use the BPSU system. A hard copy is on the research facilitators' desk and it is also saved on the shared 'H' drive of the College server.

Recorded database problems in the past five years:

The orange card system is managed by a bespoke integrated surveillance system, written for the BPSU by the scientific coordinator and Geoff Branch and Co (Appendix 3) and called the "Epidemiology Programme". The programme is based on an MS Access platform and consists of two linked databases containing five active tables, queries and reports. The programme logs and categorises current case and card reports. It analyses data received to produce reports and graphics. The system also generates the monthly mailing list and monitors compliance with the system.

A full user manual exists and a hard copy is on the research facilitators' desk and it is also saved on the shared 'H' drive of the College server.

At the time of this evaluation, the BPSU office did not systematically keep individual case study reports data, electronically, after completion of a study; such records were only kept by the study investigators. It should be noted that the BPSU office DOES NOT hold any patient clinical information.

Programming problems:

One problem arose in June 2007 when a new member of staff was being trained in the use of the system without the supervision of the scientific coordinator and some data was deleted. It was possible to use RCPCH back up protocol* to restore the data and there was no loss of data.

* College back up protocol: File back ups are made every night at midnight onto a tape and these are available the next day if required. A weekly back up and monthly back up is also taken and these tapes are held off site.

Lessons learned from this episode were documented and implemented:

1. Those with permission to change programme settings were made explicitly clear.
2. The College back up protocol needs to be written down and that staff should be made aware of the protocol
3. Monthly back-ups of the BPSU programme should also be made by the BPSU office and saved on the H: drive
4. During future development whenever extensive editing of a table is required within the programme a copy will be made and saved. This should be done only by the scientific coordinator or by Geoff Branch and Co.

4.5.2 Failures to report results 5 years after completion of study

Performance indicator	Assessment method	Expected outcome
<ul style="list-style-type: none"> ▪ Failures to report results 5 years after completion of study. 	<ul style="list-style-type: none"> ▪ Description of failures to report results 5 years after completion of study. 	<ul style="list-style-type: none"> ▪ A report describing studies that have failed to publish results 5 years after study termination

Although it is not a primary aim of the BPSU to see all studies published in peer review journals, this is encouraged. All studies will also publish their data in the BPSU annual report, which is lodged at the British Library. The average time which elapses before a paper is published in a peer review journal following the end of the surveillance period is 2.9 years, however this often includes a year of follow-up to complete data collection after surveillance has ended.

For the evaluation, PubMed was searched for any records of BPSU studies. There were only nine studies which had been completed for more than two years, yet had not published in a peer review journal. The reasons for non-publication are outlined below. All studies have presented data at meetings or in the BPSU annual report and study findings are therefore in the public domain.

1. X-linked anhydrotic-ectodermal dysplasia - Undertaken in 1986 for 3 months as a pilot study.
2. Lowe syndrome –Undertaken in 1986 for 18 months. No details of publication attempts.
3. Acute rheumatic fever – Undertaken in 1990 for 12 months. Investigator retired and publication did not result.
4. Long term parenteral nutrition –Undertaken in 1992 for 3 months. Data was presented at the RCPCH Scientific meeting
5. Haemophagocytic Lymphohistiocytosis – Undertaken in 1991 for 3 years. Data was presented at the RCPCH Scientific meeting. Even though lead investigator has since died, co-investigators still hope to publish.
6. Neonatal necrotising enterocolitis – Undertaken in 1993 for 12 months. Paper rejected by ADC, never resubmitted. Lead investigator considering re-drafting for publication.
7. Congenital cytomegalovirus – Undertaken in 2001 for 2 years. Currently considering drafting a paper.
8. Thrombosis in childhood – Undertaken in 2001 for 2 years. Paper being drafted.
9. Stroke in childhood – Undertaken in 2001 for 2 years. Paper being drafted.

Interpretation of results

- Over 93% of consultant paediatricians respond each month to the card mailing with a similar number returning questionnaires after reporting a case. These percentages have remained around this level with little variation over the years.
- Over the past three years there has been an improvement in the response rate most notably in 2005 and 2006 coinciding with the employment of a full time research administrator
- No significant problems with printing, programming or mailouts have been identified and good back-up systems are in existence and documented.

Recommendations:

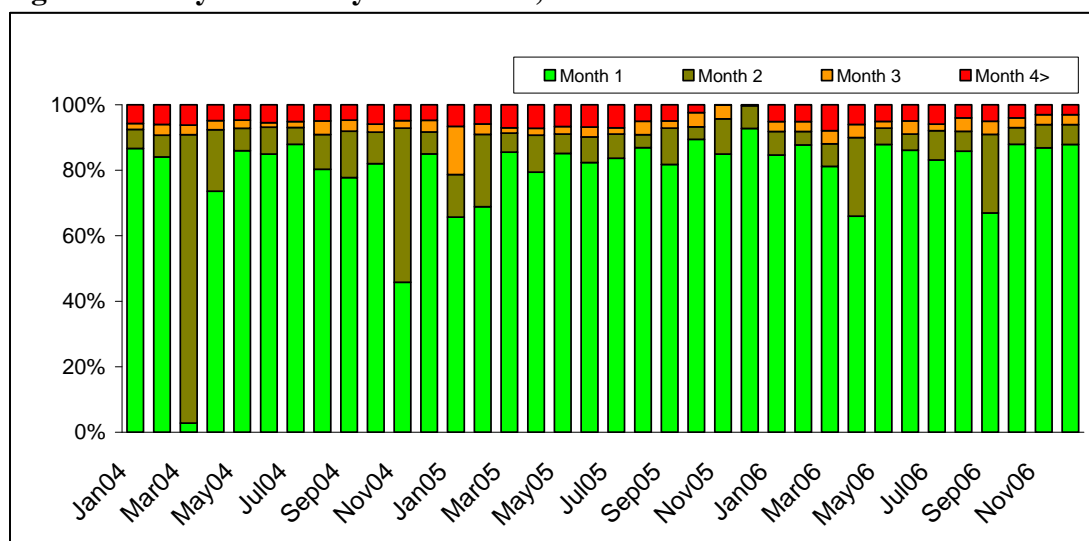
1. The card return rate is dependent on active follow-up by the Research facilitator and needs to be maintained.

4.6 Timeliness

4.6.1 Monthly card delay

Performance indicator	Assessment method	Expected outcome
<ul style="list-style-type: none"> Card response delay Delay in notifying cases 	<ul style="list-style-type: none"> Review of timeliness of surveillance processes along all points of the surveillance loop 	<ul style="list-style-type: none"> A report analysing timeliness of surveillance processes along all points of the surveillance loop

Figure 8 Delay in monthly card return, 2004 - 2006



Of all the cards returned, over 90% were returned within the first two months in 32 of the 36 months evaluated, and over 80% in 35 of the 36 months. NB Cards returned within each time frame are expressed as a percentage of all cards returned for a specific month (Figure 8).

Figure 9 Case notification delay for studies on the card in November 2007

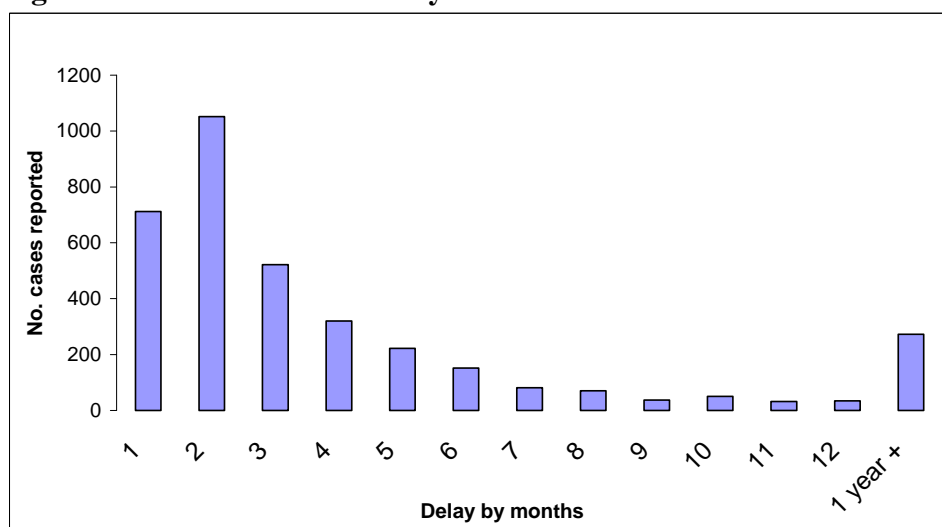


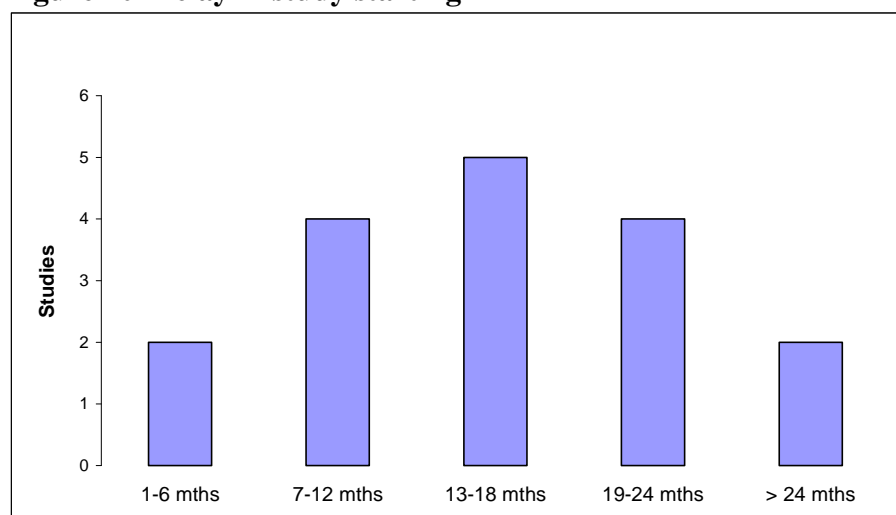
Figure 9 represents the delay in the BPSU office receiving an update on the case status of a report using data only from those studies underway at the time of the evaluation. The investigator returns most notification confirmations to the BPSU office within 3 months. This delay is dependent on the study design and management by the investigator. It does not measure the delay between the reporting clinician completing the questionnaire and its return to the investigator, which would be of interest, but would only be available for individual studies. Specific studies such as PIND require confirmation of cases via committee before the BPSU is informed therefore cases would take considerable longer to be notified. Other reasons for delay include awaiting microbiological confirmation for infectious diseases. All study investigators tend to return more notifications at the end of the study as they chase up outstanding reports, hence the increase at 12 months. These data are not systematically for each study at present.

4.6.2 Study application approval times

Performance indicator	Assessment method	Expected outcome
<ul style="list-style-type: none"> Audit study application approval times 	<ul style="list-style-type: none"> Review of timeliness of the BPSU executive committee in reaching decisions on study proposals in the different stages 	<ul style="list-style-type: none"> A report of timeliness of the BPSU executive committee in reaching decisions on study proposals in the different stages

These data were collected by looking through past minutes and the bi-monthly report tables. It is difficult to interpret the data in terms of delay being due to the BPSU process or the applicant. In some cases the delay may be due to the difficulty in agreeing a workable case definition. Study applications with the shortest processing times are from those who have applied to the system previously or are on the Executive Committee and therefore may have a better knowledge of the process, e.g. vitamin K deficiency bleeding (VKDB), congenital adrenal hyperplasia (CAH), toxoplasmosis and genital herpes.

Figure 10 Delay in study starting



Approval times for individual studies vary widely. The mean approval time is 16 months (i.e. from submission of phase 1) with a range of 6 to 34 months. The two studies with the shortest approval times were CAH and Toxoplasmosis (6 months each) while scleroderma and Langerhans cell histiocytosis (LCH) had the longest approval periods of 26 and 34 months each.

4.6.3 Perception of timeliness

Performance indicator	Assessment method	Expected outcome
▪ Perception of timeliness	▪ Closed questionnaire to participating paediatricians	▪ A report of the perception of timeliness of the BPSU among its reporting base

Table 18 Difficulty in identifying cases on receipt of questionnaire

Time between reporting a case and receipt of a questionnaire for that case	Total	%
Delayed (more than 2 weeks)	77	32
Prompt (less than 2 weeks)	138	58
blank	22	9
Denominator (respondents who had seen a case in past 2 years)	237	

Table 19 Conditions most commonly associated with delays

Progressive Intellectual and Neurological Degeneration	15
HIV/AIDS	8
Congenital Adrenal Hyperplasia	3
Malaria in Childhood	3
MCADD	3
Non-type 1 Diabetes	3
Scleroderma	3
Thyrotoxicosis	3
Idiopathic Intracranial Hypertension	2
MRSA	2
Neonatal Herpes Simplex Virus (HSV) Infection	2
Neonatal Herpes	1
Vitamin K Deficiency Bleeding	1
<i>Do not remember</i>	<i>14</i>

Interpretation of results: timeliness

- Orange cards are returned promptly by clinicians with over 90% being returned within two months of a mailout on most occasions.
- The time for approval of study applications by the BPSU varies markedly from 6 to 34 months and averages over one year.
- One-third of clinicians report a delay of more than two weeks in receiving questionnaires from investigators after reporting a case.

Recommendations

1. There is considerable delay in approving applications despite revised procedures. Different stages of the application procedure should be reviewed and assessed for improvements in the timeliness of processes.
2. Further objective monitoring of the time taken to respond to case notifications is required to confirm or refute clinicians' perceptions about delay. Electronic methods of sending out questionnaires and completing questionnaires could also be explored as a means to improving response times.

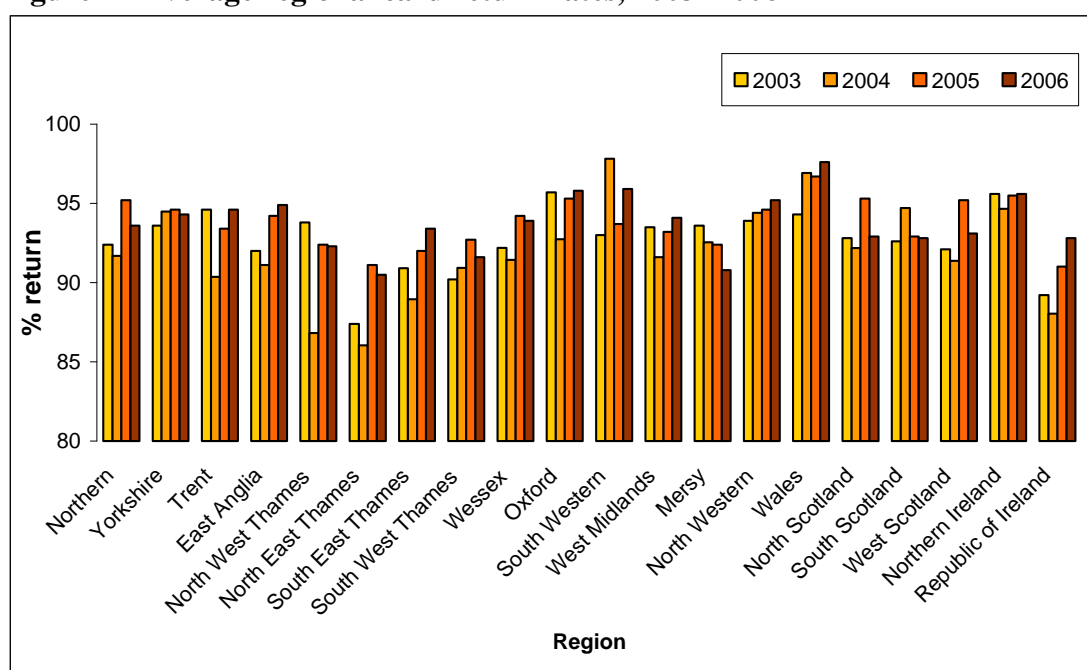
4.7 Representativeness

Performance indicator	Assessment method	Expected outcome
▪ Card return rates by region	▪ Review of participation rates by region	▪ A report analysing participation rates by region

4.7.1 Card return rates by region

The data was taken from the regional analysis of card return rates as identified in the BPSU annual reports for this period. The graph shows the last 4 complete response years for each region.

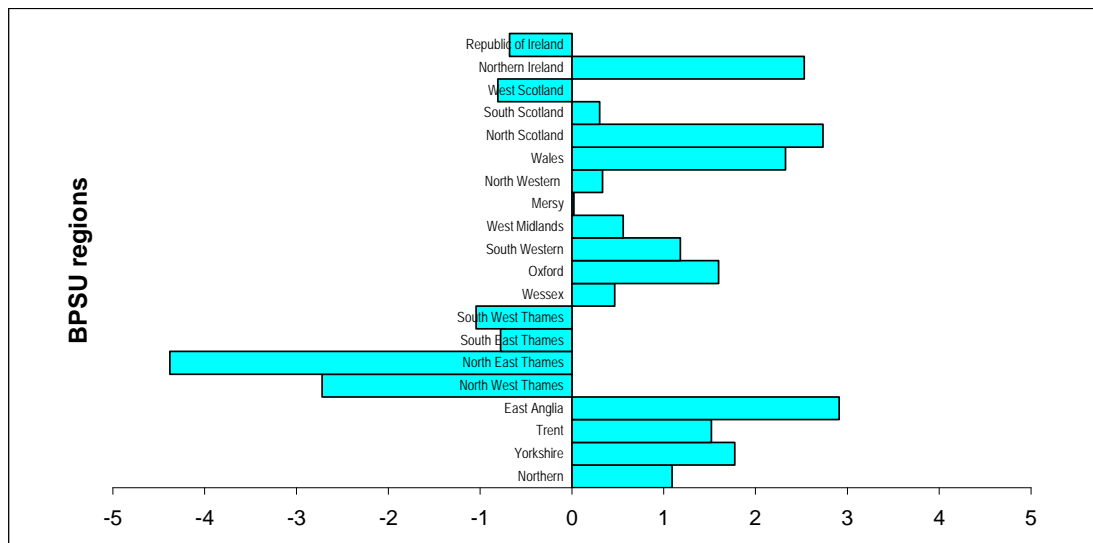
Figure 11 Average regional card return rates, 2003 -2006



Data was taken from the regional return rates as stated in the annual reports. Although there is some fluctuation, paediatricians in all the regions return over 85% of cards sent to them.

The next graph shows the regional deviation from the mean return over 20 years. The annual average mean card return for this period was 92.1%. It highlights the comparatively lower return rate within the London area. Interestingly, over 20 years East Anglia is the highest reporting region, though this has not been the case in recent years. Wales is now regularly the highest responding region, followed by Oxford and Northern Ireland.

Figure 12 Regional deviation in card-return rates over 20 years



Interpretation of results: representativeness

- Although there is some fluctuation, paediatricians in all the regions return over most of the cards sent to them.

Recommendations

1. Card return rates should be continually closely monitored so that any reduction in rates by a particular region can be rapidly identified.

4.8 Acceptance

4.8.1 Numbers and affiliation of paediatricians applying for and undertaking BPSU studies

Performance indicator	Assessment method	Expected outcome
<ul style="list-style-type: none"> Numbers and affiliation of paediatricians applying for BPSU studies 	<ul style="list-style-type: none"> Review of affiliation and location of BPSU study applicants. 	<ul style="list-style-type: none"> A report of affiliation and location of BPSU study applicants.

Applications to BPSU studies come from across the United Kingdom and Ireland. During the five year period 2003-2008 twenty-one studies were undertaken through an English institution, four from a Scottish Institute and two from a Welsh institute. No BPSU studies were initiated from Ireland.

The map shows the affiliation of the lead investigator (DGH, teaching hospital or academic institution, HPA/HPS). Thirteen studies were run from investigators based in teaching hospitals (50%), 10 from an academic institution (38%), three from the Health Protection Agency/Scotland and just one from a district general hospital.

Figure 13 Institutional affiliation of the lead investigator for studies undertaken (2003-2008)



Table 20 Institutional affiliation of the lead investigator for studies undertaken (2003-2008)

Condition - Accepted	Location of lead Investigator	Institution Type
Vitamin K deficiency bleeding III	Exeter	Teaching hospital
Congenital cytomegalovirus (cCMV)	London	Academic institution
Thrombosis in childhood	Glasgow	Teaching hospital
Internal abdominal injury due to child abuse	Cardiff	Teaching hospital
Suspected fatal adverse reactions	Nottingham	Teaching hospital
Complications to varicella requiring hospitalisation	Glasgow	Gov (HPS/HPA)
Invasive fungal infections	Dundee	Teaching hospital
Congenital Toxoplasmosis	London	Gov (HPS/HPA)
Childhood Tuberculosis	London	Teaching hospital
Severe Hyperbilirubinaemia	Wirral	Teaching hospital
Langerhans Cell Histiocytosis (LCH)	Newcastle	Teaching hospital
Thyrotoxicosis	Dundee	Teaching hospital
Non-type 1 Diabetes	London	Academic institution
Early onset eating disorders (EOED)	London	Teaching hospital
Neonatal Herpes Simplex Virus (HSV) Infection	London	Academic institution
Malaria in Childhood	London	Academic institution
MRSA	London	Gov (HPS/HPA)
Scleroderma	Manchester	Academic institution
HIV/AIDS	London	Academic institution
Progressive Intellectual and Neurological Degeneration (PIND)	Cambridge	Teaching hospital
Congenital Rubella	London	Academic institution
Fetomaternal Alloimmune Thrombocytopenia (FMAIT)	Oxford	Academic institution
Vitamin K Deficiency Bleeding	Exeter	Teaching hospital
Genital Herpes	Ipswich	Teaching hospital
Idiopathic Intracranial Hypertension (IIH)	Cardiff	DGH
Congenital Adrenal Hyperplasia	London	Academic institution
Intussusception in children less than 12 months of age	London	Academic institution

Table 21 Location from where unsuccessful applications were received

Condition – Rejected 2003-08	Location of lead Investigator	Institution Type
NEC in VLBW	Dundee	Teaching hospital
Coning following encephalitis	Rotherham	DGH
Kawasaki Disease	London	Academic institution
Rickets	Liverpool	Teaching hospital
Childhood Hypernatraemia	London	Academic institution
Gastroschisis	Oxford	Academic institution
Childhood Lymphoproliferative Disease	London	Teaching hospital
Vein of Galen Aneurysmal Malformation	Glasgow	Teaching hospital
Aicardi Goutières syndrome	Bradford	Teaching hospital
Primary Eosinophilic Gastrointestinal Disorders (PEGID)	Hertfordshire	DGH
Fractures in Children with Cerebral Palsy	Northampton	DGH
Fetal Alcohol Syndrome	Oxford	Academic institution
Reduced Levels of Consciousness - single survey	Belfast	Teaching hospital
Thrombo-embolic arterial or venous compromise in babies less than 32 weeks gestation	Newcastle	Teaching hospital
Management of neonatal deliveries and neonatal outcome in women requiring crash caesarean sections secondary to cardiac arrest	Coventry	Teaching hospital
Narcolepsy	London	Teaching hospital
Ischemic limbs & thrombolysis	Newcastle	Academic institution
Inflammatory bowel disorder	Bristol	Teaching hospital

Of the 18 rejected studies 3 (17%) were from a DGH, 10 (55%) were from a teaching hospital and 5 (27%) were from an academic institution.

4.8.2 Applications to the Sir Peter Tizard Bursary Scheme

As part of the BPSU's commitment to increase research capacity within the NHS it was decided, in 2003, to introduce a research bursary. Entitled the Sir Peter Tizard bursary (SPTB) after one of the founders of the unit, it is aimed at junior doctors and newly qualified consultants.

The bursary gives the successful applicant the opportunity to learn how to undertake epidemiological surveillance of a rare condition. Funds are also made available for relevant training opportunities. Six bursaries have now been awarded however the number of applicants for each bursary has not increased Table 2. We suspect that this may have more to do with recent changes in junior doctors' contracts which restrict time available to set aside for research. We have already seen considerable delays in getting studies started because of the lack of time available to develop the protocols

At the time of this evaluation only two of the bursary projects had been concluded so feedback from the recipients has not yet been systematically reviewed, though anecdotally involvement in the bursary has been view positively.

4.8.3 Perception of participating paediatricians on ease and usefulness of outputs of BPSU surveillance

Performance indicator	Assessment method	Expected outcome
<ul style="list-style-type: none"> Perception of participating paediatricians on ease and usefulness of outputs of BPSU surveillance 	<ul style="list-style-type: none"> Closed questionnaire to participating paediatricians 	<ul style="list-style-type: none"> Reported perception of participating paediatricians on the usefulness of outputs of BPSU surveillance

Table 22 Application to the BPSU Bursary 2003-2009

Bursary Year	No. of applicants	Condition studied	Status
2003-04	12	Thyrotoxicosis	Completed
2004-05	15	Malaria	Completed
2005-06	6	Idiopathic Intracranial Hypertension	Ongoing
2006-07	17	Toxic shock syndrome	Ongoing
2007-08	6	Glutaryl-Coenzyme A dehydrogenase deficiency	Yet to commence
2008-09	4	Surgical ligation of the patent ductus arteriosus in premature infants	Yet to commence

4.8.4 Perception of participating paediatricians on ease and usefulness of outputs of BPSU surveillance

The BPSU feeds back the results of its work through the Annual Report, bulletins, website, protocol cards and peer review journals. 48% of respondents were very satisfied with this whilst a further 48% were neither satisfied nor dissatisfied. These highlights an area where improvements could be made (Table X).

The BPSU bulletins are the most utilised BPSU output and 88% of those that read them find them useful. The BPSU website is rarely used by clinicians, and fared quite badly when compared to other sources of information. This could be considered to be as a result of a dislike of receiving information in an electronic format. However, it is apparent this is not the case as 83% of clinicians would be willing to receive information in an electronic, rather than, or in addition to, information in paper format. It is probable that part of this result is due to some clinicians are not aware of information about the BPSU being available on the web.

Several clinicians suggested that it would be useful to have brief email newsletter with hyper links to further information on the website and some asked for brief **'take home'** message for clinical practice and briefer summaries on the website. To this end BPSU activities are now being included in the Presidents email to College members. Details on these activities are hyperlinked to the relevant page of the BPSU website.

Table 23 Frequency of accessing information sources and the usefulness of these information sources

Frequency accessed	%				%	
	Often	Sometimes	Never		Useful	Not Useful
Archives of Disease in Childhood	66.7	30.5	2.8		95.3	4.7
Other peer reviewed journals	26.3	52.7	21.0		85.5	14.5
BPSU Bulletin	50.1	45.2	4.7		88.3	11.7
BPSU Annual Report	34.9	54.0	11.1		79.2	20.8
Presentations at the RCPCH Annual Meeting	11.3	60.3	28.4		79.2	20.8
Scientific meetings, conferences and congresses	9.6	52.9	37.5		77.2	22.8
BPSU website (http://bpsu.inopsu.com)	1.4	16.8	81.9		36.9	63.1
HPA Communicable Disease Report (Online)	3.8	33.3	62.9		53.0	47.0

4.8.5 Ease of completion of BPSU questionnaire

Of the 454 respondents, 237 (52%) describe having reported at least one case in the past 2 years.

Table 24 Ease of completion of BPSU questionnaire

On identifying a case did you find the BPSU questionnaire easy to complete	Total	%
YES	192	92
NO	17	8
No response	39	

Table 25 Conditions as reported within the past 2 years

Progressive Intellectual and Neurological Degeneration	52	29
HIV/AIDS	39	22
Idiopathic Intracranial Hypertension	14	8
hyperbilirubinaemia	9	5
MCADD	7	4
Neonatal Herpes Simplex Virus (HSV) Infection	6	3
MRSA	5	3
Thyrotoxicosis	5	3
Congenital Adrenal Hyperplasia	5	3
FMAIT	5	3
Malaria in Childhood	5	3
Scleroderma	5	3
Vitamin K Deficiency Bleeding	4	2
Langerhans Cell Histiocytosis	4	2
Non-type 1 Diabetes	4	2
Medium Chain Acyl CoA Dehydrogenase Deficiency	3	2
Neonatal Herpes	3	2
Genital Herpes	2	1
Intussusception	1	1

Interpretation of results: Acceptance

- Card return rates are good across the UK and Ireland although consistently lower in the London region.
- The majority of clinicians report that questionnaires are easy to complete.

Recommendations

The BPSU Executive Committee

1. should continue to scrutinise and recommend piloting of questionnaires to ensure that the burden of completion remains low. Consideration should be given to developing a more standardised format and layout to be adapted and used across different studies.
2. identify ways in which to encourage applications from outside of England and from clinicians other than those in tertiary centres
3. increase awareness of the Sir Peter Tizard bursary amongst junior doctors. Consideration may need to be given to widen the qualification criteria for acceptance into the bursary scheme
4. encourage speedier progress of the bursary protocol

5 References

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6 Appendix 1

6.1 Indicators to measure performance attributes for BPSU surveillance

Attribute	Performance indicator	Assessment method	Expected outcome
Usefulness	<ul style="list-style-type: none"> Number and classification (peer review publication or not) of surveillance and research outputs from BPSU surveys 	<ul style="list-style-type: none"> Review and classify BPSU's surveillance outputs 	<ul style="list-style-type: none"> A database of all surveillance outputs (papers, presentations, posters, reports, as well as relevant newspaper articles) from the BPSU that is accessible whenever required.
	<ul style="list-style-type: none"> Quantification of BPSU's contribution to international collaborations i.e. other surveillance units, INOPSU 	<ul style="list-style-type: none"> Review BPSU's contribution to international collaborations i.e. other surveillance units, INOPSU 	<ul style="list-style-type: none"> A report (with a time line) of the BPSU's involvement in INOPSU A list of international collaborative papers.
	<ul style="list-style-type: none"> The reporting base's perception on the usefulness of BPSU surveillance 	<ul style="list-style-type: none"> Closed questionnaire to participating paediatricians 	<ul style="list-style-type: none"> Perception of the reporting base on the usefulness of BPSU surveillance
	<ul style="list-style-type: none"> BPSU's researchers' perception on the usefulness of BPSU surveillance. 	<ul style="list-style-type: none"> Closed questionnaire to BPSU researchers' 	<ul style="list-style-type: none"> Perception of BPSU researchers on the usefulness of BPSU surveillance
	<ul style="list-style-type: none"> Hits on the BPSU website 	<ul style="list-style-type: none"> Review website hits 	<ul style="list-style-type: none"> Analysis of website hits over time
Data Quality	<ul style="list-style-type: none"> Availability and accessibility of BPSU surveillance data and output outputs 	<ul style="list-style-type: none"> Review storage systems, organisation and maintenance for surveillance data and surveillance outputs. 	<ul style="list-style-type: none"> An "End-of-study protocol" that includes all relevant data that should be collected for each study which will be stored in an "End of study directory".

			<ul style="list-style-type: none"> ▪ An “End of study directory” for each study that will contain: <ul style="list-style-type: none"> ○ A minimum dataset for each study with a line listing of each case reported with final classification. ○ All other relevant information and material on each study.
			<ul style="list-style-type: none"> ▪ A database with summary information of all completed studies ▪ Agreed processes on where to store these and how to manage them
	<ul style="list-style-type: none"> ▪ Reported card return and case-reporting behaviour 	<ul style="list-style-type: none"> ▪ Closed questionnaire to BPSU researchers’ 	<ul style="list-style-type: none"> ▪ Reported card return and case-reporting behaviour of the reporting base
Flexibility	<ul style="list-style-type: none"> ▪ Number of past studies that were a direct response to new public health objectives and consensus view on success of these responses. 	<ul style="list-style-type: none"> ▪ Review of BPSU studies in terms of its “response to new public health objectives” 	<ul style="list-style-type: none"> ▪ Review of BPSU studies in terms of their “response to new public health objectives”
	<ul style="list-style-type: none"> ▪ The BPSU's flexibility in adapting its methodology in order to undertake specific studies for which routine BPSU methodology is not adequate. 	<ul style="list-style-type: none"> ▪ Review BPSU's flexibility in adapting its methodology in order to undertake specific studies for which routine BPSU methodology is not adequate. 	<ul style="list-style-type: none"> ▪ A report of the BPSU's involvement in studies where flexibility in its methods were necessary.
	<ul style="list-style-type: none"> ▪ The reporting base’s willingness to report rapidly via accelerated systems in the event of a public health emergency 	<ul style="list-style-type: none"> ▪ Closed questionnaire to participating paediatricians 	<ul style="list-style-type: none"> ▪ A report on the perception of the reporting base on its ability to adapt to accelerated reporting systems
Simplicity	<ul style="list-style-type: none"> ▪ Ease of data flow through the system 	<ul style="list-style-type: none"> ▪ Review the BPSU surveillance loop for any obvious bottlenecks 	<ul style="list-style-type: none"> ▪ A description of the BPSU surveillance loop for any obvious bottlenecks.

	<ul style="list-style-type: none"> ▪ Ease of application process 	<ul style="list-style-type: none"> ▪ Closed questionnaire to BPSU researchers 	<ul style="list-style-type: none"> ▪ Perception of BPSU researchers on the ease of the application process
	<ul style="list-style-type: none"> ▪ Availability of processes and procedures for staff induction and training to maintain the resilience of the system. 	<ul style="list-style-type: none"> ▪ Review availability of processes and procedures for staff induction and training 	<ul style="list-style-type: none"> ▪ Induction and training manual for research division staff and the research facilitator and medical advisers
			<ul style="list-style-type: none"> ▪ Documentation of surveillance processes.
	<ul style="list-style-type: none"> ▪ Time spent on maintaining the system 	<ul style="list-style-type: none"> ▪ Description of the time spent on maintaining the system 	<ul style="list-style-type: none"> ▪ A report of the time spent on maintaining the system by the research facilitator
	<ul style="list-style-type: none"> ▪ Reported difficulties by paediatricians in reporting cases 	<ul style="list-style-type: none"> ▪ Closed questionnaire to BPSU researchers 	<ul style="list-style-type: none"> ▪ A report on reported difficulties by paediatricians in reporting cases
Stability	<ul style="list-style-type: none"> ▪ Periods of full functionality of surveillance system since inception. 	<ul style="list-style-type: none"> ▪ Review the times the system has been fully functional since inception and any interruptions of surveillance and its causes 	<ul style="list-style-type: none"> • A report describing any failures/delays in the sending out of the orange card (e.g. due to workforce absences etc) in the past 5 years.
			<ul style="list-style-type: none"> ▪ Procedures to ensure resilience of the system to these failures.
			<ul style="list-style-type: none"> ▪ Protocols on management of system failures, should they occur.
	<ul style="list-style-type: none"> ▪ Times of full functionality of the database supporting surveillance since inception. 	<ul style="list-style-type: none"> ▪ Description of any computer/database failures and procedures to ensure stability, resilience and accuracy of databases in the future 	<ul style="list-style-type: none"> ▪ A report describing any failures of the database
			<ul style="list-style-type: none"> ▪ Procedures to ensure resilience of the system to these failures (data back-up procedures)
			<ul style="list-style-type: none"> ▪ Protocols on what to do to manage such failures, should they occur.

	<ul style="list-style-type: none"> ▪ Failures to report results 5 years after completion of study. 	<ul style="list-style-type: none"> ▪ Description of failures to report results 5 years after completion of study. 	<ul style="list-style-type: none"> ▪ A report describing studies that have failed to publish results 5 years after study termination
	<ul style="list-style-type: none"> ▪ Card returns by month 2004 – 06 ▪ Card return rates by year since 1986 - 2006 	<ul style="list-style-type: none"> ▪ Review of participation rates over time 	<ul style="list-style-type: none"> ▪ A report analysing participation rates over time
Timeliness	<ul style="list-style-type: none"> ▪ Card response delay ▪ Delay in notifying cases 	<ul style="list-style-type: none"> ▪ Review of timeliness of surveillance processes along all points of the surveillance loop 	<ul style="list-style-type: none"> ▪ A report analysing timeliness of surveillance processes along all points of the surveillance loop
	<ul style="list-style-type: none"> ▪ Audit study application approval times 	<ul style="list-style-type: none"> ▪ Review of timeliness of the BPSU executive committee in reaching decisions on study proposals in the different stages 	<ul style="list-style-type: none"> ▪ A report of timeliness of the BPSU executive committee in reaching decisions on study proposals in the different stages
	<ul style="list-style-type: none"> ▪ Perception of timeliness 	<ul style="list-style-type: none"> ▪ Closed questionnaire to participating paediatricians 	<ul style="list-style-type: none"> ▪ A report of the perception of timeliness of the BPSU among its reporting base
Representativeness	<ul style="list-style-type: none"> ▪ Card return rates by region 	<ul style="list-style-type: none"> ▪ Review of participation rates by region 	<ul style="list-style-type: none"> ▪ A report analysing participation rates by region
	<ul style="list-style-type: none"> ▪ Reported card-return rates by paediatricians 	<ul style="list-style-type: none"> ▪ Closed questionnaire to participating paediatricians 	<ul style="list-style-type: none"> ▪ A report on reported card-return rates by paediatricians
Acceptance	<ul style="list-style-type: none"> ▪ Numbers and affiliation of paediatricians applying for BPSU studies 	<ul style="list-style-type: none"> ▪ Review of affiliation and location of BPSU study applicants. 	<ul style="list-style-type: none"> ▪ A report of affiliation and location of BPSU study applicants.
	<ul style="list-style-type: none"> ▪ Perception of participating paediatricians on the usefulness of outputs of BPSU surveillance 	<ul style="list-style-type: none"> ▪ Closed questionnaire to participating paediatricians 	<ul style="list-style-type: none"> ▪ Reported perception of participating paediatricians on the usefulness of outputs of BPSU surveillance

7 Appendix 2

7.1 Publications

Study	Full titles July 1986-August 2007
Acute Flaccid Paralysis	Salisbury DM, Ramsay ME, White JM, Brown DW. Polio Eradication: Surveillance Implications for the United Kingdom. <i>The Journ. of Infect. Dis</i> ; 175 (Suppl 1): S156-9
AIDS/HIV	Ades AE, Davison C, Holland FJ, Gibb DM, Hudson CN, Nicoll A, Goldberg D, Peckham C. Vertically transmitted HIV infection in the British Isles. <i>Brit. Med. J</i> ; 306: 1296-1299
	Gibb DM, Davison CF, Holland FJ, Walters S, Novelli V, Mok J. Pneumocystitis carinii pneumonia in vertically acquired HIV infection in the UK. <i>Arch. Dis. Child</i> ; 70: 241-4
	Dunn DT, Nicoll A, Holland FJ, Davison CF. How much paediatric HIV infection could be prevented by antenatal HIV testing? <i>J. Med. Screen</i> ; 2: 35-40
	Gibb DM, Faulkner W, Nokes I, Appleby S, Holland FJ, Berry T, et al. Coverage of routine neonatal metabolic screening in children born to women known to be infected with HIV-1. <i>Communicable Disease Report</i> ; 5; R123-4
	MacDonagh SE, Masters JM, Helps BA, Tookey PA, Ades AE, Gibb DM. Antenatal HIV testing in London: policy, uptake and detection. <i>BMJ</i> ; 12: 532-3
	Molesworth A, Tookey P. AIDS and HIV infection (data to end of January 1997). <i>CDR Review</i> ; 7(9), R132-34
	Gibb DM, MacDonagh SE, Tookey PA, Duong T, Nicoll A, Goldberg DJ, Hudson CN, Peckham C. Uptake of interventions to reduce mother-to-child transmission of HIV in the United Kingdom and Ireland. <i>AIDS</i> ; 11, F53-F58
	Gibb DM, MacDonagh SE, Ramyani G, Tookey P, Peckham CS, Ades AE. Factors affecting uptake of antenatal HIV testing in London: results of a multicentre study. <i>BMJ</i> , 316, 259-61
	Nicoll A, McGarrigle C, Brady, AR, Tookey P, et al. Epidemiology and detection of HIV-1 among pregnant women in the United Kingdom: results from national surveillance 1988-96. <i>BMJ</i> ; 316, 253-258
	Williams AJ, Duong T, McNally LM, Tookey PA, Masters J, Miller R, Lyall EGH, Gibb, DM. Pneumocystis carinii pneumonia and cytomegalovirus infection in children with vertically acquired HIV infection. <i>AIDS</i> ; 15, 335-39
	Cliffe S, Tookey P A, Nicoll A, Antenatal detection of HIV: national surveillance and unlinked anonymous survey. <i>BMJ</i> ; 323: 376-7
	Dunn D. Short-term risk of disease progression in HIV-1 infected children receiving no antiretroviral therapy or zidovudine monotherapy: a meta-analysis. <i>Lancet</i> ; 362: 1605-11
	Gibb D M, Duong T, Tookey P A, Sharland M, Tudor-Williams G, Novelli V, Butler K, Riordan A, Farrelly L, Masters J, Peckham C S, Dunn D T. Decline in mortality, AIDS and hospital admissions in perinatally HIV-1 infected children in the United Kingdom and Ireland. <i>BMJ</i> ; 327: 1019-0
	Renewing the focus. HIV and other sexually transmitted infections in the United Kingdom in 2002, London: Health Protection Agency
	Brown AE, Sadler KE, Tomkins SE, McGarrigle CA, Scott LaMontagne D, Goldberg D, Tookey PA, Smyth B, Thomas D, Murphy G, Parry JV, Evans BG, Gill ON, Ncube F. Recent Trends in HIV and other STIs in the United Kingdom: data to the end of 2002. <i>Sex Transm Infect</i> ; 80 (3): 159-66
	Doerholt K, Duong T, Tookey P, Butler K, Lyall H, Sharland M, Novelli V, Riordan A, Dunn D, Walker AS, Gibb DM. Outcomes for HIV-1-infected infants in the UK and Republic of Ireland in the era of effective antiretroviral therapy. <i>Pediatr Infect Dis J</i> ; 25(5): 420-6
	Townsend CL, Tookey PA, Cortina-Borja M, Peckham CS. Antiretroviral therapy and congenital abnormalities in infants born to HIV-1 infected women in the United Kingdom and Ireland, 1990-2003. <i>J AIDS</i> ; 42(1): 91-94
	Health Protection Agency Centre for Infections. The UK Collaborative Group for HIV & STI Surveillance. A complex picture. HIV and other sexually transmitted infections in the UK: 2006. <i>CDR Weekly</i> ; 16(47)

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	Townsend CL, Cliffe S, Tookey PA. Uptake of antenatal HIV testing in the United Kingdom: 2000-2003. <i>J Public Health</i> ; 28: 248-52
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	Hankin C, Newell M, Tookey P. The long-term follow-up of antiretroviral therapy-exposed uninfected children born to HIV-infected women: parents' and health professionals views, <i>AIDS Care</i> : 19 (4): 482-6
	Hankin C, Lyall H, Peckham C, Tookey P. Monitoring death and cancer in children born to HIV-infected women in England and Wales: use of HIV surveillance and national routine data. <i>AIDS</i> ; 23;21(7): 867-9
	Townsend C, Cortina-Borja M, Peckham CS, Tookey PA. Antiretroviral therapy and premature delivery in diagnosed HIV-infected women in the United Kingdom and Ireland. <i>AIDS</i> ; 21(8): 1019-26.
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British Paediatric Surveillance Unit	Hall SM, Glickman M. British Paediatric Surveillance Unit: Sixth Summary Report, <i>Communicable Disease Report</i> ; 88/42
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	Hall SM, Glickman M. British Paediatric Surveillance Unit Fourth Summary Report, <i>Communicable Disease Report</i> . 88/02,
	Hall SM, Glickman M. British Paediatric Surveillance Unit: Fifth Summary Report, <i>Communicable Disease Report</i> ; 88/19
	Hall SM, Glickman M. British Paediatric Surveillance Unit: Seventh Summary Report, <i>Communicable Disease Report</i> 89/15; 89/15
	Hall SM, Glickman M. British Paediatric Surveillance Unit: Eighth Summary Report, <i>Communicable Disease Report</i> : 29
	Hall SM, Glickman M. Report from the British Paediatric Surveillance Unit. <i>Arch. Dis. Child</i> ; 65: 807-809
	Lynn R, Hall SM. The British Paediatric Surveillance Unit: activities and development in 1990 and 199. <i>Communicable Disease Report</i> ; Vol 2, Review No. 13: R145-148
	Hall SM, Roberts C. The British Paediatric Surveillance Unit, <i>Bulletin of the Royal College of Pathologists</i> : 82; 12-17
	Hall SM, Nicoll A. The British Paediatric Surveillance Unit - a pioneering method for investigating the less common disorders of childhood. Report of a seminar held in June 1995, <i>Child: Care, Health and Development</i> ; 24: 129-143
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Book Chapter	Ross EM, Lynn R Ed B Valman. Achievements of the BPA - the British Paediatric Surveillance Unit, <i>The RCPCH at the Millennium</i> : 63-67
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	Rahi J, Dezateux C. Capture-Recapture Analysis of Ascertainment by Active Surveillance in the British Congenital Cataract Study; <i>QVS</i> . 40 (1): 236-239
	Rahi J S, Tookey PA. Congenital and Infantile Cataract in the United Kingdom: Underlying or Associated Factors. <i>IOVS</i> ;41: 2108-2114
	Rahi JS, Dezaeux C and the BCCIG. Measuring and interpreting the incidence of congenital ocular anomalies :lessons from a national study of congenital cataract in the UK. <i>Invest Ophthalmol Vis Sci</i> ;42: 1444-1448
	Rahi JS, Botting B and the BCCIG. Ascertainment of children with congenital cataracts through the National Congenital Anomaly System in England and Wales, <i>Brit J Ophthalmol</i> . 85; (9): 1049-1051
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	Dezateux C, Godward S. Validation of the reporting bases of the orthopaedic and paediatric surveillance schemes. <i>Arch. Dis. Child</i> . 75: 232-236
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Letter	Bedford H, Booy R, Dunn D, DiGuseppi C, Gibb D, Gilbert R, Logan S, Peckham C, Roberts I, Tookey P. Autism, inflammatory bowel disease, and MMR vaccine (Letter). <i>Lancet</i> ; 351: 907
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Congenital Toxoplasmosis	Hall SM. Congenital Toxoplasmosis (Regular Review). <i>Brit. Med. J</i> ;306: 291-297
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Creutzfeldt-Jakob disease/Alpers Syndrome	Water Naude J, Verity CM, Will RG, Devereux G, Stellitano L. Is variant Creutzfeldt-Jakob disease in young children misdiagnosed as Alpers' syndrome? An analysis of a national surveillance study. <i>JNNP</i> ; 75: (6):910-3.
Diabetes (in under fifteens)	Lessing DN, Swift PGF, Metcalfe MA. Baum JD. Newly diagnosed diabetes: a study of parental satisfaction. <i>Arch. Dis. Child</i> ; 67: 1011-1013
	Metcalfe MA, Baum JD. Incidence of Insulin Dependent Diabetes in Children Aged Under 15 Years in the British Isles During 1988. <i>Brit. Med. J</i> ; 302: 443-7
Diabetes (Neonatal)	Shield JPH, Wadsworth E, Baum JD. The genetic contribution to disease pathogenesis in childhood diabetes is greatest in the very young. <i>Diabetic Medicine</i> ;12(5): 377-9
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	Wadsworth EJ, Shield JP, Hunt LP, Baum JD. A case-control study of environmental factors associated with diabetes in the under 5s. <i>Diabetic Medicine</i> ; 14(5):390-6
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Fatal adverse drug reactions	Cheng K, Masters S, Stephenson T, Cooke R, Ferner R, Ashworth M, Nunn T. Identification of suspected fatal adverse drug reactions by paediatricians: a UK surveillance study, <i>Arch Dis Child</i> , doi:10.1136/adc.2006.107789,
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	Adak B, Lynn R, O'Brien S. HUS Surveillance What does it tell us about VTEC? <i>SCIEH Weekly Report</i> ; 34:14
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Haemophilus influenzae type b (Hib)	Booy R, Moxon ER. Haemophilus influenzae type b. <i>Arch. Dis. Child</i> ; 68: No. 4, 440-441
	Booy R, Heath PT, Slack MPE, Begg, N, Moxon ER. Vaccine failures after primary immunisation with Haemophilus influenza type-b conjugate vaccine without booster. <i>Lancet</i> ; 349: 1197-202
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8 Appendix 3

8.1 Suppliers

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