

## ***British Paediatric Association***

# **The British Paediatric Surveillance Unit**

The British Paediatric Surveillance Unit (BPSU) was set up to provide a method of nationwide disease surveillance particularly orientated to the study of rare childhood disorders, including infectious or infection related conditions of uncertain aetiology or epidemiology. It is a joint venture of the British and Irish Paediatric Associations, the Communicable Disease Surveillance Centre (CDSC) of the Public Health Laboratory Service (PHLS), and the Department of Epidemiology of the Institute of Child Health, London. It was first set up in July 1985 and became fully operational in July 1986. This report summarises the history and operation of the BPSU and presents some results from the activities of its first year.

### **History**

The BPSU developed from the collaboration between the BPA and CDSC to improve the surveillance of infection and associated conditions in children that could not be monitored through existing data collection systems, such as laboratory reporting. This began in 1981 with Reye's Syndrome and was later extended to include haemolytic uraemic syndrome, Kawasaki disease, and haemorrhagic shock encephalopathy syndrome.

Although the voluntary clinical reporting system for these conditions met with enthusiastic support, it was felt that a more active method of case gathering would achieve the higher levels of ascertainment necessary for reliable documentation of trends in rare disorders. Furthermore as the BPA and its members were receiving an increasing number of requests from individual research workers to report a range of rare conditions there was a need to improve the efficiency of research without losing the goodwill of clinicians being asked to report patients under their care.

### **Objectives**

The BPSU has two principal objectives: firstly, to involve paediatricians in the reporting and surveillance of uncommon childhood conditions of importance for public health (particularly those associated with infection) and those rare conditions for which

ascertainment on a national scale is necessary for research. This would include studies, for example, of incidence and prevalence that would generate aetiological hypotheses and provide data for monitoring preventive programmes. Secondly, to provide a unified reporting scheme that is simple, flexible, minimises paperwork, and yet is capable of rapid response to an 'epidemiological emergency'—for example, the detection and monitoring of 'new' diseases of environmental origin, or the surveillance of a rare adverse vaccine reaction.

### **Organisation and funding**

The unit is located in the BPA Office and is staffed by a full time administrator and a part time medical coordinator and clerical officer. It is managed by a Steering and a Scientific Advisory Committee, the three parent bodies being represented on both of these. The BPSU depends almost entirely on charitable and commercial funding for its existence and it now seeks a financial contribution, where possible, from the investigators whose studies it helps.

### **Methodology**

#### **(1) THE MAILING OPERATION**

A report card listing the current 'menu' of reportable conditions is sent monthly to all consultant BPA and Irish Paediatric Association members. After having indicated cases of any of these disorders seen in the preceding calendar month or, if none have been seen, having checked a 'nil return' box, the cards are returned to the BPSU. 'Positive' notifications are forwarded to the appropriate research workers who then contact the reporting paediatricians for further information. The mailing operation is sufficiently flexible to allow for surveys to be conducted in one or more regions if this is appropriate to a particular investigator's study design.

#### **(2) INCLUSION OF STUDIES**

The Unit's Scientific Advisory Committee meets monthly to consider proposals from investigators. An introductory booklet containing guidelines for

these submissions has been produced. Investigators are encouraged to discuss their plans at an early stage with the medical coordinator and help with study design and constructive criticism of methodology has been an important BPSU activity. In its review procedure the committee considers certain points including: the importance of the research question; the estimated incidence of the condition; the acceptability of the response (usually a questionnaire) to a 'positive' return; the proposed duration of the study and demonstration of adequate resources for its completion. Once accepted a condition can, if necessary, be added to the report card for the next month. A summary protocol for the study that includes case definition, reporting instructions, and the name and address of the investigator(s) is also sent at this stage to all respondents, who have been provided with a special folder to facilitate the storage of protocol cards as studies start and finish. Several paediatricians have commented on the educational usefulness of these protocols, which could heighten diagnostic awareness of rare disorders.

#### (3) MONITORING THE BPSU

The effectiveness of the Unit can be measured in several ways. First is the subjective or objective opinion of the investigators. The latter might include a comparison with a previous method of case ascertainment or with a concurrent alternative system—for example, death certificates, computerised hospital inpatient data. Second is the monitoring of the proportion of 'positive' reports that are subsequently confirmed (investigators are asked to make returns to the BPSU indicating 'true' cases, duplicate reports, and reporting errors). Third is the response of members in returning cards. Members who consistently fail to return their cards are sent a series of reminder letters seeking reasons for non-response and requesting future cooperation. Finally, the perceived value of the Unit as a research tool could be expected to be reflected in continuing applications for new studies to be included.

#### (4) BPSU REPORTS

The BPSU has produced two annual reports (available from its office) and also regular quarterly reports in the Communicable Disease Report of the PHLS. This paper is the first of a series to be produced six-monthly in Archives and to which investigators will be asked to contribute observations from their studies. In addition tabulations of regional response rates have been included in mailings periodically in order to introduce a 'competitive' element to participation.

### BPSU Activities July 1986 – June 1987

#### (1) PARTICIPATION IN THE SCHEME

The response rate, among about 800 paediatricians who are sent the month card, rose from 73% in the first month to 89% after one year (this is calculated as the percentage returned within 90 days after each mailing). This rate has remained stable over the last months of the year. There were noticeable regional differences that ranged from 100% in some months (North Scotland, East Anglia, Trent) to a minimum in one month of 68% (West Scotland, December 1986).

Because lack of response may have reflected shortcomings of the scheme (either administrative or due to members' dissatisfaction), persistent non-participants (non-return for three consecutive months) have been contacted to ascertain their reasons. Numbers in three such cohorts declined from 69 to eight and only a handful of these expressed criticism of the BPSU as their reason for non-compliance. It is unlikely that 100% response rate will be consistently achieved given the vagaries of change of address, leave, and retirement. Continuing close attention to non-respondents should, however, minimise the non-response rate and ensure that the mailing list is kept up to date.

#### (2) CONDITIONS REPORTED

The table indicates the conditions that have been included on the cards during the year, numbers reported, and the numbers that were 'valid'. Those 'not yet known' reflect cases still under investigation by the research workers.

All current investigators have expressed satisfaction with the BPSU, especially those who previously ascertained such cases with a different system. The effect on reporting of Reye's syndrome, Kawasaki disease, and haemolytic uraemic syndrome is shown in the figure—a particularly dramatic increase was seen in Kawasaki disease reports. The initial increase in Reye's syndrome was discouraging as this coincided with the withdrawal of paediatric aspirin preparations. Further analysis showed, however, that many of these initial reports were 'old' cases and duplicates and the continued decline of Reye's syndrome, despite improved ascertainment, has been an important measure of this intervention. The trends shown for haemolytic uraemic syndrome reflect the seasonal distribution of this condition with its late summer–autumn peak.

#### (3) PROPOSALS FOR NEW STUDIES

The Unit received 11 preliminary proposals for new surveys over the year. Six of these were subsequently submitted formally to the Scientific

Table *Conditions included on the reporting cards and number of notifications*

Condition	Total reports July 1986– June 1987	No (%) valid		
		Yes*	No†	Not known‡
AIDS in childhood¶	37	18 (49)	13 (35)	6 (16)
Neonatal herpes¶	21	8 (38)	9 (43)	4 (19)
Reyes syndrome	49	20 (41)	19 (39)	10 (20)
Kawasaki disease	120	96 (80)	10 (8)	14 (12)
Haemolytic uraemic syndrome	51	35 (69)	1 (2)	15 (29)
Haemorrhagic shock encephalopathy syndrome	17	12 (71)	2 (12)	3 (18)
Subacute sclerosing panencephalitis¶	33	25 (76)	4 (12)	4 (12)
X-linked anhydrotic ectodermal dysplasia§¶	13	2 (15)	0	11 (85)
Insulin-dependent diabetes§	20	20 (100)	0	0
Lowie syndrome	15	5 (33)	1 (7)	9 (60)
Total	376	241 (64)	59 (15.7)	76 (20)

\* Meets case criteria.

† Reporting error, duplicate, revised diagnosis.

‡ Still being followed up (30 September 1987).

§ Three month surveys.

|| South Western Region only.

¶ Figures include cases ever seen (AIDS) or seen past year (others) as well as new cases in past month.

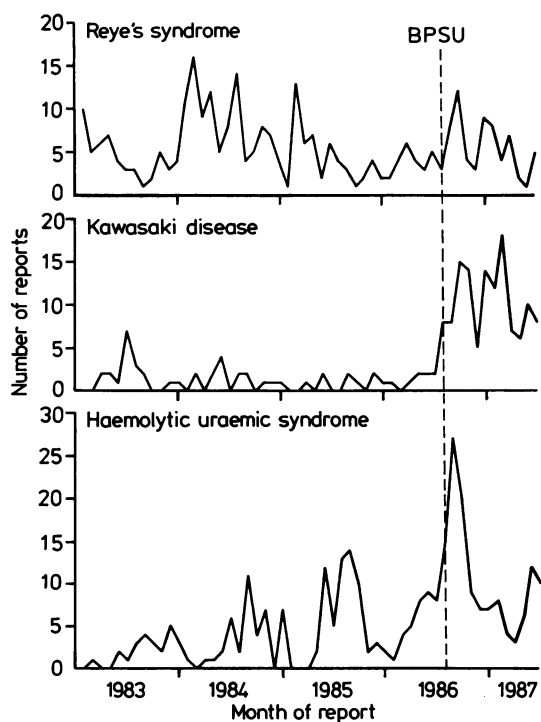


Figure *The effect of the BPSU system on reporting.*

were accepted and will be added to the 'menu' in due course, others are still under consideration.

#### Comment

The BPSU is a unique approach to the surveillance of the less common communicable and infection related diseases in children and to the study of rare paediatric disorders. Its first year has gone remarkably well for such a new venture, judging from response rates, satisfaction of investigators, and the many important new research proposals. Its current and future success depends very largely on the continued support of British paediatricians who give their time to helping national studies that, by their nature, are usually unable to acknowledge individual contributions. It will therefore be important to retain this goodwill by limiting the number of reportable conditions on the card at any one time; by ensuring that the studies included are important, interesting, and of limited duration so that the conditions change periodically, and by providing feedback. This should in turn increase both professional and public knowledge of rare paediatric disorders that, taken together, account for a considerable proportion of childhood morbidity and mortality in the British Isles.

Advisory Committee. Conditions to be studied included a range of rare metabolic, haematological, neoplastic, and infectious disorders and one other proposal was for a survey of near drowning. Some

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