

The BPSU

The British Paediatric Surveillance Unit: the first 20 years

Richard Lynn, Euan Ross

The BPSU has made a major contribution to paediatric medical epidemiology across the world

In July 2006 the British Paediatric Surveillance Unit (BPSU) completed its twentieth year of surveillance into uncommon childhood disorders. During this period the BPSU has provided a mechanism to respond to acute public health events in children and has emerged from its small base to become a major contributor to paediatric medical epidemiology across the world. All this has been achieved by an efficient system that permits the simultaneous running of multiple surveys via the circulation of a monthly report card. These achievements are the result of a willingness by UK and Irish paediatricians to work in collaboration for the betterment of child health across the whole of the British Isles.

This short article reviews the development of the unit and how studies undertaken have impacted on public health.

HISTORY

In the 1970s the then British Paediatric Association (BPA) was growing rapidly through consultant expansion and realising that its mission was expanding from a membership organisation into a wider role in the promotion of child health. Simultaneously, the Communicable Disease Surveillance Centre (CDSC) of the Public Health Laboratory Service (PHLS) in London headed by Dr Spence Galbraith needed to develop a system to speedily recognise and monitor newly emerging diseases, especially those which were infection associated but for which no specific diagnostic laboratory investigation was available. These conditions would not be readily identified by the PHLS (now the Health Protection Agency) laboratory reporting system.

Paediatricians had already been active in support of the collection of important data relevant to public health. In the early 1970s they had been invited to return information about cases of lead intoxication by Professor David Barltrop and colleagues at St Mary's Hospital. In 1976–79 paediatricians in England, Scotland and Wales participated in the National Childhood Encephalopathy

Study (NCES) co-directed by Professor David Miller and Dr Euan Ross at the Middlesex Hospital Medical School. The aim was to identify those children between 2 and 35 months of age with serious acute neurological illness and independently review their immunisation histories at the time when the safety of pertussis immunisation was under intense scrutiny.

The NCES used the strategy of ascertaining cases of acute infant neurological illness via a card posted every month to all consultant paediatricians, which not only requested reports of cases but importantly also asked for a nil return if no cases had been seen that month. This is known as "active" case ascertainment. This need for nil return is fundamental to the success of the BPSU. In the course of 3 years, consultant paediatricians reported 1182 cases of infant encephalopathic disease.¹

Dr Martin Bellman, a member of the NCES, studied the reported cases of Reye's syndrome (RS) at the time when international interest in RS was increasing following publication of the first United States studies associating it with aspirin. The US Centers for Disease Control surveillance scheme was showing that it was a significant public health problem in association with winter flu epidemics. Dr Bellman approached Dr Galbraith to see if the PHLS could collaborate with British paediatricians via a national surveillance scheme. Dr Susan Hall working at the PHLS was assigned to develop this project. Resources to undertake a monthly active card reporting system were not available. However, following discussions with Dr David Harvey (then Honorary Secretary) at the BPA, in August 1981 a letter was sent to all consultant paediatricians requesting reports of RS.

The early 1980s saw the extension of this "passive" method of reporting to include haemolytic uraemic syndrome (HUS), Kawasaki disease and haemorrhagic shock encephalopathy syndrome. However Dr Hall and others felt that under-ascertainment could be occurring,

so active surveillance was again considered.

Discussions were held in 1984 between what were to be the parent bodies of the BPSU: the BPA, PHLS and the Institute of Child Health (London). A small steering committee was set up under the chairmanship of Sir Cyril Clarke, and including Sir Peter Tizard, Euan Ross, Catherine Peckham and David Baum, as well as Dr Susan Hall who became the first medical coordinator. The steering committee, which later became the BPSU Executive Committee (BEC), decided to implement active surveillance using the NCES methodology. It was felt that the introduction of such a system would optimise the completeness of case ascertainment necessary for assessing trends in rare disorders. It was also hoped that such a system would reduce the number of requests for case reports received by clinicians from individual researchers.

The objectives of the BPSU are to facilitate research into uncommon childhood disorders for the advancement of knowledge, to allow paediatricians to participate in surveillance, to increase their awareness of rare childhood disorders and to respond rapidly to public health emergencies.² From the outset it was agreed that simplicity was essential and that as little burden as possible should be placed on reporting paediatricians and every effort made to maintain their interest and collaboration. The BEC of today still adheres to these principles.

In July 1986 the first BPSU orange card was dispatched to 800 consultant paediatricians in the UK and Ireland. It listed six disorders: AIDS, Lowe syndrome, HUS, neonatal herpes, subacute sclerosing panencephalitis (SSPE) and X-linked anhydrotic ectodermal dysplasia.

ACHIEVEMENTS

By any measure the BPSU has been a success and has far exceeded the expectations of its founders. By 2006 the Unit had surveyed over 60 rare conditions in its first 20 years. Willingness to participate has been demonstrated with 4000 clinicians having received over 350 000 cards in total, with an average card return of 90%. More than 20 000 confirmed case reports have been made with over 85% of questionnaires returned. The data have contributed to more than 150 peer review papers and a similar number of national and international presentations, thus raising the awareness of these conditions with both clinicians and the public. The methodology has been so successful that several other specialty groups such as ophthalmology, neurology and obstetrics have duplicated it in the UK. Internationally there are now 12 other

paediatric surveillance units,³ which in 1998 collaborated to form the International Network of Paediatric Surveillance Units (INoPSU). Collectively these units have facilitated the surveillance of over 150 rare paediatric conditions. Covering a child population of over 50 million with 10 000 clinicians involved, INoPSU has the potential to be a powerful tool for future research into such conditions.

Perhaps the greatest measure of success is the impact results have had on public health in relation to clinical governance, initiating and monitoring public health interventions and rapidly responding to public health emergencies and informing policy decisions.

As early as 1986 the BPSU was monitoring the effects of new warnings about aspirin and RS. Some 16 years later and even after dramatic falls in RS reports, BPSU data contributed to a Committee of Safety of Medicines decision to extend the aspirin warning to include all children under 16 years of age. The biliary atresia survey (1993–1995) demonstrated important differences in outcome according to the size of the unit undertaking surgery and the number of cases operated on, with those centres that handled the most cases having the best outcomes. This information led to an NHS Executive Directive restricting the number of centres where surgery can be undertaken.⁴

The BPSU has maintained surveillance of diseases targeted by vaccination programmes as well as those with late sequelae. Surveillance of congenital rubella and acute flaccid paralysis (polio vaccine) and *Haemophilus influenzae* b vaccine (Hib) failures has also reinforced the success of national immunisation programmes.

The BPSU has also provided a mechanism for responding to and investigating emerging public health concerns. At their onset emerging diseases tend to be rare and may remain unrecognised, potentially allowing the condition to spread. The HUS survey, undertaken in the 1980s through the BPSU, was one of the first studies to confirm the link between *Escherichia coli* 0157 and paediatric HUS in the UK. The study was replicated in the late 1990s in response to the Pennington Report,⁵ which highlighted the effectiveness of the BPSU methodology in identifying *E coli* 0157 outbreaks. The BPSU reacted to concerns in the mid 1990s over the increase in BSE in the national herd of cattle and the potential implications for child health; since 1997 the BPSU has contributed to monitoring of variant Creutzfeldt-Jacob disease in children

through the Progressive and Intellectual Neurological Disorder survey.

Among non-infectious emergencies, the Unit assessed the impact of changing the route of administration of vitamin K in newborn infants following concern about a possible link between vitamin K injections and the subsequent development of childhood cancers, as well as looking at outcomes following labour/birth in water.

The BPSU has regularly supported the work of the National Screening Committee (NSC). Data derived from several studies have contributed to decisions on whether screening for various conditions should commence or not, or whether they should be discontinued. Surveillance of paediatric HIV and AIDS revealed that in many cases the infected infant was the first member of the family to be diagnosed. Findings from the paediatric and associated obstetric surveillance system, together with the evidence that it was possible to reduce the risk of transmission considerably if women were diagnosed early in pregnancy, led to the introduction of routine antenatal screening for pregnant women, a substantial improvement in diagnosis rates in pregnancy and a reduction in the proportion of infants with vertically acquired infection. By contrast, vertical transmission of *Toxoplasma gondii*, group B streptococcal infection and herpes simplex virus was too low to justify initiation of screening in pregnancy. Surveillance of congenital syphilis and syphilis in pregnancy, however, identified a continuing burden of infectious syphilis in mothers, leading to a decision of the NCS to continue antenatal syphilis screening.

The BPSU system does allow for the occasional "off the wall" idea. Readers may recall a BPSU study into toy chemistry set poisoning undertaken by Dr Ted Mucklow from the Isle of Wight, in collaboration with the National Poisons Unit. This study quantified the role of faulty packaging of potentially deadly chemicals in poisoning in children. Data from this study contributed to a change in EU legislation on the packaging of such "toys".

CONCLUSIONS

In setting up the BPSU it was hoped that paediatricians could make a contribution to the understanding of rare paediatric disease epidemiology. Few could have expected the impact the BPSU has had on public health, not only nationally but also internationally through the establishment of similar such units across the globe. These achievements have been recognised in recent years through grants

from the Department of Health, which have secured the medium-term future. The BPSU continues to look to the future and dissemination of information continues to improve through the expansion of the website (<http://bpsu.inopsu.com>). The Unit is also supporting trainees with the award of a yearly bursary in honour of Sir Peter Tizard.

It is important to note that the impact of the BPSU is due to the enthusiastic efforts of paediatricians and others who have supported the surveillance projects through the return of the monthly orange card and the completion of questionnaires. As the late Sir Cyril Clarke said in 1991 "British and Irish paediatricians can feel justly proud of themselves as the pioneers and key enactors of this unique reporting system".

ACKNOWLEDGEMENTS

The BPSU has from the start realised that it could only succeed with the active cooperation of all consultant members. Their continuing wholehearted cooperation made the BPSU possible. The committee has asked us to convey their thanks to the many funders of the BPSU which include an anonymous charity obtained via the good offices of the late Sir Cyril Clarke, WellChild via the late Sir Eric Stroud, and now the Department of Health.

Arch Dis Child 2007;**92**:744–745.
doi: 10.1136/adc.2006.105908

Authors' affiliations

Richard Lynn, British Paediatric Surveillance Unit of the Royal College of Paediatrics and Child Health, London, UK

Evan Ross, Child Studies Unit, King's College, Strand, London, UK

Correspondence to: Richard Lynn, British Paediatric Surveillance Unit of the Royal College of Paediatrics and Child Health, 50 Hallam Street, London W1W 6DE, UK; richard.lynn@rcpch.ac.uk

Accepted 23 April 2007

Competing interests: None.

REFERENCES

- 1 Miller DL, Ross EM, Alderslade R, *et al*. Pertussis immunisation and serious acute neurological illness in children. *BMJ (Clin Res Ed)* 1981;**282**(6276):1595–9.
- 2 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. *Child Care Health Dev* 1998;**24**:129–43.
- 3 Elliott EJ, Nicoll A, Lynn R, *et al*. Rare disease surveillance: an international perspective. *Paediatr Child Health* 2001;**6**:251–60.
- 4 McKiernan PJ, Baker AJ, Kelly DA. The frequency and outcome of biliary atresia in the UK and Ireland. *Lancet* 2000;**355**(9197):25–9.
- 5 Advisory Committee on the Microbiological Safety of Food. Report on Vero cytotoxin-producing *Escherichia coli*. London: HMSO, 1985.