

Document amended and agreed on by participants at the first meeting of the International Network of Paediatric Surveillance Units, Ottawa, Canada, June 14th 2000, based on an earlier document.

The International Network of Paediatric Surveillance Units (INoPSU)

This document outlines the principals behind the development of an International Network of Paediatric Surveillance Units (INoPSU).

1. Background and Rationale

Rare diseases and infections are, paradoxically, a numerically important cause of morbidity and mortality in childhood in industrialised countries. They are also increasingly recognised as a cause of concern in resource poor countries. Individually uncommon, together they number thousands, and many result in severe sequelae. Many are characterised by chronicity and by high rates of disabling sequelae or death. Most pose a large financial and emotional burden for affected children, their families and health systems.

The difficulty in recognising rare disease can result in delayed diagnosis, increasing the risk of preventable complications or death. Emerging infections will initially be rare and may remain undetected and infection spread extensively before action is taken. All such diseases are difficult to study as their low frequency often means that little may be known about their aetiology, clinical spectrum, sequelae, pathological features, diagnosis, treatment and management. In order to generate a sufficient number of cases to derive meaningful data, the study of rare diseases requires collaboration of many clinicians providing health care to large populations over extensive areas.

To address this problem in the UK and Eire, the British Paediatric surveillance Unit (BPSU) was initiated in 1986. In 1992, Australia, Netherlands and Germany started similar Units. In 1994 Switzerland and Malaysia followed and more recently Units have been established in Canada (1996), Wales (1996), Papua New Guinea (1996), New Zealand (1997) and Latvia (1997).

The eleven Units (Table) provide an efficient, effective framework for case-finding for investigators who wish to study rare conditions in children. These include infections, infection-related conditions, vaccine-preventable diseases, congenital and inherited (genetic) diseases, unusual injuries or therapies and rare complications of common diseases. The Units frequently encourage, facilitate or elicit studies but only occasionally undertake research themselves.

Applications to conduct a study are usually considered by panels of experts with scientific, epidemiological and public health expertise, as well as paediatricians participating in the system. Some proposals are judged unfeasible or insufficiently resourced. Those conditions for which study protocols are accepted are placed on a report card which is sent monthly to all participating clinicians.

Thus, the Units have active surveillance schemes allowing simultaneous, prospective monthly collection of national data on multiple conditions. Surveillance is usually undertaken for between 1-3 years depending on the protocol, although some conditions of particular public health importance remain under long-term surveillance. Investigators may pay a fee covering a proportion of central costs to conduct a study through the surveillance unit (Table). Units are usually related to the national body representing paediatricians as this has the most accurate database of paediatricians eligible to report. This arrangement also tends to maximise reporting as paediatricians then 'own' the system. Other bodies concerned with child and public health are frequently represented on Units' committees.

TABLE - Data from National Paediatric Surveillance Units - End 1997

Country paediatric dept.)	Year Established	Mailing List (paediatricians or			
		Reply Paid Study	Response Rate	Fee paid to unit for	
Australia	1992	921	Yes*	93%	No
Britain/Eire	1986	1729	No	94%	Yes
Canada	1996	2100	Yes	82%	No
Germany	1992	496 (approx)	No	94%	Yes
Latvia	1997	n/a	n/a	n/a	n/a
Malaysia	1994	340	Yes	60%	Yes
Netherlands	1992	416	Yes	92%	Yes
New Guinea	1996	40	Yes	60%	No
New Zealand	1997	163	Yes	96%	No
Switzerland	1995	41 (approx)	No	98%	No

*(some reporting by e-mail)

The principle of 'active' surveillance is that the initiative for notification of a case comes from the Unit, not from the reporting clinicians (respondents). The precise structure of the mailing list for each surveillance unit varies to suit local circumstances. A report card listing conditions being studied is sent every month from the surveillance Unit either to all consultant paediatricians (working both in hospitals and privately) or to all heads of paediatric departments. Other specialists working in child health (e.g. paediatric surgeons, dermatologists or orthopaedic surgeons) may also be circulated. Respondents are asked to indicate on the card the number of cases of the listed disorders seen in the previous month and to return the card to the surveillance unit. It is important that the card is returned even if no cases have been seen, in order to monitor compliance with the scheme, and to maximise case ascertainment.

Mailing lists vary considerably in size but response rates in many of the existing Units approach or exceed 90% per month (Table). In some countries monthly cards are reply-paid, for some conditions in some countries telephone reporting is used and in Australia a proportion of paediatricians- report by e-mail. When a case is reported, the Unit notifies the investigator responsible for that study, who then contacts the reporting clinician to request further information. Many studies use other data sources for validation. Some Units require that researchers inform them of the outcome of a report of a case so as to calculate the proportions of preliminary reports which status (confirmed case, duplicate, not meeting the case definition etc.) and data have been received (completion rates).

The existing European Paediatric Surveillance Units have been meeting regularly since 1992 to discuss research protocols. The Director of the Australian unit has met with those of Units in New Guinea, New Zealand and Malaysia. In 1996 the proposal to form an International Network of Paediatric Surveillance Units within and outside Europe was accepted in principle by all Units existing at that time. Units regularly contact each other for results, sharing of protocols, putting

researchers in touch with each other and a common international report is shared as part of national reports. Given the existence of at least ten national paediatric surveillance Units undertaking similar work and the level of informal contact, the Units agreed in 1998 that the time had come to formalise these links into a network.

1. Agreement

1. *To form an International Network of Paediatric Surveillance Units (INoPSU) to link the pre-existing Units.*
2. *That the BPSU should, in the first instance, act as the 'server' of such a network (1998-2001).*

2. Mission

1. The mission of the International Network of Paediatric Surveillance Units (INoPSU) is the advancement of knowledge of uncommon childhood infections and disorders through the participation of paediatricians in surveillance on a national and international basis so as to achieve the aims and benefits listed below.

3. Aims and Benefits

1. to facilitate communication and co-operation between existing national paediatric surveillance Units, researchers and investigators and assist in the development of new and existing Units.
2. to facilitate sharing of information and collaboration between researchers from different nations and scientific disciplines.
3. to share information on current, past projects and anticipated and protocols and on conditions that have been nominated for surveillance but were not selected
4. to encourage the use of identical protocols to potentially enable simultaneous or sequential collection of incidence data on rare paediatric disorders in two or more nations. Thus through development and use of common case definitions and research protocols to potentially provide large and diverse study populations from which to obtain data so as to enable comparison of incidence, aetiology, management, efficacy of preventative strategies, outcomes and changing patterns of rare diseases over time.
5. to pool results, analyses and conclusions, and facilitate their dissemination to national and international health authorities so as to raise awareness of rare conditions to encourage early diagnosis and appropriate treatment and management and detect the emergence of new conditions.
6. to potentially establish international cohorts from identified cases which could, support future research.
7. to share and distribute information of educational benefit to constituent Units notably on study and surveillance methodologies, statistical techniques and models of evaluation for Units.
8. to peer-review and evaluate existing and proposed Units and to discuss areas of concern relating to surveillance data validation, ethics and confidentiality.

9. to identify rare disorders of mutual interest and public health importance for co-operative surveillance through each national Unit.
10. to facilitate development and clarification of internationally recognised diagnostic criteria for rare diseases, which will help standardise their management.
11. to collaborate with and provide information to other groups interested in rare childhood disease, such as parent support groups.
12. to particularly estimate of the incidence of selected serious vaccine-related reactions and of vaccine failures and evaluation of new vaccine policies.
13. to respond promptly to international emergencies relating to rare childhood conditions where national and international studies can make a contribution to science or public health.
14. to vigorously encourage the promulgation of the benefits of surveillance to the whole community including the general public, patient groups, health care staff and decision makers.
15. to promote guidance to national units and others as to how surveillance can be carried out without prejudicing data protection, patient confidentiality and ethical standards.

4. Principle and Membership

1. National Units participating in INoPSU will contribute to and draw on other Units for advice and information while remaining independent and responsive to individual national requirements for monitoring specific disorders.
2. The Network will consist of national paediatric surveillance Units. There will be full and associate members.

Full members will consist of:

- * the founding Units (see note 7.)
- * those meeting the following pre-set criteria of being “A national unit undertaking active, clinician-based paediatric surveillance, possessing a scientific committee and producing a regular annual report, preferably annually”.

Associate members will consist of:

- * National Paediatric Associations/Societies preparing to set up surveillance and Units that have yet to meet agreed criteria.
- * other medical speciality Units using similar surveillance methodology
- * other international organisations working in the area of rare disease e.g. the European organisation for Rare Disease, North American Organisation for Rare Disease, EUROCAT, organisations for clinical geneticists etc.
- * other concerned bodies, notably the International Paediatric Association.

1. Mechanism

1. All existing national Paediatric Surveillance Units will be members of INoPSU which will primarily function electronically with occasional face to face meetings.

2. Each Unit will nominate an international link person.
3. One Unit will be appointed as a 'server' for the network. initially for a fixed term of three years. It will host the Network's web-site which will link to other Unit's web-sites and the Web-sites of national paediatric bodies.
4. The Web-site will include:
 - * contact details of the Units,
 - * summary protocols (titles, research questions, case definitions and investigator contact details only) of current, and past and anticipated surveys
 - * a bulletin board for shared discussions.
1. There will be a small (3-5 person) group (the Network Secretariat) selected from the Units but including persons from different regions which will be responsible for ensuring that INoPSU is active, meets its aims, and that its mechanism works.

7. Member Units

Australian Paediatric Surveillance Unit (APSU)

British Paediatric Surveillance Unit (BPSU)

Canadian Paediatric Surveillance Program (CPSP)

German Paediatric Surveillance Unit (ESPED)

Latvian Paediatric Surveillance Unit (LPSU)

Malaysian Paediatric Surveillance Unit (MPSU)

Netherlands Paediatric Surveillance Unit (NSCK)

New Zealand Paediatric Surveillance Programme (NZPSU)

Papua-New Guinea Paediatric Surveillance Unit (PNGSU)

Swiss Paediatric Surveillance Unit (SPSU)

Welsh Paediatric Surveillance Unit (WPSU) – joined June 2000

8. Associate Members

British Ophthalmic Surveillance Unit (BOSU) – joined June 2000