Neonatal follow-up programs and follow-up studies: Historical and current perspectives

Reg Sauve MD MPH FRCPC[1], Shoo K Lee MBBS PhD FRCPC[2]

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The present report reviews some highlights in the history of neonatal intensive care and neonatal follow-up programs, particularly developments and reports that were based on experiences in Canada. Early outcomes reported from 'preemie baby units' were distressing, but attention has consistently been paid to preterm infant outcomes, even from the early days of neonatal intensive care units. Most current follow-up programs have goals related to 'audit' functions, education and clinical roles, but existing literature related to these functions is limited. Several reports have provided guidance in terms of neonatal follow-up research issues, and these strengthen the place of follow-up studies in outcomes research.

Key Words: *Follow-up; Neonatal; NICU; Outcomes; Programs*

Les programmes de suivi néonatal et les études de suivi : Des perspectives historiques et courantes

Le présent rapport contient une analyse de quelques faits saillants de l'histoire des soins intensifs néonatals et des programmes de suivi néonatal, notamment l'évolution et les comptes rendus fondés sur les expériences au Canada. Les premières issues déclarées dans les unités de prématurés étaient inquiétantes, mais on s'est toujours attardé au sort des prématurés, même aux premiers temps des unités de soins intensifs néonatals. La plupart des programmes de suivi actuels ont des objectifs reliés aux fonctions de vérification, à la formation et aux rôles cliniques, mais les publications à cet égard sont limitées. Plusieurs rapports ont fourni une orientation quant aux enjeux de la recherche sur le suivi néonatal et soulignent l'importance des études de suivi dans les recherches sur les issues.

The following discussion briefly explores the historical background of neonatology and neonatal follow-up, the goals of neonatal follow-up programs and the current place of neonatal follow-up studies as important examples of outcomes research.

HISTORICAL BACKGROUND OF NEONATAL INTENSIVE CARE AND FOLLOW-UP

During the past century, the care of preterm infants evolved from the Tarnier's agricultural incubators adapted for use in human infants to the well-described incubator baby displays at World's Fairs and Coney Island, New York (1) to the survival of occasional preterm babies from the display incubators to the first 'preemie baby units' in the 1920s and 1930s (2). With such ignominious beginnings, it was of little surprise that the early days of intensive care for preterm babies were met with extreme skepticism. In fact, there were catastrophic disasters described in early reports of the outcomes of preterm infants treated using ventilators; there was an 'epidemic of blindness' resulting from unmonitored use of supplemental oxygen; and brain damage was common because of inadequate attention to jaundice, nutrition and infections (3). Also, prolonged isolation of babies and separation from their parents led to many long-term interaction and behavioural problems.

But even during those early years, while neonatology was developing as a specialty and neonatal intensive care practices were being refined, there was attention to the long-term outcomes of the small 'preemies'. An intriguing and important early outcome report by Hess (2) on babies born in the 1930s was followed by carefully conducted and reported follow-up studies by Lubchenco et al (4) and Drillien (5) describing infants cared for in the 1940s and 1950s. Their reports of surviving preterm babies delivered the tragic news that up to two-thirds were disabled. These early follow-up studies laid some of the groundwork for neonatal intensive care and neonatal follow-up as they exist today.

Several Canadian investigators contributed important findings in outcome studies of groups of very low birth weight infants or term infants who experienced neonatal complications between 1960 and 1990. An early example was a report on the fate of the expremature by Grewar et al (6) from Winnipeg, Manitoba. Buck et al (7), in a classic report, described the 12-month outcomes of children of mature birth weight from the Ontario Perinatal Mortality Study, stressing important methodological issues, such as the inclusion of comparison or control subjects in neonatal outcome studies. Dunn et al (8) produced several reports from Vancouver on the sequelae of low birth weight.

[1]*Departments of Pediatrics and Community Health Sciences, University of Calgary, Calgary;* [2]*Department of Pediatrics, University of Alberta, Edmonton, Alberta*

Correspondence: Dr Reg Sauve, Departments of Pediatrics and Community Health Sciences, University of Calgary, 3330 Hospital Drive Northwest, Calgary, Alberta T2N 4N1. Telephone 403-220-4294, e-mail rsauve@ucalgary.ca

Fitzhardinge et al (9,10) published a series of important neonatal outcome papers on cohorts of very low birth weight, mechanically ventilated or small for gestational age infants. Later, Low et al (11) provided important information on the quantification and outcomes of children born following intrapartum asphyxia, and Robertson and Finer (12) described important long-term effects of hypoxic-ischemic encephalopathy.

Additional reports from Canada over the past two decades have continued to provide methodological insights into the design of neonatal outcome studies, including the use of geographically based populations (13,14) and the introduction of more appropriate testing approaches (15). Currently, neonatal follow-up programs in Canada are moving toward the development of a national neonatal follow-up database in conjunction with the Canadian Neonatal Network (16). Increasingly, multicentre, randomized controlled clinical trials have incorporated the measurement of long-term outcomes of infants, relying on the structure and procedures that already exist in Canadian neonatal follow-up programs (17-19). Also, reports on older children, adolescents and adults have added an important new dimension to Canadian follow-up studies (20).

GOALS OF CURRENT NEONATAL FOLLOW-UP PROGRAMS

Neonatal follow-up activities are a key part of the efforts to ensure the best possible outcomes for infants receiving care in neonatal intensive care units (NICUs). The goals of neonatal follow-up programs are more comprehensive than the goals of neonatal follow-up studies per se, but the two overlap; one could say that neonatal follow-up programs can exist without the participation of patients in well-designed neonatal follow-up studies, but neonatal studies cannot easily be done without relying on the framework of neonatal follow-up programs. For clarity, follow-up programs and follow-up studies are discussed separately.

NEONATAL FOLLOW-UP PROGRAMS

Audit

Audit is often defined as "a systematic and objective examination of evidence of the performance of a program, activity or function in order to facilitate decision-making" (21). Outcomes research has been described as the study of "the end results of the structure and processes of health care on the health and well-being of patients and populations" (21). However, the terms 'audit' and 'outcomes research' are often used interchangeably in neonatal follow-up activities. Most neonatal follow-up programs describe audit as a major goal. In this context, the term 'audit' is used to describe the systematic study and reporting of long-term outcomes of high-risk infants with the intent of making changes to improve outcomes in the future. Audit information is used to provide unit-specific information on the outcomes of preterm and other critically ill newborns to be passed on to health care workers in NICUs, obstetrics and family medicine, as well as to parents anticipating the birth of a preterm infant or one who is likely to experience a critical neonatal illness.

Education

Neonatal follow-up programs may provide educational experiences for medical or nursing undergraduate or graduate clinical trainees, as well as for learners in other health care disciplines, such as physiotherapy, psychology, occupational therapy, and speech and language pathology. A unique component of neonatal follow-up programs from an educational viewpoint is the unique multidisciplinary opportunities that they provide for learning how to assess 'normal infants', those who are near normal and those who have emerging signs of disabling conditions.

Clinical

It has been pointed out that the effectiveness of the clinical roles of neonatal follow-up programs has not been studied using randomized clinical trials (22). Part of the reason for this may be that the clinical roles vary a great deal between different follow-up programs. Individuals working in follow-up programs are often faced with the dilemma of attempting to maintain a research focus and, hence, avoid active involvement in the care of their subjects while simultaneously having to answer parents' questions about general health and development issues. Often, they are faced with infants who arrive at the clinic with acute illnesses requiring medical attention. In addition, there is an obligation for follow-up staff to facilitate referrals and, sometimes, to provide interim care for the infants and children who turn out to have disabilities until they can begin active involvement in a treatment or support program. Many clinics have developed plans to deal with such issues. However, there is no simple or uniform formula within the neonatal follow-up program context across Canada.

Most of the infants and children seen in follow-up clinics have their own family doctors or paediatricians. It is critical to avoid confusion and miscommunication, and to make the best use of all resources in the care of these infants and children. An approach to this is to maintain excellent communication with family physicians and community paediatricians, and, when applicable, with public health nurses and personnel in other health areas to provide the best possible service to infants and their families.

In some centres, unique programs exist to provide ongoing expert respiratory care to infants with chronic lung disease, including those who have been discharged home on oxygen. One would expect that such care would not be provided in follow-up clinics but would be coordinated with routine follow-up visits. Ophthalmological treatment may be provided in conjunction with follow-up clinics or separately. Audiology testing is usually done in follow-up clinics and needs to be linked with ongoing audiology or otolaryngology treatment services. Feeding and growth issues are common, especially in the smallest of infants, or in those with significant chronic lung disease, cardiac disease or other difficulties affecting food intake. In some cases, dietitians are part of a

neonatal follow-up team, or are available on a consultation basis or as part of a 'feeding team'. Developmental interventions – for example, in the areas of physiotherapy, occupational therapy, speech and language pathology, psychology or education – are often recommended for infants who were preterm. At times, therapies in these areas may be initiated through follow-up clinics that also provide interim care. Many of the infants and children followed through follow-up clinics are also being seen by neurologists, gastroenterologists, nephrologists or other medical and surgical specialists. Coordination of the care of each such child is the role of the family physician or paediatrician, but follow-up clinic personnel can often provide help in such situations.

Individuals working in follow-up clinics need to continually pay attention to the importance of bias that can inadvertently be introduced into their 'audit' activities. Follow-up clinics should avoid long-term care involvement, and should try to keep evaluations and treatment activities separate. The multidisciplinary nature of most clinics and the maintenance of blinding in the case of follow-up examinations being done as part of clinical trials may help to some extent.

NEONATAL FOLLOW-UP STUDIES

Neonatal follow-up research is usually focused on studies of the outcomes of specific groups of infants, determination of relationships between early findings and later outcomes, studies of different types of interventions, cost-benefit and cost-effectiveness evaluations, and perinatal or neonatal clinical trials. Elements from disciplines such as epidemiology, psychology, health services research, economics and psychometrics are typically incorporated into follow-up studies. Some of the well-recognized issues in methodological design and approaches of neonatal follow-up studies have included selection of a sample, choice of comparison group, consideration of social class, loss to follow-up, separating preterm from small for gestational age infants, optimal ages at testing and issues in reporting of results (23). Saigal (24) described common types and sources of bias to be considered in follow-up studies, including biases in selection of the inception cohort, biases in measuring exposure and outcomes, and biases in the analysis and interpretation of data.

Most current Canadian neonatal follow-up studies are set up as cohort studies, which follow clearly defined groups of infants using population-based or geographically based populations (13,14) or, in a few cases, as randomized controlled clinical trials (17-19). The outcomes of interest include survival and a variety of long-term outcomes in areas such as physical growth, health measures, cognitive function, hearing ability, visual function, fine and gross motor skills, physical and neurological examination, and social and behavioural function. Some studies include quality of life and cost measures, while most incorporate neonatal and infancy factors that are of potential value as predictors of specific outcomes, or for analyses of comorbidities or confounding factors. Most of the studies have followed infants beyond infancy to early or late childhood, but some have reported findings up until early adulthood (20).

CONCLUSION

The practices of neonatology and neonatal follow-up have arrived at a highly developed place in Canada, on the background of a rich history from which we continue to take lessons. Developments in follow-up programs and approaches to follow-up research ensure that we keep our goals in mind to ensure the best outcomes possible for infants requiring care in NICUs.

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LETTER TO THE EDITOR

Equipoise with respect to wrapping premature newborns immediately after delivery

To the Editor;

We noted the recently updated neonatal resuscitation program guidelines (1) and synopsis (2) and thought it was important to clarify whether or not equipoise exists with respect to wrapping premature newborns immediately after delivery.

We certainly appreciate the attention given to recent efforts to reduce hypothermia in premature newborns (3,4). As our recent systematic review illustrates (5), we are reasonably confident that wrapping reduces heat loss in this population. While there has often been a lag between evidence and practice, it seems this research has already had an impact: two recent surveys (6,7) confirm 20% to 29% of neonatal intensive care units now routinely wrap this population, albeit with great variation in how this practice is applied. We think it is important to stress that we do not yet know the long-term significance of this intervention. The Canadian Institute of Health Research and the Stollery Children's Hospital Foundation have funded a large, international, multicentre study to examine the effects on wrapping premature newborns on morbidity and mortality, which is presently underway in collaboration with the Vermont Oxford Network (8). Until the results of this trial are known, we think equipoise exists with regard to the long-term outcome of wrapping premature newborns, and we are not yet ready to recommend this practice to be included as part of the standard of care.

Sunita Vohra MD FRCPC MSc
Associate Professor of Pediatrics,
University of Alberta, Edmonton, Alberta
Co-PI, VON HeLP trial

Maureen Reilly RRT RRCP
Neonatal Intensive Care Unit,
Sunnybrook and Women's College Health Sciences Centre,
Toronto, Ontario
Co-PI, VON HeLP trial

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The authors respond;

Thanks for your letter regarding equipoise in wrapping preterm babies in plastic. There is evidence that cold preterm babies do not do as well as warm babies. There is good evidence that admission temperature is better with wrapping. We do not believe that centres should be asked to accidentally cool babies. The International Liaison Committee on Resuscitation statement reflects the scientific evidence.

Nalini Singhal MD FRCPC
Division of Neonatology, Department of Pediatrics,
University of Calgary, Calgary, Alberta

Allan de Caen MD FRCPC
Division of Pediatric Critical Care, Department of Pediatrics,
University of Alberta, Edmonton, Alberta