

Paediatric palliative care research in Canada: Development and progress of a new emerging team

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Paediatric palliative care is a field distinct from adult palliative care, although there are many overlaps in language, approach and philosophy. Several features, however, distinguish paediatric palliative care. The illnesses that affect children are different from those that are most predominant in the adult population. In addition, the role and involvement of the family, while always important in palliative care generally, is heightened in paediatric palliative care. In this new and growing interdisciplinary specialty, paediatric palliative care professionals recognize that children exist within a family system, with individual members making up the components (1). These distinguishing characteristics mean that we cannot simply translate general or adult palliative care research to the paediatric arena. Canada has been a leader in the development of clinical paediatric palliative care, with programs in the major Canadian geographical centres, as well as North America's first free-standing children's hospice, Canuck Place Children's Hospice (Vancouver, British Columbia). At the same time, there is widespread acknowledgement that not enough research has been undertaken within paediatric palliative care to provide an adequate evidence base for practice. To increase research capacity in Canada, in 2004, the Canadian Institutes of Health Research funded a research program entitled 'Transitions in Pediatric Palliative and End-of-Life Care' through a New Emerging Team grant.

WHY THE NEED FOR PAEDIATRIC PALLIATIVE CARE RESEARCH?

Children requiring palliative care suffer from a variety of physiologically diverse conditions that are variable in life expectancy and call for tailored treatments (2-4). Research on diseases such as cancer, neuromuscular and neurodegenerative diseases, metabolic disorders and genetic syndromes has led to improved understanding of the cellular disruptions underlying these diseases, advances in diagnostic techniques, and emerging curative or life-prolonging treatments. However, there has been little research relevant to paediatric palliative care in the areas of symptomatic pathophysiology, trajectory of disease progression, psychosocial

considerations, family experiences and access to health care resources (5).

In 2004, Gold (6) reported on the status of paediatric research in Canada by compiling a list of all research articles published in 2003 involving paediatric patients in Canada. There were a total of 262 articles in the analysis that included topic, journal name and geographical location of the principal author. None of the articles were classified as 'end-of-life' or 'palliative care', although there were three articles classified as 'death'. In addition, none of the articles were published in any of the recognized palliative care journals. While there have been some reports on paediatric palliative care research before and after 2003, published work still remains very limited (7). The *International Journal of Paediatric Palliative Care*, published by the Association for Children's Palliative Care (previously called ACT), compiles an annual review of literature in the field. There has been a significant increase since 2001 in both the volume and depth of articles published (8).

Currently, there is an emphasis on the practice of evidence-based medical practices and guidelines. The development of guidelines for palliative care for children living with a life-threatening, life-limiting, terminal illness is still in its relative infancy. Most guidelines are based solely on consensus or expert opinion because there is a paucity of evidence to guide the development of policy and minimum standards (unpublished data). The American Academy of Paediatrics released a statement (9) in 2000 concerning paediatric palliative care. They suggested that continued development of paediatric palliative care through research and education was essential for the improvement of services.

In addition to the guidelines, the need for quality measurements for health care in children are lacking in paediatric palliative care. The Agency for Healthcare Research and Quality, and numerous sponsors held a meeting in 2002 to review and assess the state of the science and practice of quality measurements for children's health care, to identify gaps in this area and to develop priorities for future investment. This group noted that since 1997, there have been considerable efforts to improve the quality of child health

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TABLE 1
Grants received to date

Project title	Funding source
Transitions in Pediatric Palliative and End-of-Life Care	Canadian Institutes of Health Research (CIHR)
Caregiving Parents of Children with Life-Limiting Illnesses: Beyond Stress and Coping to Growth	CIHR
An Analysis of Patient & Family Flow in Pediatric Palliative Care	University of British Columbia (Vancouver, British Columbia)
Parental and Professional Decision-Making Regarding Benefits and Burdens of Interventions for Children with Life-Limiting Conditions	University of British Columbia, Health Research Resource (Vancouver, British Columbia)
Transitions for Youth with Chronic Health Conditions	Michael Smith Child & Youth Research Network (Vancouver, British Columbia)
Parental and Professional Decision-Making Regarding Benefits and Burdens of Interventions for Children with Life-Limiting Conditions	Michael Smith Child & Youth Research Network (Vancouver, British Columbia)
Nutritional Failure in Pediatric Palliative Care	CIHR
Health Transitions in Adolescents with Chronic, Life-Limiting Cardiorespiratory Diseases Workshop	CIHR

care. Before the meeting, the group had commissioned an article to review the current literature (11). The article used the Institute of Medicine framework for quality to characterize the availability of measures. This framework includes six ‘quality domains’ and four ‘consumer perspectives’ (staying healthy, getting better, living with illness and end of life). In assessing the 396 measures currently being applied to children’s health care, only 17% were categorized as applying to children living with chronic illness or disability. There was a complete lack of available measures for children at the end of life.

DEVELOPMENT OF THE RESEARCH PROGRAM

The Transitions in Pediatric Palliative and End-of-Life Care new emerging team proposed to undertake research that would create a holistic picture of transitions of children and families living with life-limiting conditions. In undertaking this research, the team committed to four main goals – to build capacity by establishing and supporting new investigators; to create an innovative model of collaborative multidisciplinary work supported via electronic collaboration; to establish an evidence base for improved policy and practice; and to generate resources for families, health service providers and policy makers.

The composition of the team was crucial to its development, especially given previous concerns reported by Carter et al (12) about the lack of coordinated interdisciplinary services in paediatric palliative care. Through discussions with multiple stakeholders, a team was sought to encompass the broad spectrum of paediatric palliative care,

such as the physical, emotional and social components. This approach aligns well with converging trends in family research and paediatrics because it recognizes that family constitutes the most important social context, and it parallels the movement of organizations toward family-centred care with an emphasis on practice and research on family strengths and processes. The composition of the core research team, therefore, required a multidisciplinary group that included physicians, nurses and social workers. This core team consisted of five members who are affiliated with several teaching institutions. These affiliations allow for the support of trainees and new investigators in diverse fields, such as medicine, nursing, social work, psychology and health care management, to enhance capacity in paediatric palliative care research.

The paediatric palliative care community within Canada is small. Therefore, collaboration and partnerships are critical if the research evidence in paediatric palliative care is to advance in a timely manner. The team works jointly on some projects to examine the content and meaning of transitions from multiple perspectives to ensure that the biological, psychological, social and spiritual aspects of families’ experiences are all uncovered. In other cases, specific team members collaborate with researchers and clinicians outside the core group to further develop research in paediatric palliative care and mentor junior researchers. The team recognizes the value of such collaborations and partnerships; multiple links to the palliative and paediatric communities were sought.

RESEARCH THEMES

The Transitions in Pediatric Palliative and End-of-Life Care research team chose ‘transitions’ as a unifying theme because the palliative trajectory for children and families who live with life-threatening conditions is marked by numerous transitions, regardless of whether the condition is acute, subacute or chronic. These transitions affect the child or youth, the family, the health professionals and the community. Some transitions are clinical-biomedical in nature as the disease progresses. Others are transitions in care, for example, from cure to palliation. Some transitions occur because of natural growth and development, from infancy through adulthood, while also living with a life-threatening condition. There are also spiritual transitions from the time of diagnosis, through death and into bereavement. The focus of the present research is on these major transitional themes as experienced by patients, families and health care professionals within paediatric palliative care.

PROGRESS TO DATE

Since its inception, the research team has filed 18 grants, nine of which have been successful (approximately \$1.5 million in funding) (Table 1). Several workshops and stakeholder meetings have been held to facilitate communication, consensus building, and dissemination of information and ideas. These meetings allow for exchange of ideas, planning of future studies and opportunities to highlight the research of trainees

TABLE 2
Workshops and stakeholder meetings

Workshops and meetings	Summary	Date and location
Transitions for Youth with Chronic Health Conditions	This workshop explored the research questions surrounding transitions experienced by youth with chronic cardiorespiratory health conditions (physical, mental and developmental)	March 2008 in Vancouver, British Columbia
Stakeholder Meeting 2007	This stakeholder meeting brought together the core research team members, research trainees, affiliated students and stakeholders	November 2007 in Toronto, Ontario
Nutritional Failure in Paediatric Palliative Care	This workshop brought together participants to develop a common description and understanding of the clinical phenomenon of nutritional failure at the end of life in a paediatric population, and to develop new research initiatives in this area	February 2006 in Vancouver, British Columbia
Stakeholder Meeting 2006	This meeting provided an opportunity to brainstorm research ideas and make connections among researchers	September 2006 in Montreal, Quebec

TABLE 3
Research trainees and affiliated students (2007–2008)

Discipline	University (location)	Level
Research trainees		
Social work	University of British Columbia (Vancouver, British Columbia)	PhD
Psychology	Simon Fraser University (Burnaby, British Columbia)	PhD
Nursing	University of Toronto (Toronto, Ontario)	PhD
Nursing	University of British Columbia (Vancouver, British Columbia)	Masters
Social work	Wilfrid Laurier University (Waterloo, Ontario)	Masters
Education	University of Ottawa (Ottawa, Ontario)	MD/Masters
Affiliated students		
Nursing	University of California, San Francisco (California, USA)	PhD
Social work	University of British Columbia (Vancouver, British Columbia)	PhD
Nursing	McMaster University (Hamilton, Ontario)	PhD
Paediatrics	University of Melbourne (Victoria, Australia)	PhD
Nursing	University of Calgary (Calgary, Alberta)	PhD
Nursing	University of California, San Francisco (California, USA)	PhD
Nursing	University of California, San Francisco (California, USA)	PhD
Interdisciplinary studies	Dalhousie University (Halifax, Nova Scotia)	MD/PhD
Social work	University of Toronto (Toronto, Ontario)	Masters
Bioethics	University of Toronto (Toronto, Ontario)	Masters

MD Medical doctor; PhD Doctor of philosophy

(Table 2). To increase research capacity in Canada, the grant has funded or is affiliated with many students at various degree levels. Funded students receive a Canadian Institutes of Health Research-equivalent stipend for up to three years of study, in addition to funding to attend relevant conferences. Affiliated students receive their stipends from other sources, and the grant provides funding for attending conferences or small amounts of project funding to complete studies for their degrees (Table 3).

CHALLENGES IN CONDUCTING PAEDIATRIC PALLIATIVE CARE RESEARCH

Several challenges in conducting paediatric palliative care research have been encountered to date. Because the core investigators work in five different sites in three different cities and have multiple collaborators and partners, it was important that a robust communication and information

system support their work. Therefore, a shared electronic workspace called 'CoLab' was developed. The central component of 'CoLab' is a Web-based electronic workspace that offers document handling, e-mail, live chat and archive functions. Another helpful aspect of 'CoLab' includes the use of a telephone, video-conferencing, standard e-mail and Web-based reference management tools.

It must also be acknowledged that there are difficulties in end-of-life research and especially in research that involves children. Evidence-based medicine is at the core of current medicine, yet paediatric palliative care is one of the so-called 'orphaned fields' of medicine in which medical research is weak and diverse, financial incentives are lacking and the evidence regarding etiology and treatment of disease is much less clear (13-15). Research in end-of-life care has been described as more constrained by social, cultural and financial barriers than ethical concerns. Fine (16) stressed that "involvement

in research may have a therapeutic, anticomiogenic effect on dying patients and their families". Therefore, it is imperative that researchers design research protocols involving children at the end of life to ensure participation maximizes resources, time and emotional investment. This must be accomplished in addition to using appropriate research methodology and transparency regarding conflicts of interest (17). Finally, clinical personnel must be made aware of the benefits of research with this population so that they support such research and do not inhibit patients' and families' abilities to choose to enrol in research (18). This phenomenon became evident in the first multicentre project that required multisite ethics. A total of 11 research ethics board reviews resulted in the project not beginning recruitment until year 2 of the study (unpublished data).

THE FUTURE OF PAEDIATRIC PALLIATIVE CARE RESEARCH

Research in paediatric palliative care has developed significantly over the past several years. It is imperative to continue to build capacity in paediatric palliative care research that will be sustainable and ongoing in the foreseeable future. Some specific objectives including the creation of a sustainable enterprise to support new and emerging research in the field that corresponds to three specific areas (clinical-biomedical, family and health services) within the theme of transitions have been initiated. Tools such as a shared virtual electronic workspace, video-conferencing and live symposia support this work. Future developments include a database that makes information accessible to investigators, institutions and communities, and an extension of the research themes into a wider age range, including the transition to adult health care services for adolescents with life-limiting diseases.

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