THE INFLUENCE OF A PEDIATRIC PALLIATIVE CARE PROGRAM ON HEALTH CARE UTILIZATION AND COSTS

by

Tania Conte

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Abstract

Pediatric palliative care program (PPCP) is believed to increase system efficiency. British Columbia (BC) relies on a free-standing hospice-based PPCP, and its effects on health care utilization and costs remain an outstanding question. This study aimed to gather evidence in a combined analysis of data from literature and BC PPCP.

A systematic review was conducted through an electronic search of Medline, Embase, CINAHL, LILACS, and grey literature. Comparative studies reporting admissions, length of stay, and health care costs between PPCP users and usual care were included. Additionally, a similar comparison was applied to the data from BC PPCP using a retrospective matched-pairs cohort design (matched by ICD code and age at death) with a 3-year observational period prior to death. Data were obtained from Canuck Place Children's Hospice and BC Children's Hospital databases, and complemented by estimates from Canadian Institute for Health Information. A cost impact of the overall inpatient care provided by the hospice was presented.

The review did not demonstrate a decrease in utilization by PPCP users yet suggested a shift to other health care settings, and potential cost saving in the Canadian context (1 article). The cohort study (n=11 pairs), suggests that children in both groups had similar upward trends in inpatient utilization and cost. However, PPCP users showed more inpatient care in the last year of life (especially critical care in the last 2 months), compared to their controls and to the period prior to referral. Post-referral, a shift in health care setting utilization from hospital to hospice was observed, representing approximately 50% of the costs. Without this shift PPCP users would have cost 32% more with a median monthly increment of $7,163 per child. All inpatient care provided by the hospice in the fiscal year 2011-2012 represented a potential cost saving ranging from approximately $1.1M to $4.3M. The findings of this study suggest that PPCP users may present higher health care needs, and that the shift of inpatient care to the hospice optimized resource use, offering a more holistic approach to EOL care, relieving hospital resources to meet other demands.

Preface

I was involved in the conceptualization, study design, data collection, analysis, and interpretation of all three studies in this research.

A version of Chapter 2 has been accepted for publication ["Effects of Pediatric Palliative Care Programs for Children with Life-Threatening Conditions in Health care Resources Utilization and Costs: a Systematic Review of Comparative Studies", CMAJ Open 2014, to be released] for which I was the corresponding author, responsible for all the major areas such as the systematic search and review, data synthesis and manuscript composition. Logan Trenaman and Dr. Harold Siden contributed as additional reviewers of the systematic review, and along with Negar Chavoshi and Dr. Craig Mitton contributed significantly to the analysis and interpretation of the results, and multiple editing to the manuscript. Abstracts of this systematic review were presented at Health Technology Assessment International Meeting (HTAi) 2014 and 20th International Conference in Palliative Care.

In Chapters 3 and 4, I was involved in the design, data collection, analysis and interpretation of the cohort study results with support a multidisciplinary team. Dr. Craig Mitton was my supervisor involved from the early stages of conceptualization, interpretation of the results, and made substantial contributions to the structure of the research. Dr. Harold Siden and Negar Chavoshi participated significantly in the matching process, interpretation and editing of the findings, and Shanon Erderlyi contributed with the coding for the analysis and graphs. Cindy Stutzer and Rod Rassekh consulted with the cancer matching process. Tanice Miller and Joan Hill contributed with operating cost data from the hospice. Multiple members of PHSA Decision Support team participated retrieving and processing confidential data on utilization and cost for the hospital facility. This research was granted approval from the Research Ethics Board of Children's and Women's Health Centre of British Columbia (certificate numbers CW13-0210 / H13-01162) and Canuck Place Research Review Committee.

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List of Abbreviations

ACT: Association of Children's Palliative Care [Together for Short Lives]

BC: British Columbia

BCCH: British Columbia Children's Hospital

CADTH: Canadian Agency for Drugs and Technologies in Health

CMAJ: Canadian Medical Association Journal

CHPCA: Canadian Hospice Palliative Care Association

CIHI: Canadian Institute for Health Information

CMDB: Canadian MIS Database

CNPCC: Canadian Network of Palliative Care for Children

CI: Confidence Interval

CPCH: Canuck Place Children's Hospice

DNAR: Do Not Attempt Resuscitation

EMR: Electronic Medical Records

EOL: End-of-Life

GPs: General Practitioners

ICU: Intensive Care Unit

ICD: International Statistical Classification of Diseases and Related Health Problems

IT: Information Technologies

LOD: Location of Death

LOP: Length onto the Program

LOS: Length of Stay

LTCs: Life-Threatening Conditions

MeSH: Medical Service Headings

MNPR: Mandated Nurse-Patient Ratios

NICU: Neonatal Intensive Care Unit

NOS: Newcastle-Ottawa Scale

PC: Palliative Care

PedPalASCNET: Collaborative Research in Pediatric Palliative Care

PHSA: Provincial Health Services Authority

PICU: Pediatric Intensive Care Unit

PPC: Pediatric Palliative Care

PPCP: Pediatric Palliative Care Program

QALY: Quality-Adjusted Life Year

RCT: Randomized Controlled Trials

Sd: Standard Deviation

UK: United Kingdom

USA: United States of America

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I acknowledge and thank all of you for assisting me in this journey.

Dedication

I dedicate this work to...

God first, for giving me the strength to pursue this hard journey.

To my entire family with a special feeling of gratitude to my loving parents, Rosina and Francisco who dealt with my absence and physical distance throughout difficult moments of loss with understanding.

To Ryan and the MacAulay's for grounding me here during the hard times of grad school.

To all my friends worldwide for making me feel close to their hearts even when I was far away from their eyes.

This thesis is lovingly dedicated in memory of my grandmother, Detizia.

Chapter 1: Introduction

The death of a child might be the most devastating tragedy a family can face. Care for severely sick children, and their families, is central to pediatrics; however, only recently palliative care (PC) for the pediatric population has been considered as a sub-specialty within pediatrics. Consequently, the most suitable approaches to pediatric palliative care (PPC) are relatively in the early stages of development, and continuously evolving.[1]

There is clinical interest in understanding how the different care settings where end-of-life (EOL) care is provided for children with life-threatening conditions (LTCs) have a differential impact on patients and their families (e.g. hospital, home, hospice, etc.). Furthermore, it is not clear how different approaches to EOL and management of symptoms for LTCs across providers affect both costs and health care utilization. It is known that a disproportionate amount of health care funds are being spent on individuals in their final months of life [2]. Yet, evidence to informs how best to make these allocations are scarce.

There is a prevailing belief that PPC programs (PPCP) can deliver services more efficiently by improving care offered in settings other than tertiary centres, more specifically in hospices or at home. Nonetheless, how to reallocate resources for each of these types of services and programs remains an outstanding question.

British Columbia (BC) has a holistic PPCP carried out by a freestanding pediatric hospice that coordinates the care plan for referred children across the public funded health care settings thus offering a natural experiment to study such questions for hospice-based programs.

In order to provide evidence to support PPCP evaluation, planning, and funding, a combined analysis of data from the published literature and the BC PPCP performed to study the effects of PPCP on cost and utilization. Chapter 2 presents a systematic review of the literature investigating

differences in patterns of utilization and cost among children with LTCs, who enrolled in PPCP compared to those who did not. It explores if and how research was conducted in this field, and compares the outcomes of PPCP according to different settings. Chapter 3 presents a comparison of the utilization patterns at EOL among children who died while enrolled in the provincial PPCP compared to those who were not. Chapter 4 presents a cost analysis of the utilization in this population, according to the different settings, and the impact on the health care system.

1.1 Background

1.1.1 Conceptual Framework

Health systems are organized in such a manner that the process of health care delivery affects health system outcome and consequently population health outcomes (Figure 1). Population needs, access to care, indirect effects and utilization of the health system affect these processes of delivery, which includes the organization of health care, or ways resources are deployed, and the actual process of care.[3] In the context of this research the place or program that is providing care (home, tertiary care, community hospital or hospice) to meet the needs of pediatric population with LTCs will affect health system performance. Being enrolled in any specific model of PPCP likely has an effect on differences on management approach (curative or palliative), health care utilization of other health care settings, and a variety of levels of care to meet patient needs.

Figure 1. Macro Model of Health System

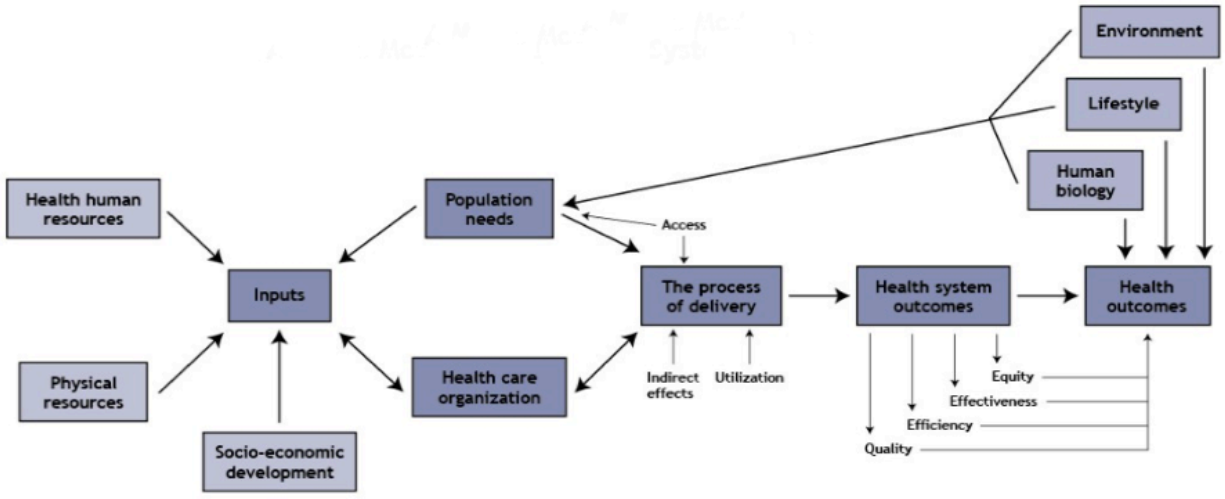


Image adapted with permission from Analyzing Health Systems: a modular approach @ Oxford University Press, p.38[3]

1.1.2 Life-threatening Conditions - Definition

According to the Canadian Network for Accessible, Sustainable and Collaborative Research in Pediatric Palliative Care (PedPalASCNET), LTCs are "conditions for which there is a likelihood of death before adulthood. It includes those for which curative treatments may be feasible but may fail, or those for which a cure is not possible and from which an affected child is expected to die. They are frequently chronic complex conditions with significant impact upon the lives of the child and family".[4] The terminology may vary in other countries where they can also be defined as terminal or life-shortening conditions (or diseases).[5]

1.1.2.1 Categorization of Life-threatening Conditions

In order to help categorize these conditions, the advocacy organization Together for Short Lives (previously called Association of Children's Palliative Care - ACT), delineates 4 large groups of LTCs (see Table 1 below). This categorization relies exclusively on diagnosis; however, severity of

disease, impact of the disease on child's functionality and family dynamics, subsequent complications, and level of care required should also be considered.[5]

The range of diagnoses under these categories is extremely wide (over 300 conditions), with a degree of overlap with severe disabilities and complex needs. Although cancer patients constitute a significant proportion of the children eligible for PPCP, the majority of the workload in this specialty is applied to inherited metabolic disease, neuromuscular diseases and acquired brain injury. Approximately 15% of these children do not have a definitive primary and obvious diagnosis, and palliative care is usually delivered over a longer time frame compared to adult palliative care.[6-9]

Table 1: Categorization of LTCs

Category 1	**Category 2**
Life-threatening conditions for which curative treatment may be feasible but can fail. Where access to palliative care services may be necessary when treatment fails or during an acute crisis, irrespective of the duration of that threat to life. On reaching long-term remission or following successful curative treatment there is no longer a need for palliative care services. Examples: cancer, irreversible organ failures of heart, liver, kidney.	Conditions when premature death is inevitable, where there may be long periods of intensive treatment aimed at prolonging life and allowing participation in normal activities. Examples: cystic fibrosis, Duchenne muscular dystrophy.
Category 3	**Category 4**
Progressive conditions without curative treatment options, where treatment is exclusively palliative and may commonly extend over many years. Examples: Batten disease, mucopolysaccharidoses.	Irreversible but non-progressive conditions, causing severe disability, leading to susceptibility to health complications and likelihood of premature death. Examples: severe cerebral palsy, multiple disabilities such as following brain or spinal cord injury, complex health care needs and a high risk of an unpredictable life-threatening event or episode.

Adapted with permission from A Guide to the Development of Children's Palliative Care Services © Association for Children's Palliative Care. [5]

1.1.3 Pediatric Palliative Care Definition and Criteria

The movement for PPCP in hospices started to be developed in the early 80's in the United Kingdom (UK), with the opening of Helen House hospice in Oxford, recognized as the pioneer in this field. [10] The UK recognized pediatric palliative care as a medical specialty in 2009. The field has a later development in North America and a number of other countries in the mid 90's. In Canada, the first hospital-based program started in 1986, and the first-hospice based program opened in 1995.[11]

PPCP provides services to individuals with LTC under the age of 19. Individuals aged ≥ 19 years are considered young adults.[5] However, there is no consistency in the age range, even within the PPCP literature, and some authors also include individuals under 20 years of age as children.[12]

According to ACT, palliative care for children and young people is defined as *"an active and total approach to care, from the point of diagnosis or recognition, throughout the child's life, death and beyond. It embraces physical, emotional, social and spiritual elements and focuses on the enhancement of quality of life for the child and support for the family. It includes the management of distressing symptoms, provision of short breaks for the caregivers, and care through death and bereavement. It is distinct from disabled children's care in a number of ways. Although many children with palliative care needs are disabled, the risk or certainty of death in childhood adds a degree of complexity and urgency to their care and the support that is needed for their family. And compared to disabled children, their need for services is more likely to fluctuate due to the particular nature of their illness trajectory, social, emotional and physical needs" (pg 7)*.[5]

Children with the same condition may require PPCP during different periods of life or progression of the disease, the rate and presentation of which can vary between them. In the same way, families' willingness to pursue treatments to significantly prolong life can range from

supportive and relieving care, to aggressive and invasive treatments. Ideally, palliative care should be proposed from the moment of the diagnosis or from the point at which it is acknowledged that curative treatment is not available. PPCP is evolving and practitioners are increasingly recognizing that each child and their family will need an individualized range of support mechanisms. [5]

Figure 2 projects the relative focus of care. The dashed line is differentiating between therapies intended to modify disease from those intended to enhance quality of life [13]. The degree in which the combination of both therapies will vary for each child and family is based on their expectations, needs, goals of care, and treatment priorities.

Figure 2. The Role of Pediatric Palliative Care

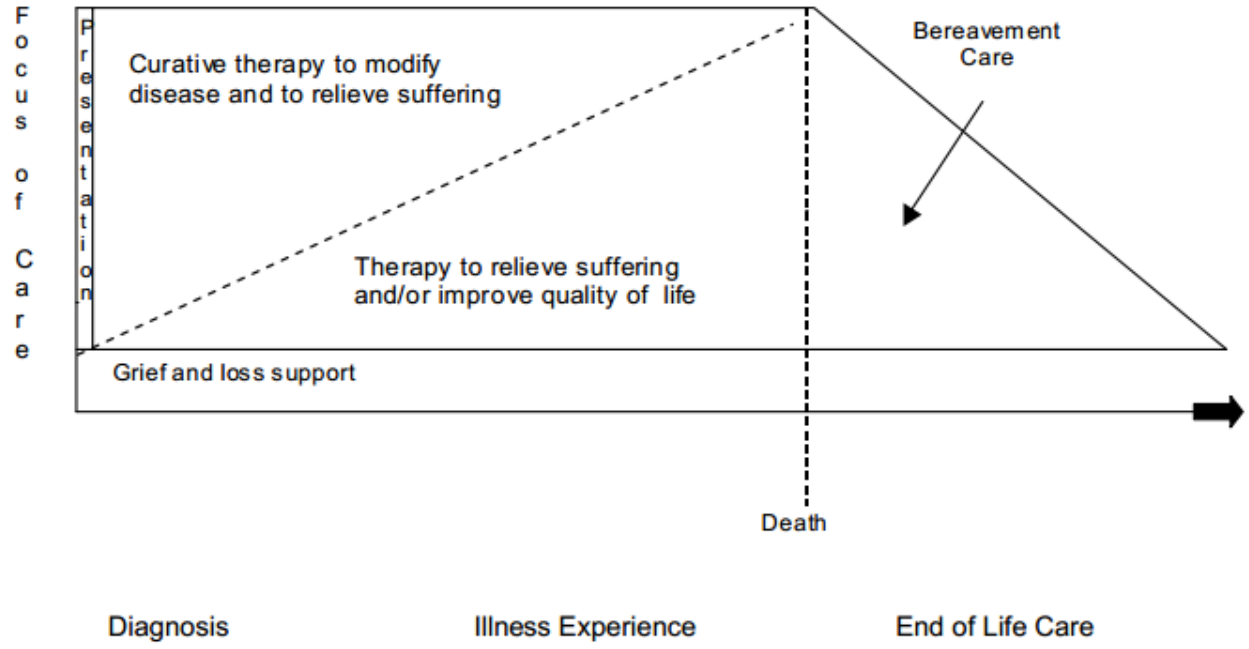


Image reproduced with permission from Pediatric Hospice Palliative Care: Guiding Principles and Norms of Practice © Canadian Hospice Palliative Care Association, Ottawa, Ontario, Canada.[13]

In some sense, every child who died in a health care provider setting have received some sort of palliative care through procedures such as pain management, intravenous hydration,

transfusions, suctioning, feeding tubes and other comfort measures. However, in order to meet the ACT criteria for PPCP, it is expected that care providers implement a broader program to attend the range of needs and coordinate the services offered beyond palliative procedures, but including other aspects of care such as respite, counselling (including expressive and play therapy), school, music therapy, recreation opportunities, sibling and parent support, and bereavement services. Tertiary care providers, hospices, outpatient clinics, community-based services, or partnership between them can all carry these programs. It is worth noting that the term "hospice" has a slightly different meaning in different countries. In Canada the term means an inpatient type setting whereas in the United States of America (USA) it is more likely to refer to a community-based palliative care team.[4] In the USA, care can be provided at home or in a designated facility, such as a nursing home, hospital unit or a freestanding hospice.

In 2006, the Canadian Network of Palliative Care for Children (CNPCC) and the Canadian Hospice Palliative Care Association's (CHPCA) launched guidelines known as the "National Principles and Norms of Practice for Pediatric Hospice Palliative Care"[13] in an attempt to facilitate access to high quality care programs regardless of where the care is delivered. The guiding principles of this approach to care are based on child/family-focused care, the value of the therapeutic relationship, continuity of care, communication and accessibility. Despite these guidelines, it is recognized that the availability of services and staff differs considerably between providers of palliative care, and hospices are more likely to provide complementary therapies. [14]

1.1.4 Estimates on Demand and Enrollment in Pediatric Palliative Care Programs

There are a variety of reasons as to why PPCP may often be underutilized. Chief among them is limitations of physicians' abilities to accurately predict a patient's life span and classify

them as being in the final stages of the disease. Furthermore, patients, families, as well as physicians have commonly (and understandably) an inclination to deny death, and thus do not seek or refer patients to PPCP, which are often perceived as services that are used when the patients are actively dying or when "nothing else can be done". Expert's opinions about when a patient should be referred, and the scope of palliative care specialty still differ; however, late referrals may limit the amount of care a PPCP can offer to patients and families.[15] A survey among medical providers comparing patient deaths, with or without hospice support, shows that 27% of the responders had a positive perception of the hospice services mainly related to non-medical support and place of death. In contrast, 12% of them provided negative comments on the service, involving feelings of loss of hope, intrusion and distrust.[16] Lack of palliative care education within many medical school programs may contribute to this discomfort with end-of-life care, and therefore may limit referrals to PPCP.[17]

More recently, the use of an evidence-based tool for identifying vulnerability factors that make children more likely to benefit from PPCP, and the use of the "surprise question", where clinicians are asked whether they would be "surprised if the child survived beyond their 18th birthday" seemed to have improved referral to palliative care programs. [8]

The prevalence of children living with LTCs worldwide ranges from 10 to 32 per 10,000 children. However, much debate is carried out regarding the appropriateness of the methods used to estimate demand for palliative care services.[1,18-20] In BC the prevalence is estimated at 17 per 10,000 individuals aged 0-24 years.[18-20] Despite scarcity of information on the pediatric population that could potentially benefit from palliative care, there is some evidence that a significant proportion of children with LTCs have died without being referred to an appropriate PPCP.

Data from the USA estimates the enrollment in PPCP ranges between 11% and 13% among infants who died in hospital. [21,22] A Canadian study reported that in 2002 there were only 7 programs of palliative care in tertiary care settings and 1 hospice across the country. The authors found that only 5-12% of children who were, in theory, eligible for PPCP in Canada had accessed those services before dying.[23] Since 2002, PPCP in Canada have grown and there are now more tertiary care programs and 7 pediatric hospices. Consequently, the estimates of those accessing services are now likely to be higher.

Evidence from the adult palliative care literature shows that these programs consistently improve patient and family satisfaction with care and may improve quality of EOL, yet data is inconclusive in regards to the impact on costs.[24-26] Moreover, there are important differences between pediatric and adult palliative care programs. From a resource use perspective, PPCP is usually delivered over a longer time frame compared to adult palliative care[7-9]; in addition, the mandate for PPCP is to provide support to siblings and parents as well. For these reasons resource utilization and cost analysis might be different from adult palliative care, however, it appears these outcomes have been understudied.

A literature search for systematic reviews investigating the impact of PPCP on health care utilization and costs yielded no results. The search found only 1 systematic review on the impact of pediatric home care for those with complex and long-term needs. Overall, the article found a decreased burden of care and costs for families, and reduced acute care admissions. However, the impact on costs is sensitive to case mix, skill mix and changes in the local health economy.[27]

Furthermore, although home care is part of the PPCP concept, children still require inpatient care from time to time. When children are enrolled in a PPCP the inpatient resource

utilization will likely vary according to the available settings such as hospital, hospice or a combination of the two.

1.1.5 The Pediatric Palliative Care Program in British Columbia and Rationale for this Study

In BC, children with LTC can be referred to a PPCP by physicians, other health professionals, family members, acquaintances or friends, as long as the family is aware and consents to the referral.[28] Once referred, the child and the family can be enrolled in the program, which is carried out by Canuck Place Children's Hospice (CPCH), in a partnership with BC Children's Hospital (BCCH).

BCCH is a tertiary teaching hospital and research facility founded in Vancouver in 1982. It provides specialized care for children from across the province, with an estimated capacity of over 200,000 children per year. BCCH uses the CPCH team as its palliative care program to assist children and families living with progressive LTCs, in those cases where a palliative referral is made and the family agrees to be enrolled in the CPCH program.[29] The enrollment is not a requirement of BCCH and in the absence of a referral and/or consent to participate in PPCP, families can expect primary clinical symptom management and the support of the hospital's counseling services, which do not constitute a broad palliative care approach according to ACT definition, but offers some degree of palliative care.

CPCH is a freestanding hospice that was founded in 1995 also in Vancouver, and is the only pediatric hospice facility in BC. It provides palliative care to BC residents from birth to 19 years of age who are living with progressive LTCs, and have been referred to the program. CPCH provides individualized palliative care for the children and support for their families, which includes around-the-clock consultation and support from a health care team, end-of-life care, pain and symptom management, respite, school, music and play therapy, and recreation opportunities. Counseling is

offered to the entire family to enhance their ability to cope with disease management and bereavement. The hospice also provides support for transition to adult care palliative services for young adults over 19 years of age.[28]

CPCH operates in Vancouver, with 9 beds and 4 family suites. The number of individuals admitted to CPCH has significantly increased over the past 8 years and, on average, the hospice provides care for approximately 250 children a year. [18,30] This capacity is still below the required amount to accommodate the provincial needs. It is estimated that there are approximately 1397 children living with a LTC in BC each year, with approximately 600 children in Greater Vancouver.[18,31,32] Given the relatively small number of beds and family suites, PPCP planning and evaluation is essential to ensure that services are delivered efficiently to those with the greatest need.

CPCH has plans to open a new unit in Abbotsford in 2015, thus, doubling the capacity. The hospice is funded primarily through donations and partnerships. Over 10 years, the Ministry of Health and Ministry of Children and Family Development funded only 26% of the hospice costs.[33*] However, CPCH operation directly affects the publicly funded health system since the inpatient admissions to the hospice facilities, and the support for families to manage the LTCs at home, reduce hospital admissions to BCCH. Needless to say, the home environment CPCH provides likely ameliorates the suffering experiences of EOL for children and families.[34]

Although home care is part of the BC PPCP concept, children still require inpatient care from time to time. When children are enrolled in the CPCH program, families can choose to have inpatient care either at the hospital or the hospice, which also depends on bed availability at the time of need. Children who required palliative care, and are not enrolled in the PPCP, still access

[*] Personal communication reproduced with permission.

health care through acute care admissions and some episodes of home care, through the regular pathways to access health services. Yet, they will most likely receive care in an uncoordinated fashion that potentially compromises the continuity of care, where critical components for family support are lacking. For the purpose of this study this way of accessing health care is referred to as 'usual care'.

It is unclear, and there is growing interest in understanding, how the enrollment in the BC PPC program affects health care utilization and costs compared to those who rely exclusively on usual care in BC. There is a prevailing belief that the PPCP can deliver services to this population more efficiently by coordinating care between the different health care settings and home care, instead of relying solely on tertiary care providers.

As the first free-standing hospice in North America, CPCH has contributed to relevant research by providing data on mortality trends and prevalence of pediatric LTC, resource utilization, and qualitative analysis of the benefits of respite care for families and children.[18,34] Likewise, CPCH continuously contributes to the limited national data on prevalence of LTC, service provision and place of death in Canada.[23,35] However, further program planning requires information on the impact of this approach to care on the costs and outcomes, and this field seems to be a critical yet understudied component of the health care system.

In order to support the program planning, evaluation and budgeting, evidence is needed. To date, no study has completed a systematic review of the literature on such outcomes of PPCP. The current project takes advantage of the natural opportunity for comparison of patients at BCCH who are and who are not enrolled in the CPCH program, and investigate how the enrollment in a comprehensive PPCP for children with LTCs affects health care resource utilization and costs.

1.2 Objectives

To gather the evidence on health care resource utilization and cost, a combined analysis of data from published literature and BC PPCP was completed. The specific objectives include a systematic review of the literature for studies on children with LTC comparing health care resource utilization and costs between PPCP users and usual care users. Additionally, a similar comparison with the data from BC PPCP was performed. These objectives were addressed via the following research questions and associated hypotheses:

1.2.1 Research Questions

1. In the published literature, how do children who accessed PPCP compare to those under usual care, in terms of health care resource utilization and cost, specifically with respect to:
    a. Admissions of any type – emergency visits, general ward, critical care, hospice;
    b. Length of stay (LOS);
    c. Health care costs of any nature - direct costs in hospital or hospice, indirect costs, total health care expenditures;
    d. Length of the last hospitalization before death;
    e. Number of invasive procedures to prolong life in the last admission;
    f. Number of resuscitation attempts?

2. In BC, how did children with LTC who died, and were enrolled in the PPCP, compare to similar children who died under usual care, specifically with respect to:
    a. Outpatient and inpatient admissions of any type – emergency visits, general ward, critical care, hospice;

b. LOS;

c. Length of the last hospitalization before death;

d. Utilization of invasive procedures to prolong life in the last admission;

e. Occurrence of resuscitation attempts?

3. In BC, how did children with LTC who died, and were enrolled in the PPCP, compare to similar children who died under usual care, with respect to direct health care cost related to outpatient and inpatient care in both settings – hospital and hospice?

4. In BC, how did the inpatient care provided by the hospice impacted costs for the publicly funded health care system?

1.2.2 Hypotheses

1. Both in the literature and in the local analysis, health care utilization will be lower among children who access PPCP due to better management of the conditions, the holistic approach to care, family training/education, and coordination of care across settings.
2. Both in the literature and in the local analysis, costs will be lower among children who access PPCP as a reflection of the influence on health care utilization.
3. In the health care settings with hospice facilities available, the costs will be lower among children who access PPCP under the assumption that hospice facilities present with lower operating costs than tertiary facilities.

The above-mentioned research questions will be addressed in 3 different chapters of research. Chapter 2 presents a systematic review of the literature. It explores if and how research has been conducted in this field, to compare the outcomes of PPCP across different settings. Chapter 3 presents a local comparison of the patterns of utilization among children who died while enrolled in the provincial PPCP compared to those who were not, across to the different settings. Chapter 4 presents a cost analysis of the utilization in this population, according to the different settings, and the impact on the health system. The specific methods and databases are described in each chapter, accordingly.

Chapter 2: Effects of PPCP for Children with LTC's in Health Care Resources Utilization and Costs: a Systematic Review of Comparative Studies

Overview

This chapter will provide an updated overview of the literature looking at how previous comparative research has studied the effects of PPCP on health care utilization and costs, results, quality of available evidence, limitations and implications for practice and research.

2.1 Objective

- To systematically review studies that have compared health care resource utilization and costs between children who accessed PPCP to those under usual care.

2.2 Methods

This systematic review was performed according to the PRISMA guidelines[36,37] following the subsequent criteria and process:

2.2.1 Inclusion Criteria

All types of comparative studies (experimental or observational studies, and secondary administrative databases analyses) were considered, regardless of length of follow-up. This decision was made because of the challenges of undertaking a randomized controlled trial (RCT) in this population. This includes ethical concerns of randomizing children to different approaches to care and stigma about palliative care. The review included published articles or abstracts from

conference procedures, retrieved through the automatized search strategies and grey literature review.

The study population of interest is specifically children, up to 22 years old, with a LTC condition, defined as any condition where there is no established curative treatment available, or where the available treatment has a high failure rate.[5] This age limit was chosen due to the lack of consistency in the upper age limit in this population,[5,12] and the fact that programs can support transition to adult care palliative services up to the age of 22.[28]

Until recently, there was a lack of standardization in what constituted a PPCP. For this reason, in this review, any studies that included a comprehensive PPCP or a common component of such programs, including PC consultation, respite care, EOL care or planning, or hospice or community-based palliative care, were eligible for inclusion.

Outcomes were chosen based on previous work in the adult palliative care literature, which has identified potential quality of end-of-life indicators that can be measured using administrative data.[38] The primary outcomes of interest were:

1. Health care resource utilization, measured as any of the following endpoints:
    a. Number of inpatient admissions of any type – emergency visits, general ward, critical care, hospice;
    b. LOS;
2. Health care costs of any nature (direct costs in hospital or hospice, indirect costs, total health care expenditures).

Secondary outcomes of interest include health care resources utilization in the *last admission before death*, measured by any of the following end points:

1. LOS;

2. Number of invasive procedures to prolong life;
3. Number of resuscitation attempts.

2.2.2 Search Methods for Identification of Studies

The Medical Service Heading (MeSH) terms for 'palliative care' return an extensive amount of publications, largely related to palliative procedures in cardiac malformations or other clinical outcomes. For feasibility, and given the relatively recent development of PPCP, the search was limited to articles published from 2000 to present. The search was completed on July 18th, 2013, and weekly-automated alerts were put in place for any publication after this date. No language limits were applied.

Studies were identified by searching electronic databases, scanning reference lists of articles, and consulting with experts in the field. This search was applied to Medline, adapted for Embase, CINAHL and LILACS. In addition, a grey literature search was undertaken, targeted at websites from specialized groups and societies in palliative care and hospice services. The search strategies for each electronic database and grey literature list are available in Appendix A.

A review of studies was undertaken independently and in duplicate (TC, LT). Disagreements were resolved through discussion, or if required, through consultation with an additional reviewer with clinical expertise in PPCP (HS).

2.2.3 Data Collection, Analysis and Quality Assessment

Data was extracted by TC and reviewed by LT to ensure consistency of reporting. Discrepancies were handled in the same manner as study identification. Studies were displayed in tables, exploring each study's approach to program evaluation. A meta-analysis was not feasible

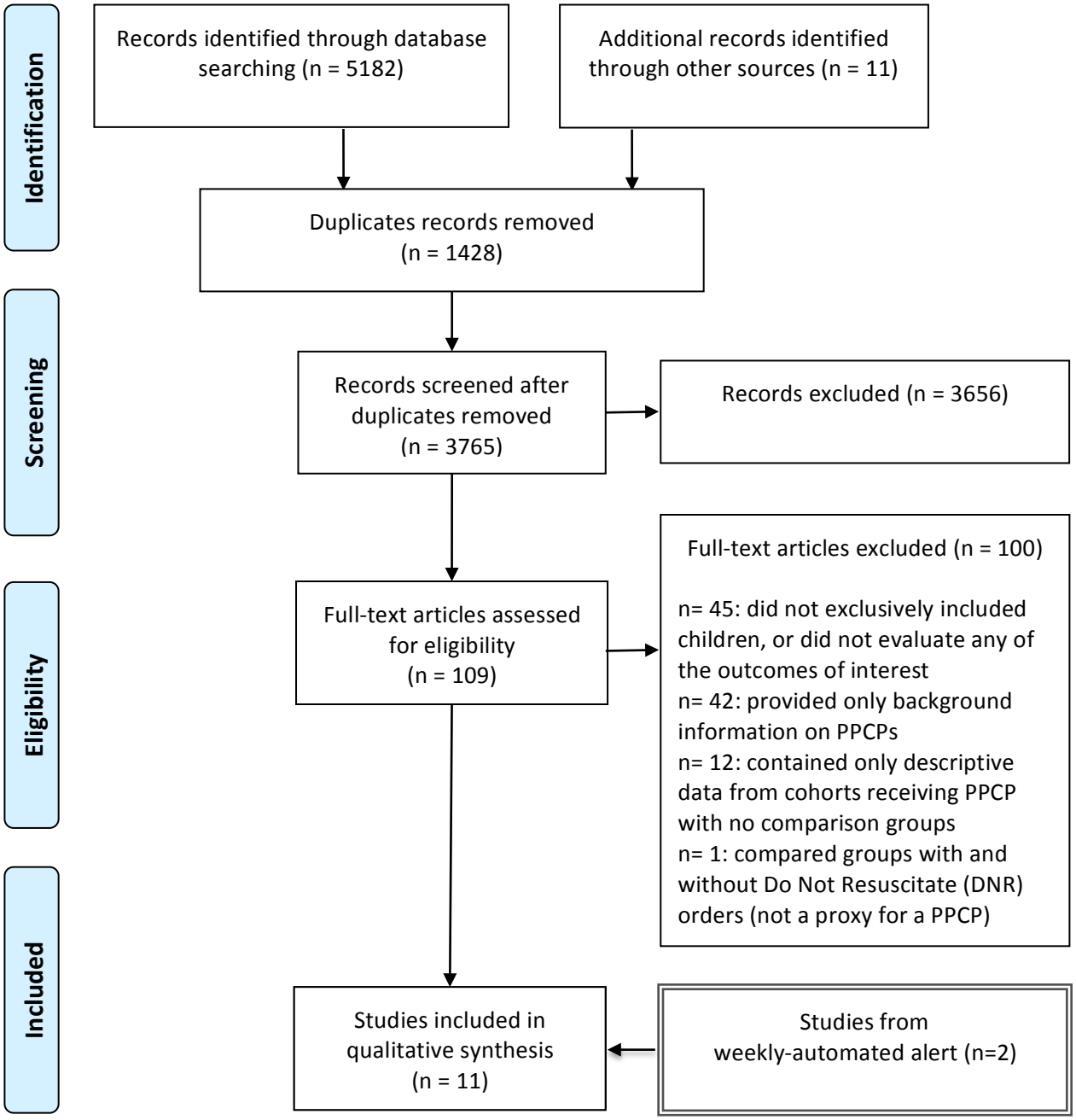
due to extensive heterogeneity in study population, outcome measurement, observation period and reporting of program components. To assess risk of bias an instrument from the Cochrane Handbook Chapter on non-randomized studies was adapted.[39] It describes the features of studies that might offer an increased risk of bias based on study type and other potential limitations. Then, the studies were ranked according to the Newcastle-Ottawa Scale (NOS)[40] for observational studies. The NOS tool assigns stars on features that may increase bias. The greater the number of stars, the higher the quality of the article (to a maximum of 9 stars). Details are described in Appendices C and D.

2.3 Results

2.3.1 Study Selection

As demonstrated in figure 3, the search strategy retrieved 5,193 references (including 1428 duplicates), with 109 reviewed at the full-text level, and 9 fulfilled the criteria for inclusion[12,21,41-47] [kappa statistic $\kappa = 0.83$, 95% CI 0.64–1.00, $p < 0.001$, indicating almost perfect agreement according to Fleiss[48]]. The primary reasons for exclusion during full text review was that the study population did not exclusively constitute children, or evaluated neither of the outcomes of interest (45 articles). Other studies were excluded for providing background information about PPCP but no evaluation (42 articles), or presented descriptive data from cohorts receiving palliative care with no comparison group (12 articles). Comparison of groups who did or did not have a Do Not Resuscitate (DNR) order (1 article) was not considered a proxy for a palliative care program. After the initial search 2 additional studies were found through the weekly-automated updates.[49,50]

Figure 3. Flow Diagram of Study Selection



2.3.2 Study Characteristics

2.3.2.1 Description of Studies

A total of 11 retrospective, observational studies were included. No RCTs were found. Full descriptive characteristics are presented in Appendix B and summarized in Table 2.

A total of 4 articles were originally classified as cohort comparisons[21,43-45] and used administrative data and survey techniques[43] to explore the differences between groups of children who had access to PPCP and those who did not. The observation period varied from the last day of admission before death to the entire period from referral to a PPCP and death. Of those, 2 articles exclusively included children who died from cancer,[43,44] and the others included children who died from any cause.[21,45] The interventions by which children were classified as being in the PPCP group were: use of a PPCP (program or hospice provider[21,44]), billing for inpatient PC consultation,[45] and parental planning of child's location of death (LOD),[43] part of an important element of PPCP called Advance Care Planning. The studies were based on data from the US [21,43,45] and UK.[44]

A total of 4 articles were originally classified as pre-post studies[12,41,46,49] and used administrative data[12,49] and chart review[41,46] to explore the differences in outcomes before and after access to PPCP. Of those, 3 studies compared the outcomes within the same group of patients before and after enrollment in a PPCP, and included children with all types of LTC's.[12,46,49] The last study compared historical cohorts of children with brain tumors from before and after the implementation of a standardized program for end-of-life (EOL) care.[41] This article could be classified as a historical cohort comparison, and the intervention as the coordination of care, since in both periods children had access to hospice providers but without the standardization of the care plan implemented by the program. The observation period varied from 12 months to 10 years. Determining the time point at which children entered the program was based on hospice use,[46]

explicit enrollment in the program,[12,49] or date of the implementation of the program.[41] The studies were based on data from the US[12,41,49] and Canada.[46]

An article originally classified as a 'case-control' is actually a cohort comparison.[47] This study from the US relied on administrative data to compare those enrolled in the PPCP with those who were not, for the 6-month period before death.

A conference abstract[50] is a cohort comparison combined with a pre-post analysis in the group who received PPC. This study was based on an administrative database in the US, and identified those in the PPCP group by the presence of a PC consultation, with a 2-year observation period.

The last article is a case-series[42] from the US, which compared costs of procedures being carried in a home-based palliative care program to those done in hospital. This included just 3 patients, and described procedures for 1 day of care.

2.3.2.2 Risk of Bias and Quality Assessment

Studies were classified as having a moderate to high risk of bias, with 5 articles receiving 5-6 stars, and 6 articles classified ≤ 4 stars. This was possibly attributed to their observational design, and the nature of the intervention, but also to the limitation of the NOS tool's ability to assess study designs other than cohorts and case-controls. Details on Appendix D and visual summary described in Table 3.

Observational studies rely on secondary analysis of administrative databases and medical reports not collected for the research purposes. Consequently, they may be incomplete or represent only one perspective (may not include all aspects of health care related costs, e.g. out of pocket expenses, uninsured drugs/procedures, alternative therapies, or full record of admissions

across providers). Access to certain content may be limited (e.g. clinical outcomes and severity of disease may not be pre-defined in records) and may not apply to the general populations (e.g. selection bias from insurance coverage/eligibility).

While many PPCP do not require referral by a physician (i.e. families can self-refer at many programs), families must accept enrollment. Enrollment can result in differential exposure to Advance Care Planning - Advance Directive conversations, and to teams with expertise in symptom management. This may cause groups to be systematically different, creating an imbalance between children in a PPCP to those under usual care, invariably affecting cost and utilization. Given that curative treatments in this population are often invasive and intensive, one would expect this bias to cause the usual care to be more resource consuming and have higher health care costs.

Information bias may have affected the allocation of patients to intervention groups for patients who were defined as "recipients" of palliative care through claims, bills or service codes. As pediatric palliative care is a relatively new and evolving specialty, the mode of recording services may have changed over time. Consequently, utilization of PC services could have been underreported or reported differently from usual care, for while procedures and practitioners are comparable, the goals of care vary (i.e. curative vs. enhancement of quality of life).

Further, history and maturation is likely to play some threat to validity as well, since the evolving nature of PPCP specialty practitioners in both PPCP and usual care may provide either type of care to children in both groups. These health care professionals might adapt their practice, either increasing curative efforts for children under the PPCP or focusing on palliative care for those in the usual care group.

Only 2 articles addressed confounding in the outcome comparison between groups. [44,49] A cohort comparison study[44] investigated the effect on the number of hospital admissions in the cohort in a pediatric hospice, from time of referral to death. Age, disease, gender and Townsend deprivation category were controlled for. The deprivation score[51] is based on unemployment, non-car ownership, non-home ownership and household overcrowding. The authors failed to address survival bias (whether the length of the study period was similar between groups) or include number of hospice admissions for the PPCP group. Therefore, the number of admissions for this group must be interpreted with caution due to shifts in admission setting rather than decreases in health care utilization. A pre-post study[49] addressed confounding by controlling for time exposed to PPCP, but did not include a control group for comparison, making it unclear whether the observed decrease in LOS and costs in the PPCP period was a consequence of the program or a natural trend among patients approaching death. Additionally, place of death and costs associated with home care were not controlled for, both of which can bias results. The remaining studies did not address confounding.

In regards to the NOS tool, it is necessary to mention that some of its questions are not applicable to other study designs beyond cohort and case-controls. Following are some difficulties encountered in applying them to the articles included in this review. For instance, within section "Selection", the question "Demonstration that outcome of interest was not present at start of study": given all the studies were retrospective and used administrative databases, the negative answer would apply to all the studies in this question. Likewise, the question "Selection of the non intervention cohort" will not apply to the studies before-and-after with no control, for not having a non-intervention group. Other critiques to the validity of the tool has been published[52] but at the moment it is still recommended by Cochrane[39] as a user friendly tool.

2.3.3 Effects of Pediatric Palliative Care Program on Outcomes of Interest

Table 3 presents a visual summary of the results from published articles and presented abstracts, ranked by quality assessment, with results aggregated by outcome measurement and study quality.

Table 2. Characteristics of the Studies

Article	Participants	Study design	Observation period	n	Intervention Group	Comparator	Outcomes
Postier et al 2014	1 to 21 years Enrolled in the home PPCP/ hospice program	RChBA **	Before: 1 year After: 1 year	425	Pre-PPCP	Post-PPCP	Change in number of hospitalizations, LOS, and total billed charges for hospital/ER stay
Fraser et al 2013	0-19 years Died from cancer	RCS	Referral to death	497	Hospice Group (n=132)	Control group (n = 311)	Total number of hospital admissions, number of planned hospital admissions, number of emergency admissions
Keele et al 2013	<18 years Died from all causes of death, at the hospital, >5 days after admission	RCS	Last admission before death	24342	PC group (n=919)	No PC (n=23423)	Age, gender, LOS, major group category diagnostic, medications, procedures in the last admission
Arland et al 2013	1 month - 19 years Died from brain tumor	RChBA #	Before: 5 years After: 10 years	114	After group (n= 92)	Before Group (n= 22)	Symptoms, hospitalizations (number, LOS), location of death
Smith et al 2013	Children (no age bracket defined) in the 10% most costly discharged patients	RCS + RChBA **	RSC: up to 2 years RChBA: undisclosed	1001	PPCP Group (n=81):	Control Group (n= 920):	Cost, demographics, use of technology
Gans et al 2012	0 to 20 years Living with a LTC Enrolled in the PPCP	RChBA **	Before: 12 months? After: 18 months	123	After PPCP	Before PPCP	LOS, medical expenditures, family's quality of life and satisfaction
Pascuet et al 2010	Children (no age bracket defined) Used 'Respite' at the pediatric Hospice	RChBA **	Before: 1 year After: 1 year	66	Before respite	After respite	LOS, ER and Outpatient visits, overall cost in hospital/hospice admission
Dussel et al 2009	Children (no age bracket defined) Died from cancer	XS survey RCS	Last month of life	140	Planned LOD (n=88)	Did not plan LOD (n=52)	EOL planning, EOL support from physicians, use of home care, hospital resources utilization, place of death
Knapp et al 2009	1-21 years Died from all causes of death	RCS	Last year of life	1527	Hospice use (n= 85)	Non-hospice use (n= 848)	Hospice use, hospice expenditures, other expenditures
Ward-Smith et al 2008	Children (no age bracket defined) Enrolled in the PPCP	RCS *	6 months before death	18	PPCP group (n=9)	Non PPCP (n=9)	Total hospital costs, LOS, differences in types of procedures
Belasco et al 2000	Children (no age bracket defined) Referred to a home PPCP	Case Series	1 day	3	Home care	Hospital care	Type of interventions delivered, place of death, comparison of charges of care

PPCP: pediatric palliative care program; RChBA: Retrospective cohort before-and-after study with no control; LOS: length of stay; ER: emergency room; RCS: Retrospective Cohort study; PC: palliative care; LTC: life-threatening conditions; XS: cross-sectional; LOD: location of death; EOL: end-of-life; * described by authors as a case-control but technically it is a cohort comparison; ** study design where the same patients are followed before and after the intervention with no controls; # Study design where different cohorts of patients are followed before and after the intervention being implemented (historical cohorts)

Table 3. Visual Summary of Results and NOS Quality Assessment

Author	Design	Outcomes			Newcastle-Ottawa Scale		
		Hospital Admissions	LOS	Cost	Selection	Comparability	Outcome
**Postier et al 2014	RChBA **	?	+	+	★ ★	★	★ ★ ★
Fraser et al 2013	RCS	?			★ ★ ★	★	★ ★
Knapp et al 2009	RCS			–	★ ★ ★	★	★ ★
Keele L et at 2013	RCS	+		+	★ ★ ★	★	★
# Arland et al 2013	RChBA #	+	+		★ ★ ★		★ ★
Smith et al 2013	RCS + RChBA **	?		–	★ ★		★ ★
Dussel et al 2009	XS survey + RCS	?	?		★ ★ ★		★
* Ward-Smith et al 2008	RCS *		?	?	★ ★		★ ★
** Gans et al 2012	RChBA **		+	+	★	★	★
** Pascuet et al 2010	RChBA **	?	–	+	★ ★		★
Belasco et al 2000	Case series			+	★		★

LOS: length of stay; RChBA: Retrospective cohort before-and-after study with no control; RCS: Retrospective Cohort study; XS: cross-sectional; * Described by authors as a case-control but technically it is a cohort comparison; ** Study design where the same patients are followed before and after the intervention with no controls; # Study design where different cohorts of patients are followed before and after the intervention being implemented (historical cohort comparison)

? No difference / Controversial + Decrease – Increase

2.3.3.1 Hospital Admissions

In total 7 articles investigated hospital admissions using different approaches.[41,43-46,49,50] Four of the articles reported the **proportion of patients admitted to hospital** (number of patients).[41,43,45,50] The remaining 3 articles reported the **number of hospital admissions** (number of events).[44,46,49]

Among the PPCP recipients, a decreased **proportion of patients with hospital admissions** was demonstrated.[41,43] Dussel et al[43] showed that a lower proportion of children with cancer, from families who had planned LOD, were admitted to the hospital in the last month of life, compared to those who did not (54% vs. 98%, p <0.001). This decrease was driven by the fact that fewer families who planned LOD chose a hospital death (28% vs. 75%, p<0.001). Arland et al[41] observed the proportion of patients admitted to the hospital after the implementation of a program for EOL care for children with brain tumors. The intervention provided a coordinated care plan. In both periods (before and after the program) children had access to the same providers, including hospices. This study showed a decrease in the proportion of patients being admitted to hospital in a 10-year period after the implementation of the program (54% vs. 29%, p<0.05). On the other hand, no effect in the **proportion of children having emergency room visits** in the last week of life was found,[43] and findings regarding the proportion of patients **using critical care** were conflicting.[45,50] Smith et al[50] investigated the receipt of PPCP among the most costly patients in a tertiary care system and the proportion of admissions admitted to critical care units (ICU). In 1 year of observation period, they found a greater proportion of PPCP users having pediatric intensive care unit (PICU) admissions (90% vs. 56%, p<0.001), and a smaller proportion of neonatal intensive care unit (NICU) admissions (17% vs. 28%, p=0.04). Keele et al[45] reported on the likelihood of being admitted to the intensive care unit (ICU) in the last admission before death. It showed that among

children who died from any cause, those who had had palliative care services were less likely to be admitted to the ICU (RR 0.29, CI 95% 0.26–0.32). Excluding the 2 articles with the lowest quality assessment (≤ 4 stars) seemed to favour PPCP.

The **effects of PPCP on the number of hospital admissions** were investigated in 3 studies.[44,46,49] Within the same article one finds different directions of the effect on number admission depending on the type of admission. No difference in *overall* admissions and emergency visits between PPCP users and controls cohorts,[44] or comparing before and after implementation of PPCP[46,49] was found. Among cancer patients, a decrease in planned hospital admissions under a PPCP (IRR 0.60, CI 95% 0.43-0.85, p=0.004),[44] and decrease in outpatients visits after the implementation of PPCP was observed.[46] There was an increase in number of hospital admissions when longer exposure to PPCP was found,[49] which is expected for longer survival period. Excluding the article with the lowest quality assessments did not change the results. Table 4 shows a summary of the results.

Table 4. Summary of Comparison of Hospital Admissions

Author	Design	NOS	VS	Proportion of Patients with Admissions
# Arland et al 2013	RChBA #	★★★★★		**Admissions to hospital** Before program (5 year): 54% vs After program (10 year): 29% (p< .05) 46% fewer hospital admissions.
Dussel et al 2009	XS survey + RCS	★★★★		**Admission to hospital** Planned LOD: 54% (47/87) vs. Did not plan: 98% (51/52), p <0.001 **Admission to emergency room** Planned LOD: 6% (5/84) vs. Did not plan: 16% (8/50), p= 0.057
Keele L et at 2013	RCS	★★★★★		**Admission to ICU – Relative Risk (95%CI)** Lower among PPCP group: RR 0.29 (0.26–0.32)
Smith et al 2013	RCS + RChBA **	★★★★		**Admissions to PICU** PPCP group: 90% (73/93) vs. Control group: 56% (522/920), p<0.001 **Admissions to NICU** PPCP group: 17% (14/93) vs. Control group: 28% (262/920), p=0.04
Author	**Design**	**NOS**	**VS**	**Number of Admissions**
**Postier et al 2014	RChBA **	★★★★★★		**Average number of hospital admissions** Pre-PPCP: 3.09±3.6 vs. Post-PPCP 3.18±4.3, p = 0.538 Interaction - level of PPCP exposure, Cancer or not, study period (p < 0.001), adjusting for other demographic and clinical characteristics.
Fraser et al 2013	RCS	★★★★★★		**Total hospital admissions** (controlled for age, disease, gender, deprivation category) IRR 0.79 (CI 95% 0.59-1.05), p=0.10 **Planned hospital admissions** IRR 0.60 (CI 95% 0.43-0.85), p=0.004 **Emergency hospital admissions** IRR 1.15 (CI 95% 0.84-1.58), p=0.375
** Pascuet et al 2010	RChBA **	★★★		**Median number of ER visits/monthly** After - 0.03 (95% CI, -0.09 to 0.02, p=0.20) **Median number of Outpatients visits/month** After: - 0.5 (95% CI -1.0 to - 0.05, p=0.029)

NOS: Newcastle-Ottawa Scale; VS: Visual Summary; RChBA: Retrospective cohort before-and-after study with no control; XS: cross-sectional; RCS: Retrospective Cohort study; LOD: location of death; ICU: intensive care unit; RR: relative risk; PICU: pediatric intensive care unit; PPCP: pediatric palliative care program; NICU: neonatal intensive care unit; p: p-value; IRR: incidence rate ratio; ER: emergency room; CI: confidence interval; * Described by authors as a case-control but technically it is a cohort comparison; ** Study design where the same patients are followed before and after the intervention with no controls; # Study design where different cohorts of patients are followed before and after the intervention being implemented (historical cohort comparison)

 No difference / Controversial  Decrease  Increase

2.3.3.2 Length of Stay (LOS)

In total 6 studies investigated the influence of PPCP on days spent in hospital or LOS.[12,41,43,46,47,49] The majority of studies showed a decrease in **hospital LOS** after the implementation of PPCP[12,41,46,49] and between PPCP users and controls.[43] Arland et al[41] looked at the EOL period in children with brain tumors, finding a 25% decrease in the mean LOS per hospital admission and a 66% decrease in LOS per patient after the implementation of a PPCP. Gans et al[12] found a decrease of 1.2 days in mean LOS in hospital per patient per month after the enrollment in a community-based PPCP. Postier et al[49] investigated changes in the mean LOS in hospital after children enroll in a home-based PPC/hospice program and showed interaction between time of exposure to the PPCP and disease, demonstrating that non-cancer patients with at least 6 months of enrollment in the PPCP had a significant decrease in total LOS by an average of 38 days. Dussel et al[43] showed a trend in lower median LOS in hospital during the last month of life, among children with cancer who planned LOD (17 days vs. 21 days, respectively, p <0.494). Only 1 study found no difference between groups. Ward-Smith et al[47] compared children enrolled in a PPC program for at least 6 months with those not enrolled, and did not find any difference in mean LOS in the last 6 months before death**.** However, statistical significance was either not reached or tested in some articles.[12,41,43,47]

The only study that explicitly included **combined LOS in hospital and hospice** found an overall increase in LOS. Pascuet et al[46] used a more complete approach measuring hospital and hospice admissions and found an increase in median number of total inpatient days per month after the enrollment of children in a PPCP carried by a hospice provider (variation: 0.9 days, p=0.013). The decrease in the median LOS in hospital per month after the enrollment (- 2.9 days, CI 95% -4.5 to -1.3 days, p=0.001) was compensated by an increase in the median LOS in hospice per

month (2.4 days, min 0.08- max 26.5 days). This result shows a shift in health care setting other than a decrease in health care resource utilization, and the effects on cost of health care will be presented.

Excluding the 3 articles without statistical treatments[12,41,47] resulted in complete discordance between studies. Table 5 shows a summary of the results.

Table 5. Summary of Comparison of LOS

Author	Design	NOS	VS	Outcome Report
**Postier et al 2014	RChBA **	★★★★★★		**Mean total LOS** Pre-PPCP: 34.09±59.7 days vs. Post-PPCP: 19.37±34.0 days (p < 0.001). Interaction - level of PPCP exposure, Non-cancer, and study period (p < 0.001).
# Arland et al 2013	RChBA #	★★★★★		**Average LOS per hospital admission** After group: 3.03 days vs. Before group: 4.05 days Decrease 25%. No test applied **Average LOS per patient in the total group (not only among those admitted to the hospital)** After group: 1.25 days/patient vs. Before: 3.68 days/patient Decreased 66%. No test applied
Dussel et al 2009	XS survey + RCS	★★★★		**Median (IQR) LOS in days - last month of life** Planned LOD: 17 (4-27) Did not plan LOD 21 (6-28), p=0.494
* Ward-Smith et al 2008	RCS *	★★★★		**Mean LOS (min-max)** PPPC: 4 days (5 to 17 days) Non-PPPC: 4 days (5 to 18 days)
** Gans et al 2012	RChBA **	★★★		**Average number of days spent in the hospital (per member, per month)** Before program: 4.0 vs. After program: 2.8 Reduction of 32%. No test applied.
** Pascuet et al 2010	RChBA **	★★★		**Median number of hospital Inpatient days/month** After: - 2.9 (95% CI -4.5 to -1.3, p=0.001). **Median number of hospice days/month (min-max)** After: 2.4 (0.08-26.5) **Median number of Total Inpatient days /month** After: Variation: 0.9 (p=0.013).

LOS: length of stay; NOS: Newcastle-Ottawa Scale; VS: Visual Summary; RChBA: Retrospective cohort before-and-after study with no control; PPCP: pediatric palliative care program; XS: cross-sectional; RCS: Retrospective Cohort study; IQR: interquartile range; LOD: location of death; CI: confidence interval; * Described by authors as a case-control but technically it is a cohort comparison; ** Study design where the same patients are followed before and after the intervention with no controls; # Study design where different cohorts of patients are followed before and after the intervention being implemented (historical cohort comparison).

 No difference / Controversial  Decrease  Increase

2.3.3.3 Health Care Costs

In total 8 studies measured the influence of PPCP on health care costs[12,21,42,45-47,49,50] with summary of results in Table 6. It is unclear whether costs of health care differ when studies refer to them as charges, expenditures or costs. Studies were heterogeneous and had conflicting results. Reduced costs were found in 6 articles after the implementation of PPCP[12,46,49], and among PPCP users compared to their controls.[42,45,47]

Gans et al[12] measured **health care cost changes after the enrollment** in a community-based PPCP. This study found a shift in the health care resources utilization for those using PPC, with increased outpatients care costs (34%) and pharmaceutical costs (35%), and decreased inpatient care costs (- 35%). In all, enrollment in the PPCP resulted in 11% fewer mean health care costs compared to usual care, though this difference was not investigated statistically. Similarly, Pascuet et al[46] showed a decrease in costs with the shift in health care resources utilization after the enrollment of children in a PPCP carried by a hospice provider. The study demonstrates that although an increase in LOS was observed when considering days in hospital and days in hospice as inpatient days, **the total cost of inpatient care per month** decreased significantly (- $4,252/month, 95% CI -$7,551 to - $953, p=0.012). This decrease was attributed to the difference in average cost per day between providers (hospital inpatient day: $2,007 vs. hospice day: $500). Postier et al[49] found a decrease **in total hospital charges** after children enrolled in a home-based PPC/hospice program. This decrease was dependent on the disease category and the amount of exposure to the program. A significant reduction in charges, nearly $275,000 ($p < 0.001$), was observed over 12 months observation period for non-cancer patients with at least 6 months of PPCP exposure. Keele et al[45] measured **average daily charges** in the last admission before death. It showed that among children who died from any cause of death, those who had had palliative care

services had lower daily charges than those without PC consultation (PC code: $9348 vs. No PC codes: $11 806, p<0.001). Ward-Smith et al[47] compared children enrolled in a PPCP for at least 6 months with those not enrolled, and found a slight difference in **mean total hospital costs in the last 6 months before death.** The authors found a difference in the types of cost between the groups, with the PC group having more pharmacy costs and the non-PC group more radiology service costs. Belasco et al[42] observed children referred to a home based PPCP and compared the **cost of equivalent care in the hospital to home care**. The authors listed the procedures/services children were receiving at home and estimated the equivalent costs having them access the same procedures/services at the tertiary care provider. The costs were discrepant, however, for the home care cost estimates, the authors did not include costs of procedures of uninsured procedures.

Opposite results were demonstrated in 2 articles with increased costs among PPCP users compared to controls,[21,50] or no difference in costs after the implementation of PPCP.[50]

Knapp et al[21] looked at children who died from all causes of death and compared the **expenditure patterns of hospice users and non-hospice users in the last year of life.** Overall, the hospice users had higher expenditures in all types of admissions (hospice, inpatient, outpatient, emergency department) and pharmacy expenses. Likewise, subgroup analysis by cause of death (perinatal, chronic, external and other) found the same results: hospice users incurred higher expenditures. Smith et al[50] looked into the most costly patients with LTC's in a tertiary care provider for 2 years. Those who received PPC consultation presented significantly higher **total costs** ($177K vs. $103K, p<0.001) **and daily costs** ($3.8K vs. $3.4K, p=0.001) compared to those who did not. The findings remained in the subgroup that died within the study period. However, within the

PPC users no increase in the daily cost after the initial PPC consultation was confirmed (Before PPC: $3827 vs. After PPC: $4013, p=0.06).

However, most authors did not test for significance.[12,21,42,47] Excluding these articles and the studies with the lowest quality assessment[12,42,46] did not impact the discrepancy of the findings.

The 2 studies with comprehensive outcome measures across hospice and hospital expenditures had conflicting results with regard to the direction of effects: a decrease in costs in the Canadian health system context[46] and an increase in the US context.[21]

Table 6. Summary of Cost Comparison

Author	Design	NOS	VS	Outcome Report
**Postier et al 2014	RChBA **	★★★★★★		**Average charges (factor of 10,000)** Pre-PPCP: 20.97± 43.3 vs. Post-PPCP 10.91± 21 (p < 0.001) Interaction - level of PPCP exposure, Cancer/non-cancer, study period (p < 0.001).
Knapp et al 2009	RCS	★★★★★★		**Mean Expenditures** ***Hospice users*** **Inpatient** **Outpatient** **ER** **Pharmacy** **Hospice** **Total §** All causes of death $49,621 $14,414 $873 $7,449 $11,36 $83,719 Perinatal $65,814 $23,058 $971 $10,946 $11,934 $112,723 Chronic $50,283 $14,366 $929 $7,582 $10,887 $84,047 ***Non-Hospice users*** All causes of death $19,968 $12,954 $468 $3,207 $36,597 Perinatal $35,770 $35,796 $832 $5,820 $78,218 Chronic $38,232 $21,603 $632 $6,117 $66,584 **§** Total cost not presented in the primary article but added the individual mean expenditures.
Keele L et at 2013	RCS	★★★★		**Average daily charges (adjusted for geography)** PPCP group: $9348 ($6070–$15,318) vs. No PPCP group: $11 806 ($8017–$18352), p<0.001
Smith et al 2013	RCS + RChBA **	★★★★		**Change in median daily cost (IQR) - Within PPCP group** Before PPCP: $3827 ($2.9-3.9K) vs. After PPCP: $4013 ($3.1-5.1K), p=0.06. **Daily Cost 2010/2011** PPCP group: $3.8K ($3.1K-$4.7K) vs. Control: $3.4K ($2.7K-$4.5K), p=0.001
* Ward-Smith et al 2008	RCS *	★★★★		**Mean hospital costs (min-max) - Excluding surgical charges** PPCP: $78,780 ($33,283 to $130,970) vs. Non-PPCP: $81,780 ($28,970 to $135,432)
** Gans et al 2012	RChBA **	★★★		**Average Total Medical Expenditures (per member, per month)** Before program: $15,653 vs. After program: $13,976 Overall decrease 11% - nearly $1 million in 18 months [Inpatient care: decrease 35% ($3,571); Outpatient Care: increase 34% ($1,398); Pharmaceutical costs: increase 35% ($495)]
** Pascuet et al 2010	RChBA **	★★★		**Change in total inpatient cost/month (Hospital + Hospice)** After: - $4,252/month (95% CI, - $953 to -$7,551, p=0.012). **Unit costs/day:** Hospital Inpatient day: $2,007 vs. Hospice day: $500
Belasco et al 2000	Case series	★★★		**Average charges per day** AML patient (toddler): Hospital: $4,283 vs. Home: $17 Neuroblastoma patient (pre-school child): Hospital: $2,300 vs. Home: $325 Multiple chronic diseases patient (teenager): Hospital: $8,258 vs. Home: $1,308

NOS: Newcastle-Ottawa Scale; VS: Visual Summary; RChBA: Retrospective cohort before-and-after study with no control; PPCP: pediatric palliative care program; RCS: Retrospective Cohort study; IQR: interquartile range; AML: Acute myeloid leukemia; CI: confidence interval. * Described by authors as a case-control but technically it is a cohort comparison; ** Study design where the same patients are followed before and after the intervention with no controls; # Study design where different cohorts of patients are followed before and after the intervention being implemented (historical cohort comparison)

 No difference / Controversial  Decrease  Increase

2.3.3.4 LOS in the Last Admission Before Death

Only 1 study was found comparing LOS in the last admission before death.[45] The authors reported a shorter median LOS among children who died from any cause of death and had access to palliative care services (PC code: 17 days [IQR 9–36] vs. No PC code: 21 days [10–47], p<0.001).

2.3.3.5 Invasive Procedures to Prolong Life in the Last Admission

Only 2 cohort studies observed invasive procedures in the last admission before death. The first[45] demonstrated that among children who died from any cause, those who had a PC consultation had a significantly lower **relative risk of receiving some procedures.** These included invasive mechanical ventilation (RR 0.14, 95% CI 0.12–0.16), adrenergic (RR 0.15, 95% CI 0.11–0.2), sedatives (RR 0.25, 95% CI 0.2–0.3) and analgesics (RR 0.53, 95% CI 0.47–0.6). These children also had a higher probability of accessing noninvasive mechanical ventilation (RR 1.6, 95% CI 1.3–1.9) and intracranial pressure monitoring or an extra ventricular device (RR 2.8, 95% CI1.6–5.0). The second study[43] reported a lower **proportion** of intubations in the last 24 hours of life (21% vs. 48%, p= 0.029) and no difference in the proportion of withdrawal of other support measures (36% vs. 19%, p= 0.123), among cancer patients with planned LOD compared to those without one.

2.3.3.6 Resuscitation Attempts

Only 2 cohort studies examined resuscitation interventions. The first[45] showed that among all causes of death, those who had a PC consultation had a significantly lower **relative risk** of cardioversion (RR 0.49, 95% CI 0.38–0.62) than those who did not. The second[43] reported a lower **proportion** of cardiopulmonary resuscitation attempts (CPR) among cancer patients with a planned LOD (4% vs. 19%, p = 0.142), compared to those without.

2.4 Discussion

2.4.1 Summary of Main Results and Limitations

Table 3 presents a visual summary of the results from published articles and presented abstracts, ranked by quality assessment, with results aggregated by outcome measurement and study quality.

Among the PPCP recipients, a decreased **proportion of patients with hospital admissions** was demonstrated.[41,43] While no effect in the number of patients using emergency admissions was found,[43] findings on the proportion of patients using critical care were conflicting.[45,50] Excluding the articles with the lowest quality assessment seemed to favour PPCP. With respect to **number of hospital admissions**, no difference in overall admissions and emergency visits between PPCP users and controls,[44] nor after implementation of PPCP[46,49] were found. Among cancer patients, a decrease in planned hospital admissions under a PPCP,[44] and an increase in number of hospital admissions with longer exposure to PPCP was found,[49] which is a reflection of survival. Number of outpatients visits decreased after the implementation of PPCP.[46] Excluding the articles with the lowest quality assessments did not change the results.

In terms of **LOS,** most studies demonstrated shorter hospital stays after the implementation of PPCP[12,41,46,49] and between PPCP users and controls,[43] until inpatient time in hospice facilities was considered[46] and LOS became longer for PPCP patients. This result shows a shift in health care setting other than a decrease in health care resource utilization. However, statistical significance was either not reached or tested in some articles.[12,41,43,47] Excluding the articles without statistical treatments resulted in complete discordance between studies.

Regarding **costs of health care**, no conclusive impact of PPCP can be drawn from the

primary studies[12,21,42,45-47,49,50] due to conflicting results and heterogeneity in comprehensiveness of outcome measures. Reduced costs were found in 6 articles after the implementation of PPCP,[12,46,49] and among PPCP users compared to their controls.[42,45,47] Opposite results with increased costs among PPCP users compared to controls was observed in 2 articles,[21,50] or no difference in costs after the implementation of PPCP.[50] However most of these articles focused on costs of hospital admissions not accounting for costs in different settings. From the more comprehensive studies that included hospice and hospital expenditures, an overall cost decrease in the Canadian health system context,[46] attributed to lower costs of the hospice settings, and increase in the American one, was observed.[21] However, most authors failed to test for significance,[12,21,42,47] and excluding these articles or those with the lowest quality assessment[12,42,46] did not impact the discrepancy of the findings.

For **EOL admissions**, PPCP users had shorter LOS[45] and less aggressive care (invasive procedures and CPR).[43,45] However, focusing solely on hospital utilization has limitations, as it does not consider impacts on the health care system in totality, nor the financial burden borne by families.

Interpreting the reviewed studies was challenging due to numerous limitations of the primary articles, and study contexts. For instance, Keele et al[45] only included children who died at least 5 days after admission, therefore excluding individuals who did not choose life-extending measures to prolong their stay beyond 5 days, or those who were discharged to die at home. This definition would affect both hospital admissions and costs, leaving the true difference between programs to be greater. Further, the PPCP children were allocated based on billing codes by the 'International Statistical Classification of Diseases and Related Health Problems' (ICD) codes for palliative care (V66.7). If the services were billed under the ICD code for the primary condition or

disease, it would misclassify children under PPCP to the usual care group. The direction and magnitude of this bias is uncertain. As well, Fraser et al[44] did not measure and/or control whether children were still in disease-directed treatment in both groups - a factor for decreased planned admissions. Also, it is not clear whether symptom management admissions to the hospice were included in the overall number of admissions, making it unclear whether the results represent a shift in health care setting or a decrease in resource utilization. Smith et al's conference abstract[50] did not present sufficient information on methodology. Therefore, questions remain regarding risk of bias, selection and identification of participants, and intervention classification. Most studies focused on hospital admissions and did not account for other types of resource utilization. Studies that demonstrated a decrease in LOS among PPCP users[12,41,47,49] did not investigate the number of days spent in hospice or other facilities, which again, leaves uncertainty around shift in health care setting. The only study that measured both hospital and hospice admissions found an increase in the total combined LOS, demonstrating a shift in the setting of health care utilization from hospital to hospice.[46] Despite this shift, a significant decrease in monthly costs was observed, owing to the difference in average daily costs between settings in the Canadian health system. A similar trend was shown in the US by Gans et al,[12] who demonstrated a shift in resource utilization, from a decline in inpatient care costs to a surge in outpatient care and pharmaceutical costs. Nonetheless, an overall 11% decrease in health care costs after the implementation of the community-based PPCP occurred. However, this study did not test for statistical significance nor adjusted for survival time after program enrollment. Conversely, another American study[21] found hospice users to have higher expenditures in all types of admissions (hospice, inpatient, outpatient, emergency department) and pharmacy expenses. This study allocated patients to the PPCP group based on billing codes for hospice services. However, some patients in the non-hospice user group died at

the hospice, demonstrating the limitation of using billings to identify patients. Another important limitation was observed in Belasco et al[42] where the authors listed the procedures/services received at home and estimated the equivalent cost at the tertiary care centre. Costs were discrepant, however, for home care, uninsured procedures were not accounted for in cost estimates. It is unclear whether those costs were out-of-pocket for the families or waived by the health care providers. Therefore, charges did not appropriately reflect costs, introducing important measurement bias. It is important to mention that it is unclear whether costs of health care differ when studies referred to it as charges, expenditures or costs.

2.4.2 Overall Completeness and Applicability of Evidence

It is worth noting the considerable heterogeneity in outcome measures, observation period, scope of PPCP elements, characteristics, reporting of program components, and whether reported interventions accurately represented the enrollment of children and families in a PPCP. Moreover, the specific context and funding models for the different health systems may have affected results. Therefore, both the evidence and its applicability should be interpreted with caution.

2.4.3 Quality of the Evidence

According to the NOS tool, the overall quality of evidence is moderate to low (Table 3). In particular, the risk of selection bias to PPCP (given the nature of the referral process) is a major concern. Furthermore, information bias and misclassification is a threat to internal validity in observational studies based on secondary databases.

2.4.4 Potential Biases in the Review Process

A thorough search of the literature was performed without any language restrictions but no RCTs or prospective studies were found. Only comparative studies of palliative care against usual care were included. No indirect comparison was contemplated with single-arm observational studies. While the majority of studies found with the search strategy did not relate directly to PPC, some were studies in the PPC field but did not report the outcomes of this review, or were qualitative research or processes evaluation in the development of the PPC science, not subject to this systematic review.

2.4.5 Agreements and Disagreements with Other Studies or Reviews

This is the first systematic review on this topic. While the general interpretation of the results tends to show PPCP decreasing hospital resource consumption, more rigorous study designs with broader perspectives that include expected costs incurred in other settings should be undertaken. Focusing only on measurements of hospital admissions is limited, and does not consider the impact for the health care system in totality which it still responsible for funding other providers (hospices, community hospitals, home care providers). Also, the financial burden borne by families is unknown. Noting these limitations, this review provides an important first step towards a more comprehensive understanding of the impact of PPCPs on resource utilization across various health care settings.

2.5 Conclusion

2.5.1 Implications for Practice

The published evidence to support the planning and reallocation of resources for PPCPs by estimating its impact on the overall health care system has moderate to low methodological

quality.

The literature does, however, demonstrate that PPCPs result in no increase in hospital resource utilization and suggests a shift to other health care settings. Depending on the health care system, costs may increase, but at least in the Canadian context, one can argue that PCPPs can be cost saving. However, the paucity of evidence with broad approaches to measurement is not only in conflict, but very context dependent.

2.5.2 Implications for Research

Prospective studies are required to evaluate the overall impact of PPCPs on the health system from perspectives beyond that of the tertiary care provider, while measuring shifts in health care settings and family burden. Enhanced study designs can address the various aforementioned biases and classification issues. Standardization of outcome measures can enhance comparability and pooling of future research for increased power to better evaluate impact.

Chapter 3: Health Services Utilization: a Matched-Cohort Comparison

Overview

This chapter provides a comparative analysis of data from the BC PPCP and usual care on utilization at the hospital and hospice facilities. A matched-pairs cohort design was used to identify children in both groups, and a history of their utilization in both health care settings was retrieved.

3.1 Objective

- To compare health care utilization by children with LTC who died enrolled in the BC PPCP to that of BC children who died under usual care. The clinical question and outcomes are displayed in Table 7.

Table 7. Clinical Question and Outcomes

Population	Children who died in hospital (BCCH) or in hospice (CPCH) from a LTC
Cases (Intervention)	Children enrolled in PPCP provided by CPCH.
Controls (Comparison)	Children receiving usual care
Outcomes	*Primary outcomes:* • Number of outpatient and inpatient admissions of any type (emergency visits, general ward, critical care) in both settings (hospital and hospice) • LOS overall and per type of admission. *Secondary outcomes:* • LOS in the last hospitalization before death; • Utilization of invasive procedures to prolong life in the last admission (mechanical ventilation, vasoactive drugs and resuscitation attempt).

3.2 Methods

3.2.1 Study Design

A retrospective matched-cohort comparison of children who died from LTC, in hospice (cases) versus in hospital (controls) was designed. The matching criteria were disease code (ICD code 3-digit level) and age at death.

3.2.2 Definitions

Case: children enrolled in the BC PPCP who died between Jan-2008 and Dec-2012 from a LTC.

Control: children who died at BCCH between Jan-2008 and Dec-2012 and were never enrolled in the BC PPCP.

PPCP enrolled: children accepted by the PPCP multidisciplinary team as eligible to the program, AND parents or children (at the age of consent) had agreed to participate in the program.

PPCP NOT enrolled: children not included in the PPCP registry at the time of death, OR were not classified with LTC in the palliative care stage by the PPCP multidisciplinary team, OR parents or children (at the age of consent) had not agreed to participate in the program.

Critical care: admissions requiring critical care attention independent of the setting where it was provided. Admissions occurred at BCCH in the NICU or PICU, and those occurred at CPCH classified as levels of acuity 4-5. These levels of acuity are equivalent to admissions to the NICU/ PICU at BCCH, based on nursing workload and patients symptoms. CPCH team developed the Canuck Place Nursing Workload Measurement Tool and Acuity Scale that has been used for over 10 years, published in Siden et al supplemental material.[9]

3.2.3 Assumptions

I. All children died from a LTC as categorized in Table 1, and would have been eligible for PPCP, by definition, irrespective of referral.

II. Due to the lack of unique clinical or temporal markers to establish when enrollment in a PPPC should be initiated, one assumed that if 2 children died from the same disease at the same age, likely they would have had experienced similar disease trajectories and could be considered a match. This assumption does not hold for cancer patients where variations in numbers of relapses and remission periods must be considered. Consequently, the matching process was adapted for cancer patients.

III. BCCH and CPCH are the only pediatric hospital and hospice in BC. Accordingly, BCCH and CPCH were assumed to be the main providers of inpatient care for the study's population especially with regard to critical care, which is the most resource-intensive type of care towards EOL. Admissions to other community hospitals or facilities were assumed to be marginal to the study, with the exception of the critical care unit in Victoria General Hospital. This unit could, potentially, have admitted children before transferring them to the Mainland, and, therefore, children living in this hospital catchment area were excluded from the study to avoid risk of bias due to incomplete data.

IV. To increase comparability between matched children and eliminate the source of bias, the study's population was limited to those who died at BCCH or CPCH since no data was available from controls who died outside of BCCH (at home or other facilities); and children who died at home systematically had lower health care resources utilization and lower costs towards EOL.

3.2.4 Exclusion Criteria

i. Residents of Vancouver Island Health Authority catchment area were excluded, as they were able to access critical care on Vancouver Island (for which data were not available for this study).

ii. Children who died of non-disease conditions (e.g., trauma) were excluded, as they were not typically referred to the hospice palliative care program, except for bereavement services for the families.

3.2.5 Data Source

CPCH provided registry of children who died in the facility between 2008 and 2012 with information on demographics, disease, and program enrollment. Number, type of admissions, and acuity levels were extracted from medical charts and administrative records utilized by the hospice's multidisciplinary team.

Provincial Health Services Authority (PHSA) is in possession of the BCCH administrative database. The PHSA Decision Support Unit ran the match search internally (according to the matching criteria) and provided a list of potential matches, along with information on demographics and disease. Potential pairs were reviewed and approved by a palliative care physician expert. A pediatric oncologist provided assistance when questions arose. PHSA provided information on types and numbers of admissions from the administrative database. Medical charts were reviewed to complete information regarding secondary outcomes, which was lacking in the electronic records.

3.2.6 Ethics Approval

Approval was obtained from the Research Ethics Board of Children's and Women's Health Centre of British Columbia, under the University of British Columbia's Research Board (certificate numbers CW13-0210 / H13-01162). CPCH Research Review Committee also approved the proposal. The study was based on secondary data analyses with no involvement of research subjects or their families.

3.2.7 Sampling Procedures and Matching Process

The matching process began on data from PPCP database of children who died in hospice between 2008 and 2012 (n=100). Cancer patients represented 41% of this population. Matching children were sought among pediatric residents of BC who died in BCCH from the same disease (ICD code 3-digit level) at approximately the same age. Age difference within pairs was allowed as according to the following criteria:

Case < 1 year: Control up to 3 months older/younger;

Case 1 - 2 years: Control up to 6 months older/younger;

Case > 2 years: Control up to 12 months older/younger.

Initially 19 pairs were found, of which only 3 were cancer patients. Distinct cases and controls were used. Whenever a case or control was matched more than once, the pair with the smallest age difference was chosen and the remaining excluded. During chart review the primary diagnosis was confirmed through medical notes. Children who were not receiving palliative care and died as a consequence/complications of curative treatments were excluded, as they would not

have met the criterion of being referred to a PPCP in the course of their treatment. Finally, **only 5 pairs of non-cancer patients were obtained.**

3.2.8 Deviations from Original Matching Strategy

Because cancer patients represent a significant proportion of children enrolled in PPCP a different strategy was adopted to identify comparable pairs to include in the study.

Access to the cancer registry was granted from the pediatric oncology department at BCCH and information on children who died from cancer between 2008 and 2012 was retrieved, regardless of place of death (n=105). Distinction between children who were enrolled in PPCP (n=66) and those who were not enrolled (n=39) was achieved using PPCP registry from the hospice.

ICD codes failed to enable matching according to cancer type, and therefore, they were replaced by the description of cancer type reported in the registry's diagnostic field. A physician and a nurse were reviewed the cases one by one (using the cancer registry and electronic medical records (EMR) if needed) to ensure matched pairs were comparable.

Once children were paired by cancer type, an oncologist assessed the implications for treatment of age difference within pairs. If the age difference within a pair implied that different treatment courses for the same cancer would have been used (e.g. radiotherapy + chemotherapy + surgery vs chemotherapy + surgery), the pair was excluded.

Since the process was only able to produce 3 pairs, it was repeated for children deceased in 2013. Ultimately, **6 pairs of cancer patients were obtained.** Even tough 4 controls died outside BCCH they were include in the study, giving the scarcity of matches. Nonetheless, the last admission before death of their cases was excluded, to avoid bias towards higher utilization in the CPCH group.

3.2.9 Outcome Measures

The outcomes of interest in this local comparative analysis were:

Primary outcomes:

1. Number of outpatient and inpatient admissions of any type (emergency visits, general ward, critical care) in both settings (hospital and hospice);
2. LOS, overall and per type of admission (critical and non-critical);
3. Direct costs associated with those admissions (this specific outcome is presented in Chapter 4).

Secondary outcomes:

1. LOS in the last hospitalization before death;
2. Utilization of invasive procedures to prolong life in the last admission (mechanical ventilation, vasoactive drugs and resuscitative attempts).

3.2.10 Hypothesis

Since a systematic review of the literature (Chapter 2) revealed a high heterogeneity of health care resource utilization and costs between children who accessed PPCP to those under usual care, a hypothesis was made that the comparison between cases and controls would present differences in health care utilization. However, no speculation on the direction of the results was made, and a 2-sided test was performed in the statistical analyses.

3.2.11 Sample Size

In usual practice, the sample size used in a study is usually determined based on the expense of data collection and the need to have sufficient statistical power. However, in the current research, the objective was to compare health care utilization between cases and controls, rather than determine how many matched pairs are required to achieve a certain power to detect differences before conducting the study. Furthermore, the cohort of cases was the entire pediatric population deceased in hospice under the program between 2008 and 2012 (which itself is a small population size [n=100]) with **a final sample meeting the matching criteria of 11 pairs**. Therefore, recruiting a larger sample at the time of this study is unfeasible regardless of any calculation of sample size.

However, this sample provides information about the variability of the outcomes, which plays an important role in such sample size calculations and may provide useful guidance for the planning of future related studies. Thus a related calculation was presented after the results, to determine how large the mean difference in the outcomes would have to be, to have a reasonable chance of detecting it in a study of n = 11 matched pairs. Further, an estimate of the ideal sample size was calculate, to achieve a certain power to detect differences between groups in LOS, as this outcome is the most resource intense.

3.2.12 Observational Period

Data on the monthly utilization of health services, for up to 3 years prior to death, was obtained for all cases and controls.

Children deceased before the age of 3 were included in this study. As these subjects had less than 3 years of utilization data prior to death, the observational periods *across pairs* were

affected. Likewise, the age difference allowed between matches affects observational period *within pairs*.

These differences made the outcomes incomparable across pairs. To account for this issue, each outcome measure was weighted by the corresponding observational period for each subject, and expressed on a monthly basis. For example, if the observational periods for the 2 subjects in a pair were 7 months and 8 months, the first measured LOS was scaled by 7 and the second by 8. Through this method, the original LOS is converted to "LOS per month". This process allowed the outcomes within and across pairs to be comparable for statistical analysis.

3.2.13 Statistical Analyses

Wilcoxon signed rank test was used to analyze LOS and number of admissions, since the data did not meet the assumptions for a parametric test (paired t-test).

The assumptions for the paired t-test are that the pairs must be independent of each other, differences between pairs (z_is) can be considered a random sample, and the z_is are normally distributed. The latter assumption did not hold, as a sample size of 11 pairs may not be large enough to represent the true underlying distribution and to rely on the Central Limit Theorem. Another limitation was the requirement that z_is have the same variance. With only a small number of pairs, this essential assumption was impossible to verify and one has to rely on the sampling procedure. The fundamental question is whether the differences resulting from the pairs can reasonably be treated as if they are a representative sample.

Finally, number of admissions is a count outcome, and the distribution of a count variable is usually skewed, not satisfying the normality assumption.

Similarly, the **Wilcoxon signed rank test** was used to assess whether the median of the underlying distribution of the z_is is equal to zero. The advantage of the Wilcoxon signed rank test is that it does not depend on the exact form of the parent distribution. If m denotes this underlying median of z, the hypotheses were:

$$H_0: m = 0$$

$$H_1: m \neq 0$$

The assumptions for Wilcoxon signed rank test are that each pair was chosen randomly and independently, and the distribution of the z_is is symmetric around the median, which can be verified using box-plots. However, because of the small sample size, it was difficult to assess the symmetry assumption using box-plots, and even if all the assumptions could have been satisfied, a signed rank test may not have had sufficient power to reject the null hypothesis that the median difference in hospital utilization between pairs was 0 (zero).

Given the small sample size, a non-parametric test was chosen to compare the utilization of mechanical ventilation, vasoactive drugs, and resuscitation attempt. For these binary outcomes, the McNemar's test was employed (an appropriate choice for testing equal probability of use of those procedures)

Considering that each observation was the utilization of these procedures within each matched pair, the test assessed whether the cases had a different utilization of those procedures compared to their control. Expressing 'yes'=1 and 'no'=0 the null and alternative hypotheses were:

$$H_0: p_{10} = p_{01}$$

$$H_1: p_{10} \neq p_{01}$$

Since only the probabilities p_{10} and p_{01} were involved, only the "discordant" cells in the 2x2 tables are relevant for this test. The p-value indicates if there is sufficient evidence to reject the null hypothesis that the probabilities of using any procedure are the same for the case and control groups.

The above analysis was reasonable provided that the durations of last admission were nearly the same across pairs and within pairs. If durations were very different, different probabilities would have been estimated. If that was the case, the approach described above may not have been valid.

An $\alpha = 0.05$ was chosen to determine the significance of the results. A 2-sided test was employed to assess the significance of the results in both directions, and 95% confidence intervals (CI) were calculated. All statistical analyses were conducted using R software© (Version 0.98.507 2009).

3.2.13.1 Exploratory Analysis

To explore trends in LOS over time, a time series display was used in order to understand how both groups gradually changed behavior near death with respect to time spent in hospital and enrollment in the PPCP. As children in the CPCH group were not under the intervention during the entire study period, a comparative analysis for the period pre- and post-referral was performed. To determine the before and after period in the control group, the "length onto the program" (LOP) of cases was applied to their matched control, assuming that, their death at the same age and from the same disease, could have led to program referral for the same period of time as their match. The admissions were then classified as "before program", and "after program", and additional analysis was run to check if the groups were different at baseline before the "referral point" and

after it. Changes from pre- to post-referral period were analyzed separately for each arm of the study to understand disease trajectory (and account for any systematic selection bias in PPCP enrolment). Mean values of monthly outcomes were presented for future research purposes and economic models. However, the estimates for the differences between groups tested in the pairwise analyses to account for disease and age are based on the median values. The group means and the pairwise statistical tests may show opposite direction of results caused by skewedness in the distributions of the outcomes. For instance, the mean monthly LOS estimates might look lower in the CPCH group but the pairwise analyses show a longer monthly LOS in the CPCH group (Table 16). Whenever possible, separate subgroup analyses for cancer patients were presented. An exploratory analysis by health care setting was also carried out.

3.3 Results

3.3.1 Demographics

Despite no significant difference in age and gender distributions between groups, the PPCP group was older by 1.5-2.8 years, and the proportion of female was 2.5-5 times higher than the control group. These findings were more pronounced in the cancer group

The mean observation period was 23.4 months and the mean LOP was 89 days, with a large range of 2 to 345 days before death.

A single significant difference between the matched cohorts was found (Table 8): overall, cases had more advanced directives in place compared to controls (100% of cases with “Do not Attempt Resuscitation directive (DNAR) versus 27% of controls, $p = 0.013$), however, it could have happened just by chance giving the small sample size. The cancer sub-group drove this result as

every case had a DNAR whereas none of the controls did. In the non-cancer sub-group, the difference between groups was not significant.

Table 8. Demographics Table

	All			Cancer			Non-cancer		
	CPCH	BCCH	p	CPCH	BCCH	p	CPCH	BCCH	p
n	11	11		6	6		5	5	
Female % (n)	**72.7 (8)**	**27.3 (3)**	0.182	**83.3 (5)**	**16.7 (1)**	0.371	**60 (3)**	**40 (2)**	1.000
Mean Age in Years (sd)	7.3 (6.7)	8.86 (8.1)	0.554	11.7 (5.1)	14.5 (5.6)	0.688	1.96 (4.1)	2.04 (4.3)	0.423
Age Range in Years	(0.01, 18.7)	(0.01, 22.0)		(5.18, 18.7)	(6.84, 22.0)		(0.01, 9.3)	(0.01,9.7)	
Mean Follow-up in Months (sd)	23.4 (17.5)	23.4 (17.5)	1.000	36 (0)	36 (0)	Na	8.3 (15.5)	8.33 (15.5)	1.000
Observational Period Range in Months	(0.12, 36)	(0.12, 36)		(36, 36)	(36, 36)		(0.12, 36)	(0.12, 36)	
With DNAR % (n)	100 (11)	27.3 (3)	**0.013**	100 (6)	0 (0)	**0.041**	100 (5)	60 (3)	0.480
Mean LOP in Days (sd)	89 (112.9)	NA		138.2 (129.4)	NA		30 (54.4)	NA	
LOP Range in Days	(2, 345)	NA		(4, 345)	NA		(2, 127)	NA	

CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); sd: standard deviation; n: sample size; p: p-value; DNAR: do not attempt to resuscitate form; LOP: length of enrollment onto the program; NA: not applicable

3.3.2 Number of Admissions

Overall, no significant differences were found between groups, which it is potentially due to the low power of this study to detect true differences if they existed. In the pre-referral period, children in the CPCH group had, on average, 1.83 monthly admissions per patient versus 3.69 in the BCCH group. In the pairwise analyses (controlling for disease and age at death), CPCH children had a tendency towards fewer monthly admissions per patient overall, with the exception of emergency visits (Table 9). The results were driven by the non-cancer subgroup results (Figure 4).

Table 9. Monthly Number of Admissions per Type, and Pairwise Test (Wilcoxon) – Pre-Referral to PPCP Period[§]

Pre-referral Period Number of Admissions/Patient/Month Mean (sd)			Median Pairwise Difference			
				Confidence Interval		
Variable	CPCH	BCCH	Estimate	2.50%	97.50%	*p*
Total	1.83 (1.59)	3.69 (5.64)	-0.68	-6.83	1.23	0.492
Outpatient	1.37 (1.66)	1.55 (2.09)	-0.32	-1.71	1.51	0.813
Emergency#	0.03 (0.04)	0.01 (0.02)	0.04	0.03	0.06	0.059
Inpatient	0.43 (0.62)	2.13 (5.75)	-0.06	-8.07	0.23	0.722
ICU*	0.32 (0.67)	1.06 (2.90)	-0.17	-3.56	0.22	0.675
Non-critical	0.11 (0.10)	1.08 (2.87)	-0.10	-4.53	0.20	0.343

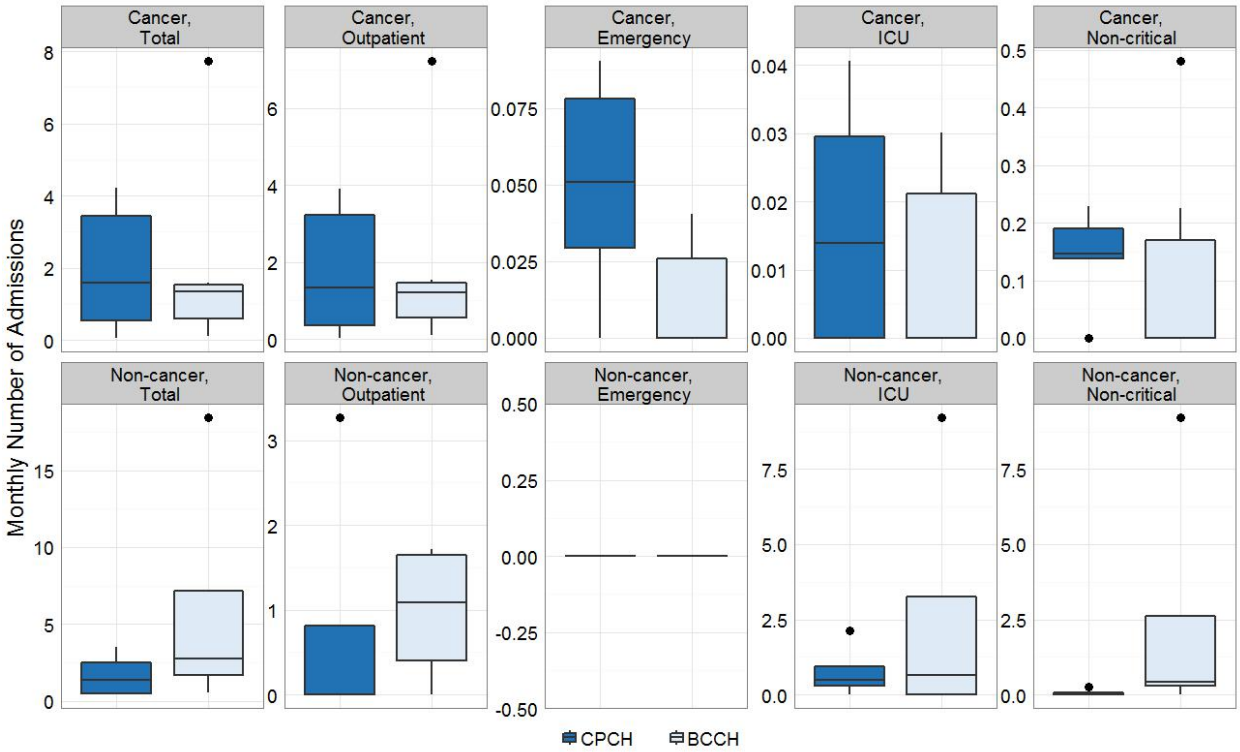
CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); CI: confidence interval; p: p-value; ICU: intensive care unit admissions; sd: standard deviation

§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth;

* CI of 95% was not achievable because of lack of data. Software R automatically calculated a CI of 90%.

CI of 95% was not achievable because of lack of data. Software R automatically calculated a CI of 80%.

Figure 4. Monthly Number of Admissions per Type - by Cancer and Non-cancer subgroup – Pre-Referral to PPCP [§]



§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth.

Conversely, when isolating the post-referral period, children in the CPCH arm had significantly more admissions, which have to be interpreted with caution since it could still be happening by chance given the multiple tests performed in this dataset. Children in the CPCH group had, on average 6.57 monthly admissions per patient versus 3.11 in the BCCH group. In the pairwise analyses (controlling for disease and age at death), the number of inpatient admissions in critical care beds drove the results with borderline significance level (p = 0.059). No statistically significant difference was found in the number of admissions in outpatient, emergency room visits or non-critical inpatient admissions (Table 10). In the subgroup analysis, the same pattern for cancer and non-cancer patients was observed (Figure 5). The individual pairwise analyses are available in appendix E.

Table 10. Monthly Number of Admissions per Type, and Pairwise Test (Wilcoxon) – Post-Referral to PPCP Period

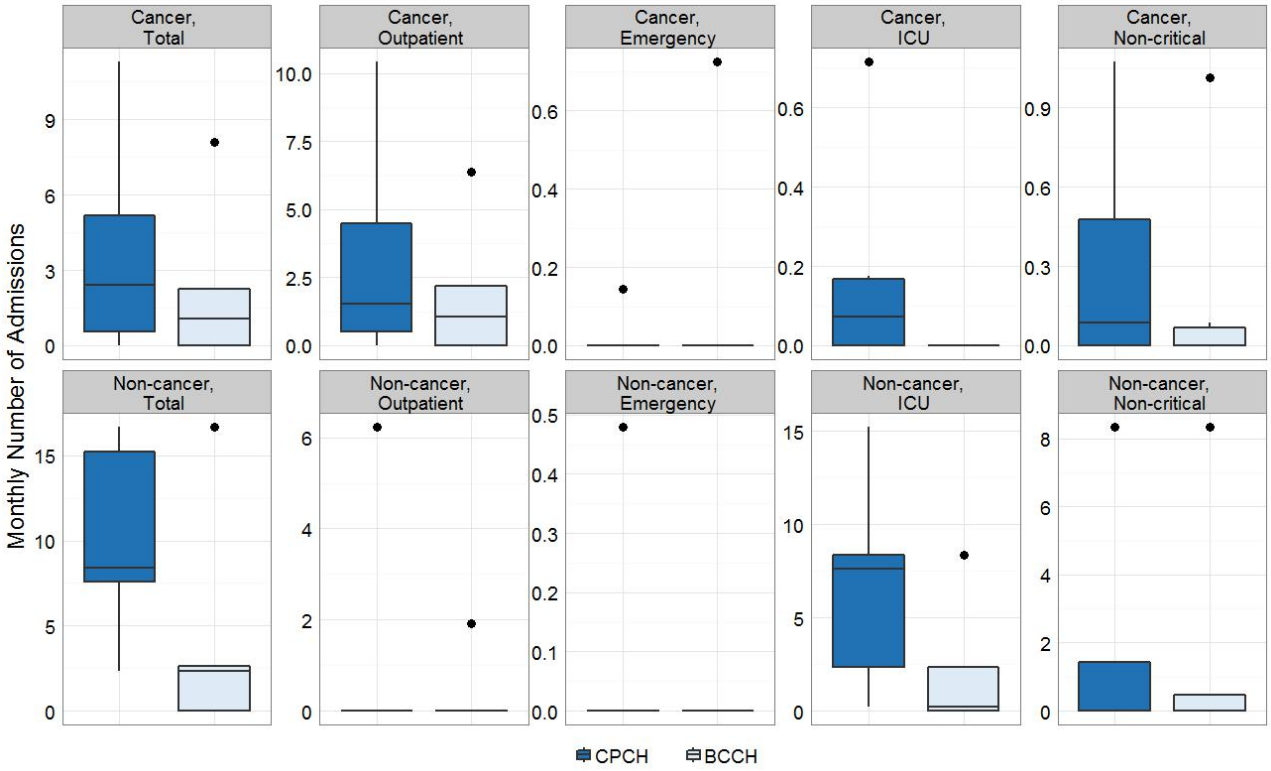
Post-referral Period Number of Admissions/Patient/Month Mean (sd)			Median Pairwise Difference			
				Confidence Interval		
Variable	CPCH	BCCH	Estimate	2.50%	97.50%	*p*
Total	6.57 (5.84)	3.11 (5.09)	3.78	1.07	9.37	**0.014**
Outpatient*	2.30 (3.52)	1.15 (1.98)	2.12	0.18	4.06	0.093
Emergency	0.06 (0.15)	0.07 (0.22)	0.00			**NA**
Inpatient*	4.21 (6.19)	1.89 (4.95)	3.66	0.26	8.50	0.093
ICU#	3.16 (5.05)	0.99 (2.53)	3.89	0.18	7.96	0.059
Non-critical#	1.05 (2.47)	0.90 (2.49)	0.40	-0.43	1.07	0.361

CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); CI: confidence interval; p: p-value; ICU: intensive care unit admissions; sd: standard deviation

* CI of 95% was not achievable because of lack of data. Software R automatically calculated a CI of 90%.

CI of 95% was not achievable because of lack of data. Software R automatically calculated a CI of 80%.

Figure 5. Monthly Number of Admissions per Type - by Cancer and Non-cancer subgroup – Post-Referral to PPCP



An opposite trend in admissions per month was found, when comparing patients in the post-referral period to themselves in the pre-referral period. In the CPCH group, the median difference was 3.08 monthly admissions greater in the post-referral period compared to before the referral (p = 0.01). The number of admissions in critical care beds drove the results (p = 0.021), mainly in the non-cancer group (Table 11 and Figure 6). Again, it has to be interpreted with caution since it could still be happening by chance given the multiple tests performed in this dataset and the selection bias inherent to this research.

Table 11. Changes in the Monthly Number of Admissions per Type, in the CPCH Arm and Pairwise Test (Wilcoxon) – Pre-Post-Referral to PPCP[§]

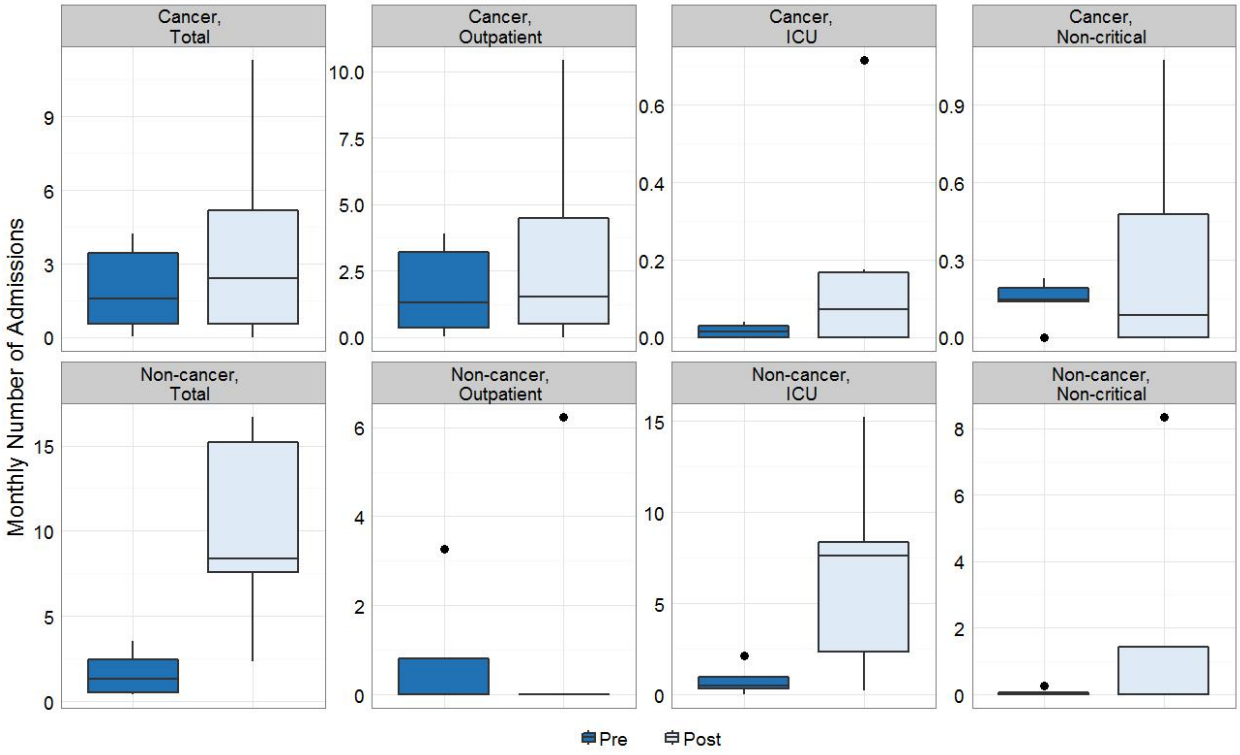
Changes from Pre- to Post-referral CPCH Arm Number of Admissions/Patient/Month Mean (sd)			Median Pairwise Difference			
				Confidence Interval		
Variable	Before	After	Estimate	2.50%	97.50%	*p*
Total	1.83 (1.59)	5.57 (5.05)	3.08	0.69	7.55	**0.010**
Outpatient	1.37 (1.66)	2.53 (3.62)	1.34	-0.52	4.92	0.205
Inpatient	0.43 (0.62)	2.97 (4.87)	1.02	0.04	7.26	**0.049**
ICU	0.32 (0.67)	2.64 (5.00)	1.05	0.11	7.68	**0.021**
Non-critical*	0.11 (0.10)	0.33 (0.53)	0.39	-0.14	0.92	0.402

CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); CI: confidence interval; p: p-value; ICU: intensive care unit admissions; sd: standard deviation

§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth;

* CI of 95% was not achievable because of lack of data. Software R automatically calculated a CI of 90%.

Figure 6. CPCH Arm - Changes in the Monthly Number of Admissions per Type, by Cancer and Non-cancer subgroup – Pre-Post-Referral to PPCP[§]



§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth.

Conversely, in the BCCH arm, no statistically significant changes were observed in the number of admissions of any type from the pre- to post-referral period. A tendency towards fewer monthly admissions per patient post-referral was seen (Table 12). This pattern was consistent in both cancer and non-cancer subgroups (Figure 7).

It is important to assess whether this increase in inpatient admissions in the CPCH arm occurred in hospital or hospice in order to understand where health care is utilized, to evaluate opportunity costs, and to plan care delivery. According to Table 13, the majority of admissions in the CPCH group post-enrollment occurred in hospice, demonstrating a shift in health care setting utilization, despite being critical care admissions.

Table 12. Changes in the Monthly Number of Admissions per Type, in the BCCH Arm and Pairwise Test (Wilcoxon) – Pre-Post-Referral to PPCP[§]

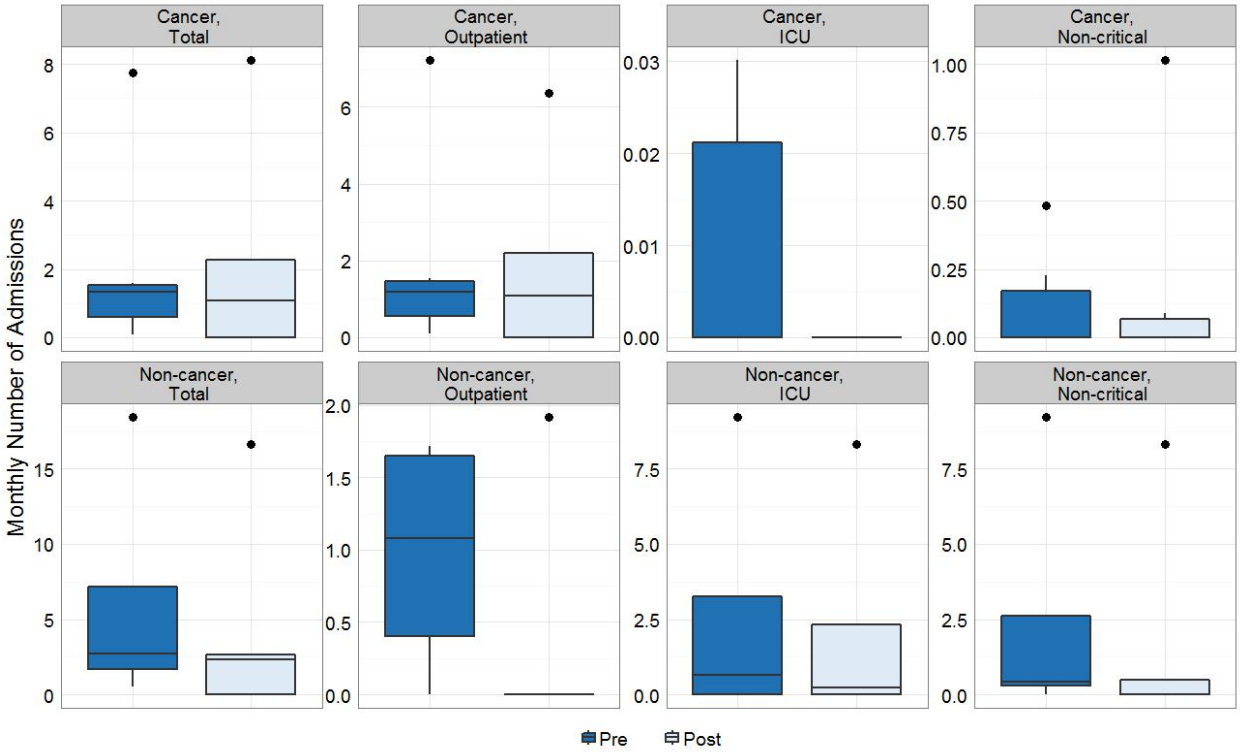
Changes from Pre- to Post-referral BCCH Arm Number of Admissions/Patient/Month Mean (sd)			Median Pairwise Difference			
				Confidence Interval		
Variable	**Before**	**After**	**Estimate**	**2.50%**	**97.50%**	***p***
Total	3.69 (5.64)	1.75 (2.52)	-0.23	-8.79	0.78	0.846
Outpatient	1.55 (2.09)	1.26 (2.05)	-0.36	-1.28	0.66	0.407
Inpatient	2.13 (5.75)	0.42 (0.77)	-0.02	-9.23	1.01	1.000
ICU*	1.06 (2.90)	0.26 (0.74)	-0.03	-4.62	1.16	0.675
Non-critical	1.08 (2.87)	0.16 (0.34)	-0.17	-4.82	0.51	0.800

BCCH: BC Children's Cohort (controls); CI: confidence interval; p: p-value; ICU: intensive care unit admissions; sd: standard deviation

§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth.

* CI of 95% was not achievable because of lack of data. Software R automatically calculated a CI of 90%.

Figure 7. BCCH Arm - Changes in the Monthly Number of Admissions per Type, by Cancer and Non-cancer subgroup – Pre-Post-Referral to PPCP[§]



§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth.

Table 13. Monthly Number of Inpatient Admissions per Facility – Entire Study Period[§]

Admissions/Patient/Month by facility – Mean (sd)			
All			
Arm	**CPCH Arm**		**BCCH Arm**
Facility	Hospital	Hospice	Hospital
Pre	0.43 (0.62)		2.13 (5.75)
Post	1.04 (2.46)	3.17 (5.04)	1.89 (4.95)
Admissions/Patient/Month by facility – Mean (sd)			
Non-cancer Patients			
Arm	**CPCH Arm**		**BCCH Arm**
Facility	Hospital	Hospice	Hospital
Pre	0.83 (0.88)		5.14 (8.89)
Post	1.91 (3.63)	6.79 (5.78)	3.94 (7.18)
Admissions/Patient/Month by facility – Mean (sd)			
Cancer Patients			
Arm	**CPCH Arm**		**BCCH Arm**
Facility	Hospital	Hospice	Hospital
Pre	0.16 (0.08)		0.13 (0.20)
Post	0.32 (0.43)	0.16 (0.28)	0.18 (0.41)

CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); sd: standard deviation

§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth.

3.3.3 LOS

To clarify the patterns in each group overtime, time series graphs demonstrate trends in resource utilization in each group and the degree of enrollment in the PPCP (Figure 8 and 9). The dark and light blue lines represent the observed total LOS per person-month at risk of being hospitalized over time, in CPCH and BCCH arm, respectively. The grey bars represent the observed proportion of patients enrolled in the PPCP per person-month at risk, over time. The groups were very similar until approximately 15 months before death. More specifically, 6 months prior to death a drastic divergence between the groups in inpatient utilization was observed. In addition to the small sample size, this later disparity between groups could be the cause of not finding significant differences in the aggregated measures. Although both groups showed a decrease in outpatient visits, and an increase in inpatient admissions towards death, CPCH group seemed to have consumed more health care resources in this period both as outpatients and inpatients (Figure 8). In terms of level of acuity of inpatient admissions, CPCH patients consistently had more non-critical admissions than BCCH patients, from 15 to 2 months prior to death. In the last 2 months, CPCH patients became more severely ill than their matches and had a higher monthly LOS in critical care beds.

Children in the CPCH arm started to enroll in the PPCP from 12 months before death, with a maximum enrollment rate of 67% even in the last month of life. This may be due to children enrolling only a few days before death or being younger than 30 days old.

Figure 8. LOS per Person-Month at Risk Over Time, by Arm: Outpatient and Inpatient Admissions

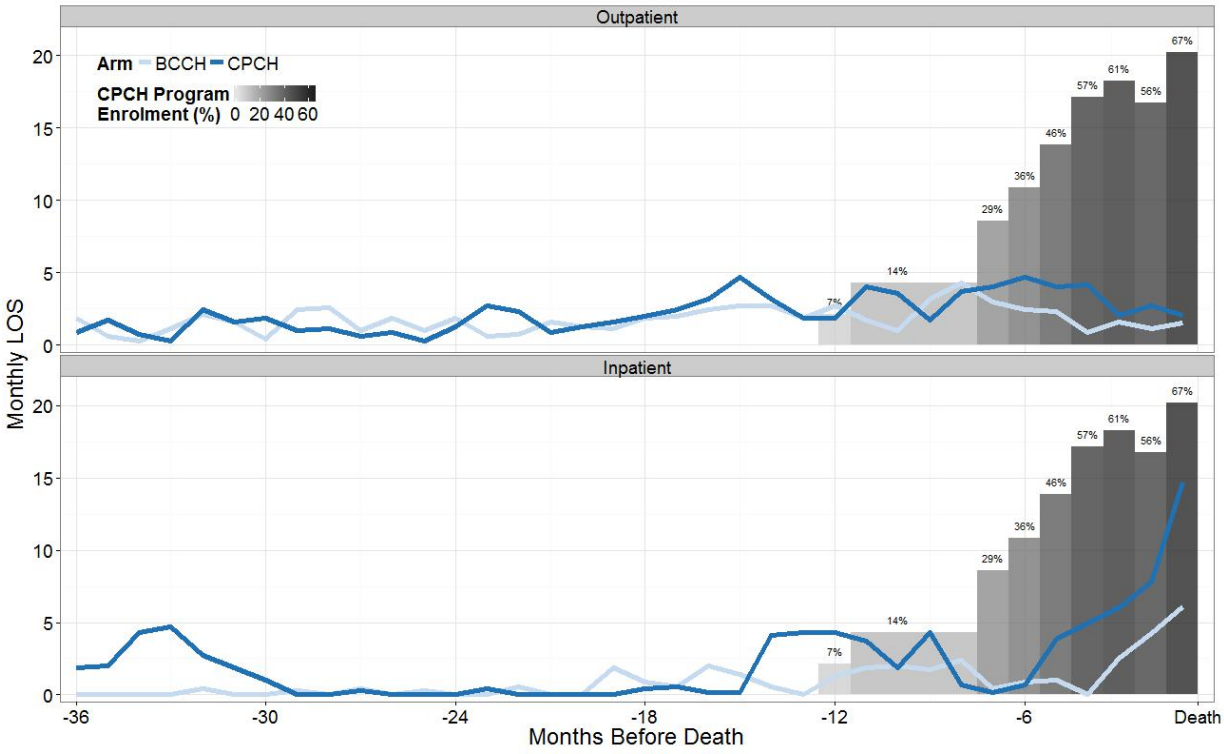
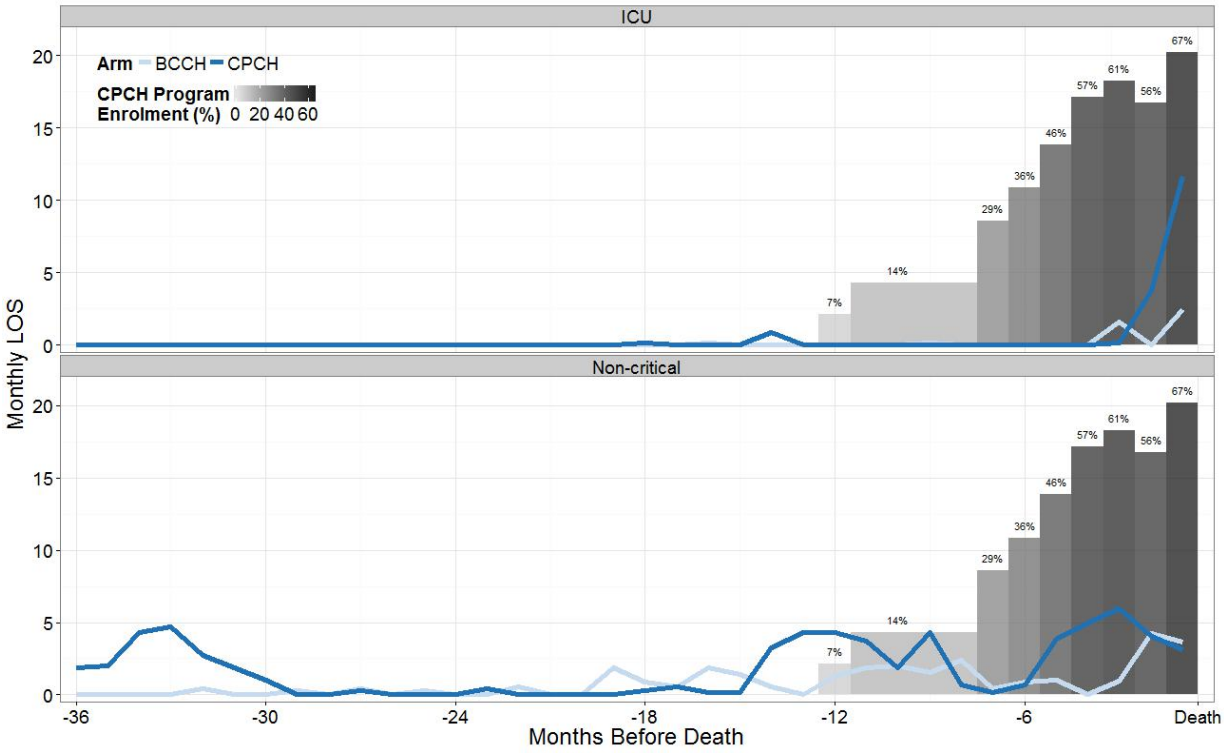


Figure 9. LOS per Person-Month at Risk Over Time Over Time by Arm: Critical and Non-Critical Admissions



No significant differences between the groups were found in any period, which it is potentially due to the low power of this study to detect true differences if they existed. In the pre-referral period, children in the CPCH group had, on average, a 10.68 days monthly LOS per patient versus 10.85 days in the BCCH group. In the pairwise analyses (controlling for disease and age at death), a tendency towards longer LOS in the CPCH arm for all admission types was observed with the exception of outpatient visits and non-critical admissions (Table 14). These results were driven by the patterns in the cancer group as well as outliers (Figure 10).

Table 14. Monthly LOS per Type, and Pairwise Test (Wilcoxon) – Pre-Referral to PPCP Period[§]

Pre-referral Period LOS/Patient/Month - Mean (sd)			Median Pairwise Difference			
				Confidence Interval		
Variable	CPCH	BCCH	Estimate	2.50%	97.50%	*p*
Total	10.68 (12.42)	10.85 (22.72)	1.91	-14.29	8.12	0.432
Outpatient	1.37 (1.66)	1.55 (2.09)	-0.32	-1.71	1.51	0.813
Emergency#	0.03 (0.04)	0.01 (0.02)	0.04	0.03	0.06	0.059
Inpatient	9.28 (13.05)	9.29 (23.08)	0.88	-13.47	8.45	0.275
ICU*	7.89 (13.71)	7.27 (20.28)	0.22	-15.09	13.05	0.529
Non-critical	1.39 (2.48)	2.02 (3.26)	-0.13	-4.53	3.89	0.906

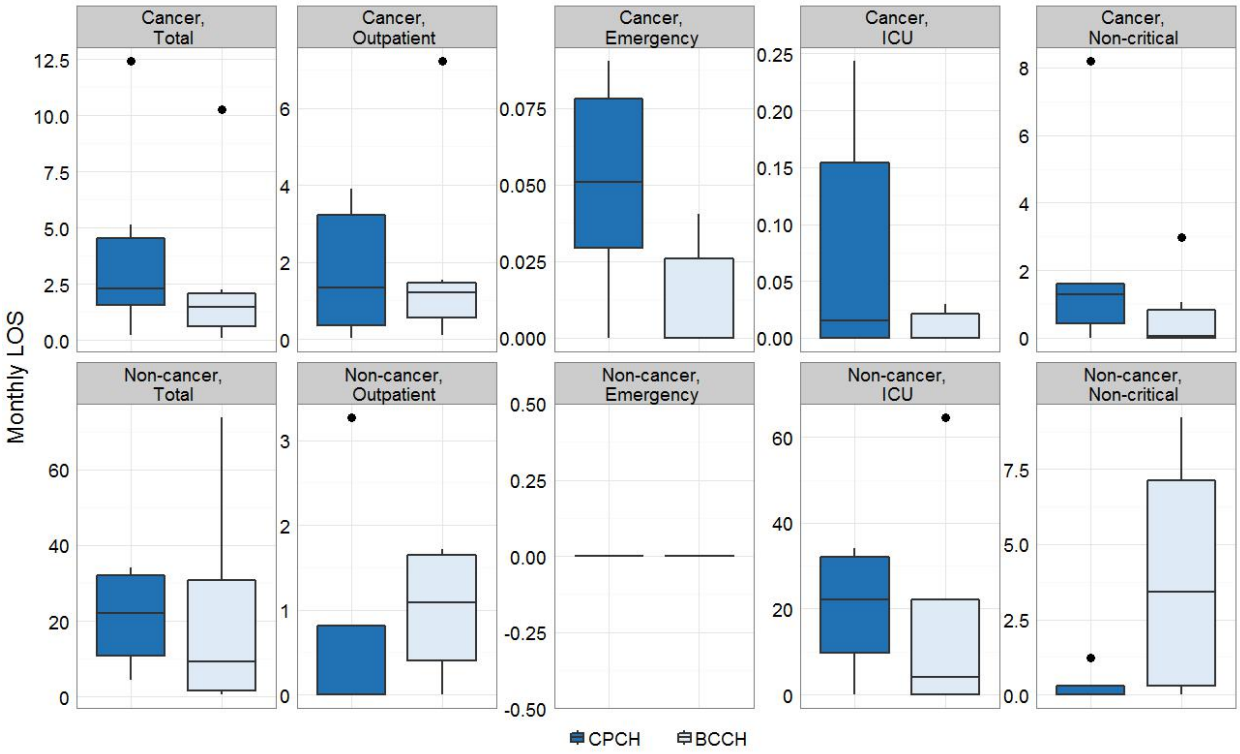
CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); CI: confidence interval; p: p-value; ICU: intensive care unit admissions; sd: standard deviation

* CI of 95% was not achievable because of lack of data. Software R automatically calculated a CI of 90%.

CI of 95% was not achievable because of lack of data. Software R automatically calculated a CI of 80%.

§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth.

Figure 10. Monthly LOS per Type - by Cancer and Non-cancer subgroup – Pre-Referral to PPCP[§]



§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth.

While in post-referral period a higher number of hospital admissions in the CPCH arm was previously found, no significant difference in LOS was observed between groups in the same period (Table 15). Children in the CPCH group had, on average, a monthly LOS of 8.53 days per patient versus 6.79 days in the BCCH group. In the pairwise analyses (controlling for disease and age at death), though not significant, a tendency to longer LOS in the CPCH arm for all types of admissions was observed, with much higher estimates of the median differences between pairs than in the pre-referral period. Again, the lack of significance in the statistical analyses might have been due to the low power of this study to detect true differences if they existed. Outliers occurred in the BCCH arm for both critical and non-critical admissions, but when the interquartile range in both cancer and non-cancer subgroups were taken into consideration, the LOS tended to be higher in the CPCH arm (Figure 11). The individual pairwise analyses are available in Appendix E.

Table 15. Monthly LOS per Type, and Pairwise Test (Wilcoxon) – Post-Referral to PPCP Period

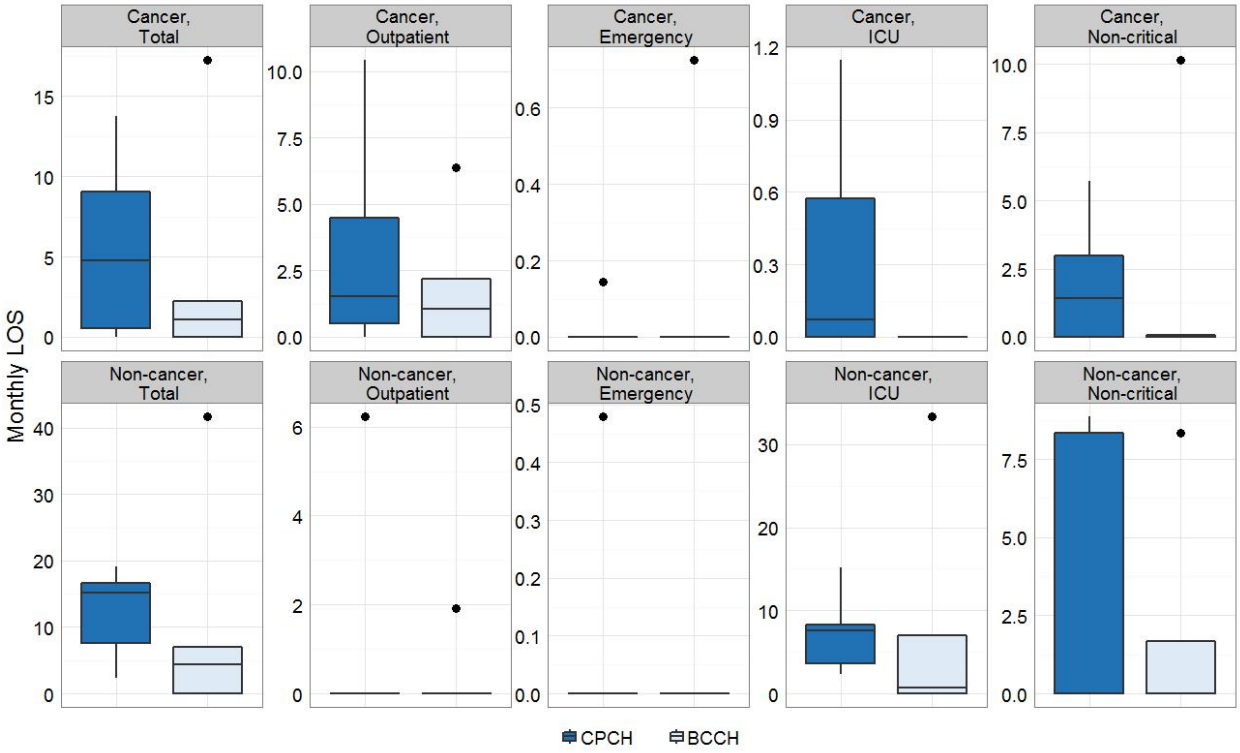
Post-referral Period LOS/Patient/Month - Mean (sd)			Median Pairwise Difference			
				Confidence Interval		
Variable	CPCH	BCCH	Estimate	2.50%	97.50%	*P*
Total	8.53 (6.94)	6.79 (12.66)	3.66	-8.70	10.47	0.308
Outpatient*	2.30 (3.52)	1.15 (1.98)	2.12	0.18	4.06	0.093
Emergency	0.06 (0.15)	0.07 (0.22)	0.00			NA
Inpatient	6.17 (6.13)	5.57 (12.46)	2.09	-10.56	10.82	0.624
ICU	3.55 (4.90)	3.73 (10.04)	0.99	-12.14	8.18	0.529
Non-critical#	2.62 (3.50)	1.84 (3.71)	2.82	-7.10	7.18	0.584

CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); CI: confidence interval; p: p-value; ICU: intensive care unit admissions; sd: standard deviation

* CI of 95% was not achievable because of lack of data. Software R automatically calculated a CI of 90%.

CI of 95% was not achievable because of lack of data. Software R automatically calculated a CI of 80%.

Figure 11. Monthly LOS per Type - by Cancer and Non-cancer subgroup – Post-Referral to PPCP



When patients were compared to themselves in the pre-referral period, non-significant but puzzling patterns between groups were found. Overall LOS per month decreased for CPCH children post-enrollment, except for non-critical and outpatient admissions (Table 16). This pattern was more evident for non-cancer patients (Figure 12).

In the BCCH group, patients had a slight overall increase in LOS after the enrollment point. However, when types of admissions and subgroups were analyzed separately, a consistent decrease was observed (Table 17 and Figure 13). This occurred due to the small sample size and the fact that half of the sample had opposite directions of the results.

The shift in health care setting utilization to hospice was confirmed in the analysis of LOS per facility, more noticeable in the non-cancer subgroup (Table 18).

Table 16. Changes in the Monthly LOS per Type, in the CPCH Arm and Pairwise Test (Wilcoxon) – Pre-Post-Referral to PPCP[§]

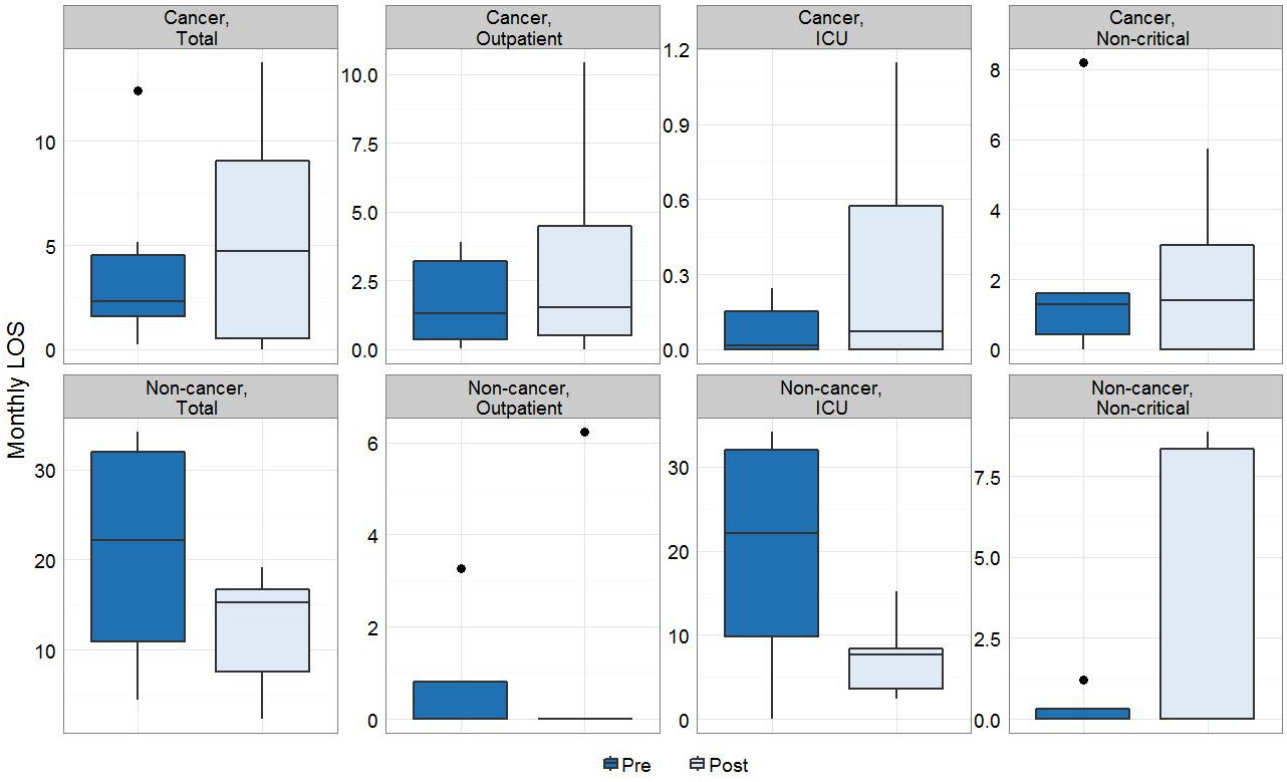
Changes from Pre- to Post-referral CPCH Arm LOS/Patient/Month - Mean (sd)			Median Pairwise Difference			
				Confidence Interval		
Variable	**Before**	**After**	**Estimate**	**2.50%**	**97.50%**	***P***
Total	10.68 (12.42)	7.72 (6.74)	-1.60	-13.39	5.85	0.432
Outpatient	1.37 (1.66)	2.53 (3.62)	1.34	-0.52	4.92	0.205
Inpatient	9.28 (13.05)	5.12 (5.32)	-2.42	-13.37	3.38	0.375
ICU	7.89 (13.71)	3.08 (4.89)	-5.03	-16.07	1.87	0.529
Non-critical*	1.39 (2.48)	2.04 (3.10)	1.13	-3.19	5.48	0.675

CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); CI: confidence interval; p: p-value; ICU: intensive care unit admissions; sd: standard deviation

§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth.

* CI of 95% was not achievable because of lack of data. Software R automatically calculated a CI of 90%.

Figure 12. CPCH Arm - Changes in the Monthly LOS per Type, by Cancer and Non-cancer subgroup – Pre-Post-Referral to PPCP[§]



§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth.

Table 17. Changes in the Monthly LOS per Type, in the BCCH Arm and Pairwise Test (Wilcoxon) – Pre-Post-Referral to PPCP[§]

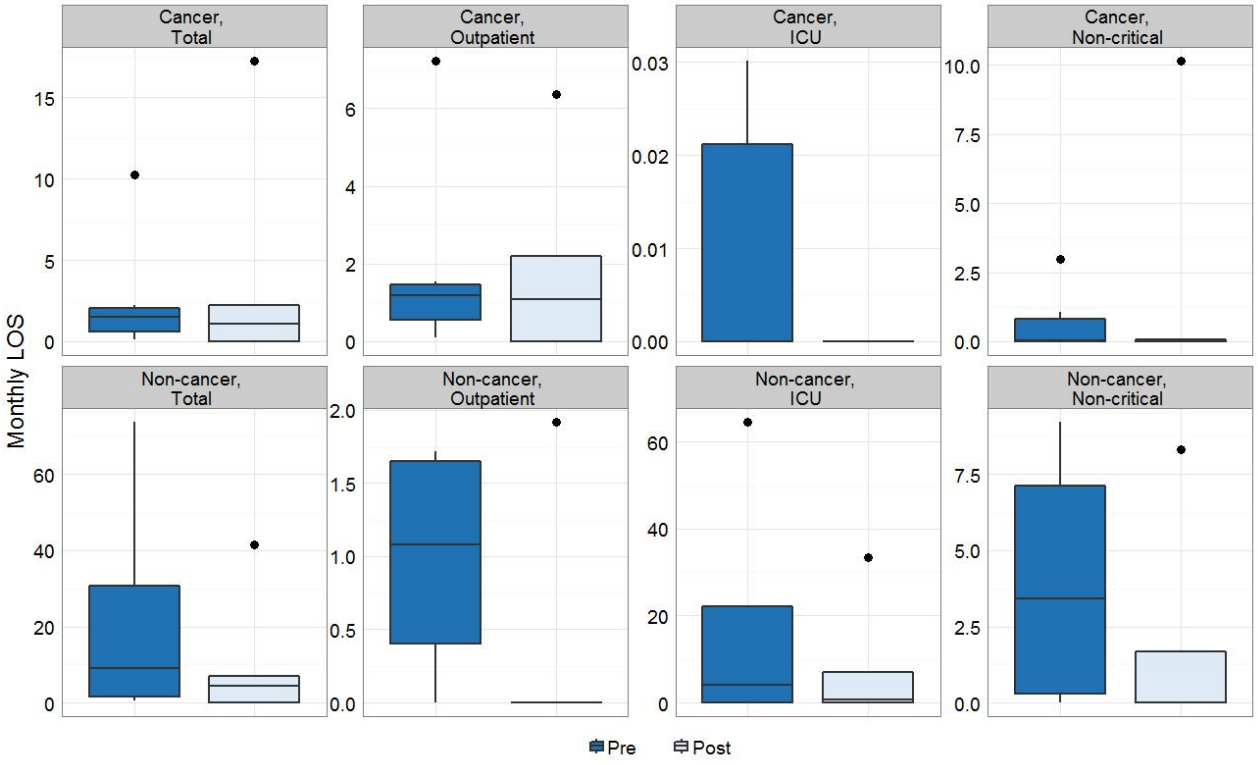
Changes from Pre- to Post-referral BCCH Arm LOS/Patient/Month - Mean (sd)			Median Pairwise Difference			
				Confidence Interval		
Variable	Before	After	Estimate	2.50%	97.50%	*P*
Total	10.85 (22.72)	3.30 (5.44)	0.17	-34.38	3.45	1.000
Outpatient	1.55 (2.09)	1.26 (2.05)	-0.36	-1.28	0.66	0.407
Inpatient	9.29 (23.08)	1.96 (3.64)	-0.41	-36.93	4.77	0.834
ICU*	7.27 (20.28)	0.77 (2.21)	-0.56	-32.28	3.50	0.529
Non-critical	2.02 (3.26)	1.19 (3.19)	-0.43	-5.15	3.53	0.529

BCCH: BC Children's Cohort (controls); CI: confidence interval; p: p-value; ICU: intensive care unit admissions; sd: standard deviation

§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth.

* CI of 95% was not achievable because of lack of data. Software R automatically calculated a CI of 90%.

Figure 13. BCCH Arm - Changes in the Monthly LOS per Type, by Cancer and Non-cancer subgroup – Pre-Post-Referral to PPCP[§]



§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth.

Table 18. Monthly LOS per Facility – Entire Study Period[§]

LOS/Patient/Month by facility – Mean (sd)			
All			
Arm	**CPCH Arm**		**BCCH Arm**
Facility	Hospital	Hospice	Hospital
Pre	9.28 (13.05)		9.29 (23.08)
Post	2.21 (3.17)	3.96 (4.82)	5.57 (12.46)
LOS/Patient/Month by facility - Mean (sd)			
Non-cancer Patients			
Arm	**CPCH Arm**		**BCCH Arm**
Facility	Hospital	Hospice	Hospital
Pre	19.93 (15.58)		22.18 (35.03)
Post	3.25 (4.45)	7.61 (4.88)	10.22 (17.81)
LOS/Patient/Month by facility - Mean (sd)			
Cancer Patients			
Arm	**CPCH Arm**		**BCCH Arm**
Facility	Hospital	Hospice	Hospital
Pre	2.18 (3.12)		0.70 (1.20)
Post	1.35 (1.51)	0.92 (1.71)	1.70 (4.13)

CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); sd: standard deviation

§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth.

3.3.4 Last Admission Before Death (EOL admission)

3.3.4.1 LOS

To analyze the EOL admission before death, the pairs in which the controls did not die at hospital were excluded. No significant difference was found between groups, which is potentially due to the low power of this study to detect true differences if they existed. However, the CPCH group tended to have a lower median difference between pairs by 3.25 days (Table 19).

Table 19. LOS in the Last Admission Before Death and Pairwise Test (Wilcoxon)

LOS Last Admission Mean (sd)		Median Pairwise Difference			
			Confidence Interval		
CPCH	BCCH	Estimate	2.50%	97.50%	*p*
4.57 (6.16)	6.86 (7.82)	-3.25	-14.00	11.00	0.688

§ This analysis contain data from 7 pairs; CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); sd: standard deviation; p: p-value; LOS: length of stay.

3.3.4.2 Invasive Procedures

When invasive procedures that prolong life were studied, the pairwise analyses tested the differences in treatment between pairs (grey cells in the 2x2 tables, Table 20). No significant differences between groups were found. However, the absolute numbers showed a more intense use of invasive procedures in the BCCH arm.

Table 20. Use of Invasive Procedures to Prolong Life in the EOL Admission and Pairwise Test (McNemar's test)

Mechanical Ventilation		
	CPCH	
BCCH	Yes	No
Yes	2	3
No	0	2
p	0.248	

Cardiopulmonary Resuscitation		
	CPCH	
BCCH	Yes	No
Yes	0	2
No	0	5
p	0.480	

Vasoactive Drugs		
	CPCH	
BCCH	Yes	No
Yes	0	3
No	0	4
p	0.480	

CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); p: p-value.

As mentioned in the methods section, the above analyses would have been reasonable provided that LOS of the EOL admission was nearly the same across pairs and within pairs. Although non-significant difference in LOS across pairs was found (Table 19), great discrepancy within pairs was observed (Table 21). As the LOS was not comparable within pairs, different probabilities were being estimated and the aforementioned analyses are not valid.

Table 21. LOS for the EOL Admission by Pairs (in days)

LOS EOL admission (days)		
	Arms	
Pair Number	CPCH	BCCH
5	1	6
55	15	3
60	1	7
78	1	4
81	1	3
302	12	1
304	1	24

CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); LOS: length of stay; EOL: end-of-life

3.3.5 Sample Size Calculation for Future Research

This study is the first of its kind to provide information on the variability of the outcomes among children who died from LTC. A broader comparative approach to inpatient health care utilization was employed, which plays an important role in sample size calculations and may provide useful guidance for planning future related studies.

Using LOS as the main outcome due to its significant impact on health care utilization, estimates of effect size and power to detect differences between groups is presented, relevant to comparative research in the PPCP field.

In the current sample (n=11 pairs), the mean difference between pairs in monthly inpatient LOS was 0.5 days with a standard deviation of 10.4 days (Table 22).

Table 22. Sample Mean LOS, Difference Between Groups and Variability

Mean LOS/Inpatient/Month (in days)	
BCCH	8.8
CPCH	9.3
Difference	0.5
sd	10.4

CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); sd: standard deviation; LOS: length of stay.

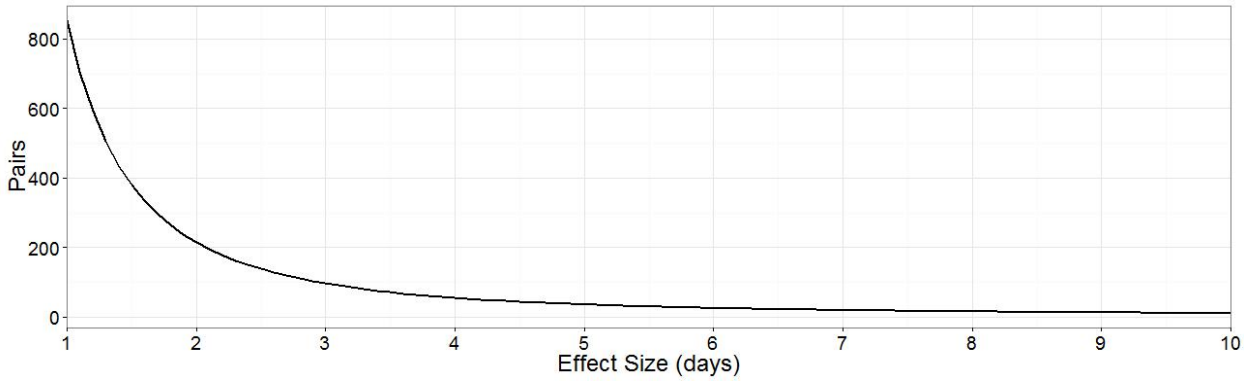
Commonly, from a health care provider perspective, a minimum difference of 1 day in LOS is needed to impact costs. Considering the variance found in this study, to detect a minimum effect size of 1 day/month/patient difference in LOS between groups, an ideal comparative cohort study would have to include 853 pairs of children, using a significance level of 0.05 and a Power of 80%. Therefore, the current study on 11 pairs, under the same parameters had a power of 6.0% to detect a minimum difference in LOS between groups (Table 23), with a 5% probability that any difference occurred by chance.

Table 23. Parametric Test Sample Size Calculation

Paired t-test Power Calculation (2-sided)	
Pairs Needed to Detect Effect Size of 1 day/Month/Patient	Power of this Study to Detect Effect Size of 1 day/Month/Patient
n = 853 α = 0.05 Power = 0.8	n = 11 α = 0.05 Power = 0.060
NOTE: n is number of *pairs*	

Figure 14 shows estimates of the different samples sizes needed to test other effect sizes, considering the variability found in this study, with a significance level = 0.05 and power = 0.80.

Figure 14. Sample Size Estimates for Different Effect Sizes - LOS



3.4 Discussion

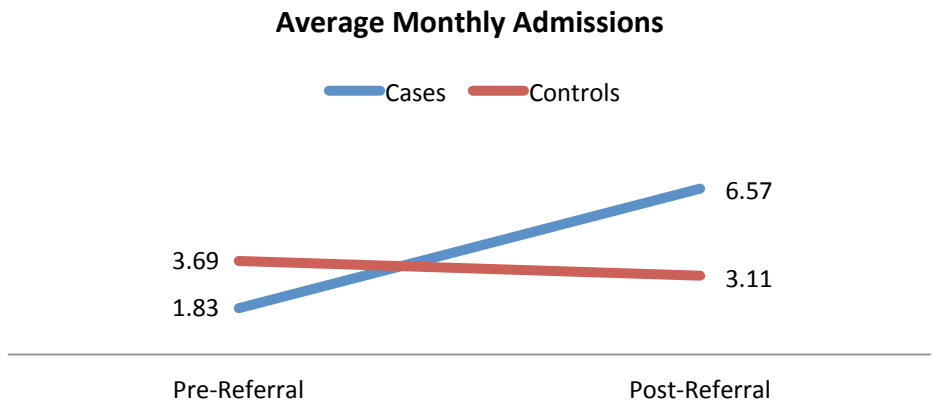
3.4.1 Summary of Results

Figure 15 summarizes the results with respect to the **number of monthly admissions per patient** in the aggregated data**.** In the comparison between groups, the only significant difference was the greater number of monthly admissions found in the CPCH group in the post-referral period (p=0.014). This difference appeared to be driven by the number of admissions in critical care beds, with borderline significance (p=0.059).

When patients were compared to themselves in the pre-referral period an opposite trend between groups was observed. In the CPCH group, the number of monthly admissions increased significantly (p=0.010), driven by inpatient admissions (p=0.049), and related to critical care admissions towards death (p=0.021). These significant findings must be interpreted with caution since it could still be happening by chance given the multiple tests performed in this dataset. Conversely, in the BCCH arm, a decrease was observed in the number of monthly admissions of any type from the pre- to the post-referral, but was not statistically significant. Finding no significant differences in the statistical analyses, not necessarily proves no difference between groups, but might have been due to the low power of this study to detect true differences if they existed.

Other than the increase in admissions in the CPCH arm after referral to PPCP, a shift in the health care setting from the hospital to hospice was observed, irrespective of critical care admissions.

Figure 15. Average Monthly Number of Admissions: Summary of Comparison Between Groups and Changes within Groups from Pre- to Post-Referral Period **

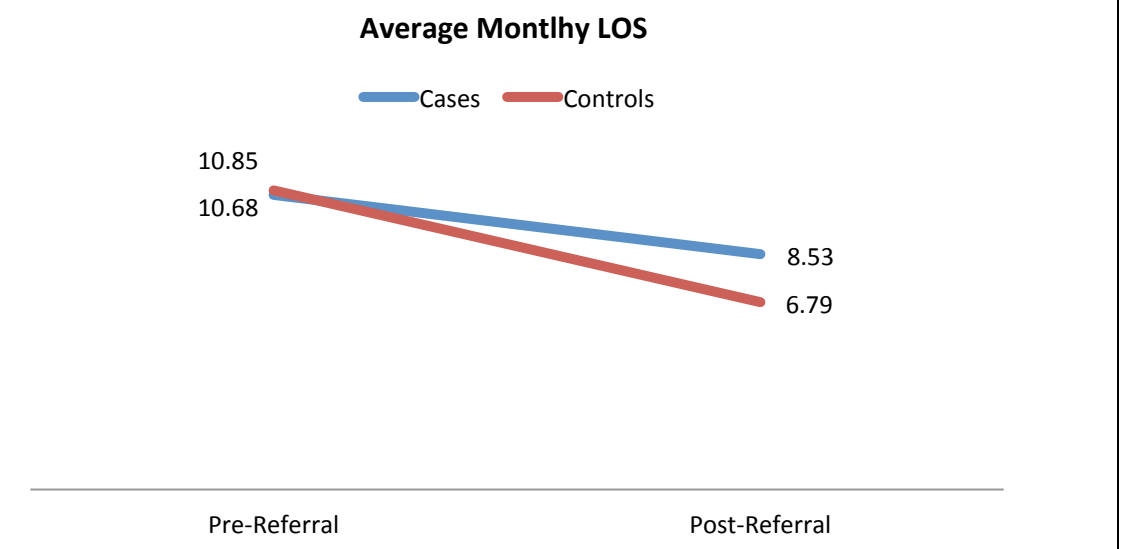


	Pre-Referral	Post-Referral	Median Change Within Group	*p*
Cases	1.83	6.57	3.08	**0.010**
Controls	3.69	3.11	-0.23	0.846
Median Difference Between Groups	-0.68	3.78		
p	0.492	**0.014**		

** Complete results in Tables 9,10, 11 and 12.

Despite the higher number of admissions found in the post-referral period in the CPCH arm, the LOS were not notably affected. Figure 16 summarizes the results with respect to **monthly LOS per patient**. The comparison between groups (controlling for disease and age at death) showed that CPCH patients presented with longer LOS before and after the program, however, did not reach a level of significance. When patients were compared to themselves in the pre- and post-referral periods, PPCP users showed a slight reduction in LOS. The control group had an inconsistent pattern.

Figure 16. Average Monthly LOS: Summary of Comparison Between Groups and Changes within Groups from Pre- to Post-Referral Period **



	Pre-Referral	Post-Referral	Changes Within Group	*p*
Cases	10.68	8.53	-1.6	0.432
Controls	10.85	6.79	0.17	1
Median Difference Between Groups	1.91	3.66		
p	0.432	0.308		

** Complete results in Tables 14, 15, 16 and 17.

A time series better demonstrates the trends in both groups showing that they had similar LOS until approximately 15 months before death. Outpatient visits decreased while inpatient admissions increased towards death in both groups, with CPCH group utilizing more health care resources. CPCH group consistently had more non-critical admissions than BCCH group, from 15 to 2 months prior to death. In the last 2 months of life patients in program users group became more severely ill than their matches and had a higher monthly LOS in critical care beds. These exploratory analyses show an opposite trend compared to the changes observed in the aggregate data when patients were compared to themselves in the pre- and post-referral periods. It may have occurred

due to the limitations of the data and referral to the program less than a month before the death occurred.

Despite non-significance in the difference between groups, analysis of LOS per facility demonstrated an important shift in health care setting from hospital to hospice, more substantially so in the non-cancer subgroup.

It is important to note that admissions of any acuity level to hospice are not completely funded by the publicly funded health care system. Historically, over 10 years only approximately 26% of the hospice operating cost is funded by government initiatives.[33†] However, the hospice operation directly affects the health system since each admission to the facility (or managed by the PPCP team in home care) avoids inpatient admissions at the hospital. The impact on costs from this shift in health care utilization will be discussed in Chapter 4.

When the **last admission before death** was separately analyzed, no significant difference was found between groups in LOS or use of invasive procedures to prolong life. However, the CPCH group tended to have a lower median difference between pairs in LOS (3.25 days), and lower absolute numbers of invasive procedures. A large difference in the LOS within pairs was observed, which made the number of invasive procedures between cases and controls incomparable.

3.4.2 Limitations of this Study

The first limitation of the study was the target population. Since only patients who died either at the BCCH or at CPCH were included in the study, an important portion of PPCP patients who opted for a home death or who died in other facilities was excluded (approximately 50% of the

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patients enrolled in the BC PPCP every year[53‡]). The decision to omit these patients was made due to the lack of an accessible registry of controls who died at home or other facilities. This excluded population might have presented a different behavior in terms of health care resource utilization. The power to detect any difference between groups with only 11 pairs of patients is only 6%. In order to recruit a larger number of patients using the same matching criteria would require a multicenter study with other similar settings of a hospice-based PPCP or a database with longer observational period. The small sample size also limited the use of parametric tests due to the great interference of outliers.

The matching criteria were also a challenge for this study. It is unclear when the goals of care shift from curative to palliative, and therefore when a child should be "enrolled" in PPCP. This uncertainty did not allow for a precise starting point from which comparisons should begin leaving the "death" as the indicator of potentially benefits from receiving PPCP. Furthermore, classifying the main disease that led to death has been traditionally done using ICD codes. ICD codes do not cover the total range of rare and terminal diseases and conditions, resulting in cases regularly being assigned unspecified codes. An attempt to use the most specific ICD code level (4 digits) failed to produce any matches in the target population, while the 3-digit level ICD code failed to find cancer matches. This might indicate that different diseases are occurring in different groups, inappropriate use of ICD codes, or that ICD codes are not specific enough to describe the diseases occurring in this population. When an alternate matching process was applied for cancer patients, based on the description of disease by the physicians in the cancer registry, roughly the same proportion of pairs as in the non-cancer group was found. Perhaps applying the same strategy for non-cancer patients, instead of using ICD codes, approximately twice as many patients would have been matched.

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Combining the ICD code criterion with age at death might have had further impact on the small sample size. Although not tested within the current data, 2 hypotheses may exist: children may have died from the same diseases but at different ages across groups; or children may have died at approximately around the same age in both groups but from different diseases. A population database is required to answer these challenges, by enabling comparisons between pediatric deaths enrolled in the PPCP to those who never enrolled, regardless of place of death.

Selection bias is a concern in any observational study, especially in PPCP studies where it is very likely to occur, affecting the results. As described previously, the enrollment criteria for PPCP depend on both referral to and acceptance of the program. Barriers to families' enrollment in programs are still a subject of inquiry and may be encompassed of a combination of factors such as culture, beliefs, values, previous experiences, and family readiness to acknowledge an incurable condition of a child.[54,55] How this self-selection influences the direction of the results is an important topic for future research within this field.

In this study, children in the CPCH arm started to enroll in PPCP 12 months before death, with a maximum enrollment rate of 67%, even in the last month of life. This may have been due to some children enrolling only a few days before death or due to their age (i.e. younger than 30 days). Before this 12-month period prior to death, all children belonged to the same group (usual care), and gradually, some migrated to the PPCP arm. Interestingly, approximately 3 months prior to program enrollment, a higher trend in inpatient admission started among children in the CPCH group. It might be hypothesized children referred to the program were more critically ill than their matches, despite both of them have progressed to death at roughly the same time. However, there is no formal indicator or score for "severity of disease" with which to test for any such systematic differences between groups. Therefore, the pairwise analyses separating pre- and post-referral

periods indicated that it is more appropriate to compare between groups rather than use aggregate results.

The estimates of LOS may have been overestimated due to 2 factors: age and measurement. The inclusion of newborns who spent their entire (short) life in ICU led to a very high estimate of monthly LOS in both arms due to the method of calculation (number of days spent in hospital divided by observational period in months). This resulted in higher mean estimates as it was assumed that if they were alive for 30 days, they would have spent those days in ICU. Additionally, measurement of LOS in the current administrative system did not take into account transfers between units (e.g., ward to critical care) and assigned a 1-day LOS in each unit, despite patients only being a fraction of the day in each unit. However, the results of the non-parametric tests should not be influenced by these challenges, as the median is not sensitive to extreme observations.

From another standpoint, the trends observed in the time series graphs and the aggregate data analysis may seem controversial due to possibilities. First, the way the data was treated to assign the admissions to pre- or post period may have affected the results. For instance, if an admission started before the referral date but continued after the enrollment on the program, this admission (and all its LOS and cost) was assigned to the pre-referral period, which could result in a higher LOS than the number of days in the pre-referral period, and an inflation of the outcome measurements. Second, the fact that 36% of the cases were referred to the program less than 1 month before dying, added to the small sample size, resulted in a very short post-referral period for those cases. These reasons may explain why there is an upward trend in the time series in both groups but when the patients in each group were compared to themselves, the changes from pre- to post-referral period were downward.

Moreover, considering the variance found in LOS f (sd = 10.4 days, Table 24), to detect a minimum effect size of 1 day/month/patient difference in LOS between groups, this study of 11 pairs has a power of 6% ($\alpha = 0.05$). It means that those comparisons that failed to find significant differences between groups not necessarily proves that the groups were similar, but might have been due to the low power of this study to detect true differences if they existed. The ideal comparative cohort study would have needed 853 pairs of children ($\alpha = 0.05, \beta = 0.80$). Since the population of children who dyes from LTC is naturally small (i.e. approximately 210-250 deaths per year due LTC [18,32]) a longer study or a multicenter collaboration with other similar settings of hospice-based PPCP would be required to reach the ideal sample size for another matched study. This study is the first of its kind to provide information on the variability of the outcomes, with a standard deviation based on a small sample with extreme observations. Using a large standard deviation only make the estimate of ideal sample size more conservative, which should be considered when planning future studies.

Finally, our statistical analyses had some limitations with respect to the risk of Type I error occuring, when the study can find significant differences between the groups but when in actuality there is none. This is due to the multiple tests performed in the same dataset. For further studies, this can be controlled using a Bonferroni correction to adjust the significance level according to the number of tests included in the analysis. It was not performed in this study as it further decreases the power, which it was proved to be very low.

3.5 Conclusions

The only significant difference between groups was the greater number of admissions in the CPCH group in the post-referral period, which was still possible to be seen by chance. Children who

were referred to the PPCP presented an increased number of monthly admissions, especially in critical care towards death (compared both to their controls and to themselves in the period prior to referral).

Despite this higher number of admissions, monthly LOS per patient was not affected in the aggregated data. Although a tendency for longer LOS in the CPCH arm was shown, no significant differences between the groups were found overall, neither in the pre- or post-referral periods, which might be attributed to the lack of research power to detect true differences due to the small sample size.

However, the exploratory analysis better display the trends. Over time, both groups had similar LOS until approximately 15 months before death. From 15 to 2 months prior to death, CPCH patients consistently had more non-critical admissions than their matches and, in the last 2 months of life, became more severely ill with a higher monthly LOS in critical care beds. This in itself might have led to the referral and acceptance of the PPCP.

Despite the need for critical care, a shift in health care setting utilization from hospital to hospice was observed, both in number of admissions and LOS. This was more evident in the non-cancer subgroup, which has been shown to have longer length of enrollment in PPCP.[9]

Regarding the EOL admission, no significant difference was found between groups in LOS or use of invasive procedures to prolong life. However, the CPCH group tended to have a lower median difference between pairs in LOS (3.25 days), and lower absolute number of procedures.

These results should be interpreted with caution due to the limitations of a small sample size and a selection bias inherent in the enrollment in PPCP. Similar matched studies would require a multicentre study with other similar settings of a hospice-based PPCP or a long time period.

Future studies that include severity of disease and the starting point from which children should be referred to a PPCP can shed light on the systematic differences between children who enroll in PPCP before death and those who die under usual care. Determining a “ground zero” for palliative care referral is fundamental to allow for unbiased comparison between groups and detangle possible confounding factors. Meanwhile, the following chapter estimates the impact on cost of the observed shift in health care setting utilization from hospital to hospice, from a publicly funded health system perspective.

Chapter 4: Cost Evaluation: a Matched-Cohort Analysis and Budget Exercise

Overview

The health care utilization analysis demonstrated a shift in admissions from hospital to the hospice. This chapter provides a comparative analysis of the overall costs of those admissions, and whether there was any impact on BC's publicly funded health system caused by this shift in health care setting. Average cost per day for each different type of admission was obtained and applied to the utilization observed from the cohort study described in the previous chapter. Differences in costs between PPCP and usual care groups were examined pre- and post-referral to the program, along with changes in costs within each group in their own disease trajectory. Then, the costs of all inpatient care provided by the hospice were replaced by the costs of accessing similar care at the hospital and the groups were examined again for differences, assuming this change could actually be made. Finally, the impact on the cost of all inpatient care provided by the hospice on an annual basis (beyond the cohort subjects) were simulated as if they would have occurred at the hospital.

4.1 Objectives

- To compare the cost of admissions of children living in BC who died from a LTC and were enrolled in the PPCP to those who died under usual care.
- To assess to what extent inpatient care provided by the hospice impacted the cost of admissions for children enrolled in PPCP within the cohort study and beyond.

4.2 Methods

4.2.1 Definitions

A number of terms are used in this chapter that must be defined for the reader at the outset, as per Table 24.

Table 24. Term Definition for Cost Analysis

Term	Definition
Cases	Children enrolled in the BC PPCP who died between Jan-2008 and Dec-2012 from a LTC.
Controls	Children who died at BCCH between Jan-2008 and Dec-2012 and were never enrolled in the BC PPCP.
Hospital	BCCH facilities including newborn admission areas, neonatal intensive care unit at BC Women's Hospital, and BCCH outpatient clinics and laboratories.
Hospice	CPCH facility.
Functional centre (or cost centre)	Accounting term to organize/segregate costs for management and control purposes.
Overhead Costs	Accounting term for those functional centres that serve many different final departments (e.g. administration, facilities maintenance, power, etc.) [56]
Final Departments (or cost pools)	The ultimate unit or department that provides services to the patients, for which the overhead costs are going to be proportionally distributed.
Final Units of Costs	Results of the distribution of each final department costs (including proportional overhead costs) by the number of services that final department provides. In this study they are the average cost of a bed per day (for inpatient admissions), and the average cost per visit (for outpatient and emergency visits).
Direct Cost	Full cost of inpatients services, including overhead costs, but excluding physicians' expenses (e.g. human resources, diagnostic, laboratory services, administration, support expenses). It is based on costs reported in functional centres related to inpatient services within the hospital or hospice operating cost report and divided by the qualifying inpatient days reported by each provider.
Partial Direct Care Costs	Costs related to the direct care outpatient visits and emergency admissions, without overhead costs and physicians compensation, divided by the number of visits reported.
Cost Allocation	Allocation method to determine the final unit of cost.
Direct Allocation	Cost allocation that ignores interaction of overhead costs, and allocated directly to final functional centres. [56]

Term	Definition
Step-down Allocation with Iterations	Cost allocation that makes full adjustments for interaction of overhead costs, and allocated to other overhead costs and final functional centres in a stepwise fashion to eliminate residual costs.
Residual Costs	Costs that are not fully distributed to final departments in the cost allocation process.
Critical Care Admission	Admissions requiring critical care attention independent of the setting where it was provided. Admissions occurred at BCCH in the NICU or PICU, and those occurred at CPCH classified as levels of acuity 4-5. These levels of acuity are equivalent to admissions to the NICU/ PICU at BCCH, based on nursing workload and patients symptoms. CPCH team developed the Canuck Place Nursing Workload Measurement Tool and Acuity Scale that has been used for over 10 years, published in Siden et al. [9]
Non-critical Admission	Admissions not requiring critical care attention independent of the setting where it was provided. Admissions occurred at BCCH in any ward (not NICU/PICU), and at CPCH classified as levels of acuity 1-3.
Combined System	A system that provides emergency, inpatient and outpatient care for LTC children with palliative care needs, with a combination of hospital and hospice facilities. In this study it a description of what really happened in the BC PPCP context.
Single System	A system that provides emergency, inpatient and outpatient care for LTC children with palliative care needs by the hospital facility, not including the hospice facility. In this study it is a simulation of the costs applying the hospital costs to the corresponding hospice admissions observed among the cases, or to the entire inpatient population at the hospice.

4.2.2 Study Design

A retrospective matched-cohort was designed as described in Chapter 3 (3.2.1 – 3.2.4, 3.2.6-3.2.8). Admissions that occurred in the observational period of the cohort study were assigned an average daily cost, in 2011-2012 Canadian dollars (per type of admission and facility were it occurred). A partial perspective of direct costs to the publicly funded health care system was adopted, based on the Canadian Agency for Drugs and Technologies in Health (CADTH) guidelines,[57] meaning that some but not all direct costs were included. A complete perspective of direct costs was not possible due to lack of data on '*Direct costs to publicly funded services (other than health care)*', '*Time costs to patients and their families*', and some '*Direct costs to patients and their families*' such as community-based services, rehabilitation, and aids and appliances provided outside of BCCH and CPCH.

4.2.3 Final Units of Costs

The final units of cost were classified per type of admission and facility where the admission occurred. All the possible final units of costs are presented in Table 25. Children under the PPCP could access inpatient care both in the hospital and in the hospice post-referral. For the purpose of this study, this scenario with a combination of inpatient care both at the hospital and at the hospice was referred to as the 'combined system'. Each final unit of costs according to the type of admission and facility where it occurred was applied to the utilization observed in both groups of the cohort study and then statistical analysis was conducted.

Table 25. Type of Admissions and Facility

Type of Admission	Facility of Admission
Outpatient	Hospital
Emergency*	Hospital
Inpatient: Non-critical Admission	
Ward	Hospital
Level 1	Hospice
Level 2	Hospice
Level 3	Hospice
Inpatient: Critical Care Admission	
NICU/PICU	Hospital
Level 4	Hospice
Level 5	Hospice

4.2.4 Characteristics of the Data, Source, Cost Components

The characteristics of the cost data for each type of admissions are summarized in Table 26. The Direct Cost for Inpatient Admissions both at the hospital and at the hospice was calculated with costs for inpatient admissions from the 2011-2012 fiscal year's operating cost without any distinction between cancer or non-cancer patients. This fiscal year was chosen since this is the most up-to-date data available from CIHI. The hospital costs were calculated by CIHI based on data from the pediatric general population[§], whereas for the hospice costs CPCH used data from an exclusive LTC population (n=153).[**] Although CPCH has more disease-specific data than BCCH, this is the only available source of information that includes overhead costs and details the applied methodology, thus a trade-off had to be made between the type of information available and the population on which the information was based.

Emergency admissions and Outpatient visits occur only at the hospital. CPCH does not provide those services and when children under the PPCP require emergency consultation, imaging

[§] Personal communication reproduced with permission from Canadian Institute for Health Information (CIHI) Canadian MIS Database (CMDB)

[**] Personal communication reproduced with permission from CPCH Finance Department

exams, and surgical procedures, patients are directed to BCCH. However, Direct Cost data on those types of admissions were not available from CIHI nor could be calculated by PHSA Decision Support due to the lack of overhead cost data. Alternatively, PHSA provided the partial direct care cost[58††] for those admissions based on data retrieved exclusively from the 22 children included in both groups of the cohort study, specified by cancer and non-cancer subgroup. These costs were those incurred when the admissions occurred (2008-2012), without overhead costs, and were not adjusted for inflation to make them all comparable to the 2011-2012 fiscal year dollars. All costs were presented in Canadian dollars.

Table 26. Characteristics of the Cost Data per Type of Admission

Facility / Setting	**Hospital**	**Hospice**	**Hospital**
Type of Admission	Inpatient Non-critical and Critical Care	Inpatient Non-critical and Critical care	Outpatient and Emergency Visits
Type of Cost	Direct Cost (include overhead costs)	Direct Cost (include overhead costs)	Partial Direct Care Cost (no overhead costs)
Unit	Average Cost per Day	Average Cost per Day	Average Cost per Visit
Source	CIHI	CPCH	PHSA
Population	General Pediatric Population	LTC Hospice Population (n=153)	LTC cohort (n=22)
Subgroup Specificity	Nonspecific	Nonspecific	Specific Cancer and Non-cancer subgroups
Dollar-Year	2011-2012	2011-2012	Real Cost 2008-2012

Details of the included cost components and cost allocation methodology are provided in Table 27. The calculation of the cost per visit for Outpatient and Emergency visits was named Partial Direct Care Costs because this primarily included drugs, supplies, and clinician costs directly

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involved in the care, except for physicians. No overhead costs such as housekeeping, food service, or facility maintenance were included. PHSA internally identified the costs for the visits from those observed in the cohort study (reported in Chapter 3) and calculated the average cost per visit following the steps described in the cost allocation process column of Table 27. PHSA did not specify how the costs of clinicians and other non patient-specific costs were allocated to the departments. Further evaluation of the costs components was not performed because no access to the raw data was granted for this project. In the same manner, PHSA did not provide the standard deviations of the average costs per visits, which did not allow testing for significance.

To calculate the average cost per day per type of inpatient admission, CIHI applied their Methodology for Calculation of Inpatient Ward and ICU Hospital Per Diem Rates,[59] which used a step-down allocation with iterations. CIHI did not include among the cost components expenses due to research, long-term care, recovery revenues, amortizations, interests on major equipment loans and long-term liabilities. Further detail on the CIHI methodology is found in Appendix F.

Likewise, the CPCH financial department mirrored CIHI's methodology to determine the final departments and cost centres. For the hospice cost allocation, CPCH did not include expenses with research, recovery revenues, amortizations, or costs with fundraising activities. Once those cost centres were excluded, CPCH excluded a proportion of the operating costs from several overhead cost centres attributed to outreach activities (bereavement program, external consultations, telephone support for outpatients and families, camping activities, program intake, etc.). Therefore, the costs presented in the cost allocation are those exclusively related to inpatient care (for patients, parents and siblings). CPCH used a direct allocation method to proportionally distribute those costs according to the number of beds occupied during the fiscal year (by weighted acuity) and the number of family members staying at the hospice accompanying those children

during admissions. In the weighted acuity occupancy 1 bed-day in level 2 corresponds to 2 bed-days in level 1; 1 bed-day in level 3 corresponds to 3 bed-days in level 1; 1 bed-day in level 4 corresponds to 4 bed-days in level 1; and 1 bed-day in level 5 corresponds to 5 bed-days in level 1. This weighted system was created by the hospice because there is a limited human resource available per day, which caps the capacity of the hospice to provide inpatient care. A child classified as level 5 consumes 5 times the resources as a child classified as level 1. Using this weighted system, the operating cost for inpatient care was distributed proportionally to the number of beds, attributing higher costs to higher levels of acuity.

Some additional adjustments were made such as in regards to contracted services for Information Technologies (IT) provided to CPCH and paid by PHSA on the hospices' behalf. While this cost is provided to the hospice for 'free' by PHSA, it may be included in BCCH's overhead costs. Therefore, to net these systematic differences and minimize bias in the cost comparison, these costs were added to the CPCH cost components before the cost distribution to the final departments. Physician compensation was not included in any of the final units of cost.

Table 27. Cost Components Included in the Cost Allocation

Type of Cost	Source	Cost Components	Cost Allocation Method/Process
Partial Direct Care Costs	PHSA	Time spent on the unit (mostly nursing compensation and supplies); Outpatient visits; Emergency stay; Medical imaging costs; Professional services costs (e.g. physiotherapy, occupational therapy, social work, etc.); Surgical costs, including patient-specific supplies for anaesthesia, surgical supplies, and surgical implants; Patient-specific pharmacy costs. PHYSICIANS' EXPENSES NOT INCLUDED	1. Retrieved all dollars in a given department (emergency, outpatient); 2. Extracted/removed all patient-specific supply costs that were tagged to a patient 3. Averaged unit cost for each department per day; 4. Multiplied each patient's utilization by the appropriate unit cost (per type of admission); 5. Added back the patient-specific costs; 6. Average unit cost for each department per day (including patient-specific costs).
Direct Cost	CIHI	Full operating cost of inpatient services (extensive list of functional centres listed in Appendix F) <u>Excluding costs from the following functional centres:</u> research costs, long- term care costs, recovery revenues, amortizations, interests on major equipment loans and long-term liabilities PHYSICIANS' EXPENSES NOT INCLUDED	CIHI Methodology for Calculation of Inpatient Ward and ICU Hospital Per Diem Step Down Allocation with Iteration (Appendix F)
Direct Cost	CPCH	Full operating costs of inpatient services Clinical Care (no physicians), Recreational Therapy, Counselling Team, Food Services, Housekeeping, Facilities Maintenance, Professional Education and Development, Executive Team, Finance Department, Information Systems, Volunteer Coordination, Human Resources Management, <u>Excluding costs from the following functional centres:</u> research costs, recovery revenues, amortizations, costs with fundraising activity PHYSICIANS' EXPENSES NOT INCLUDED	Direct Allocation to Final Departments

PHSA: Provincial Health Services Authority; CIHI: Canadian Institute for Health Information; CPCH: Canuck Place Children's Hospice; ICU: intensive care unit

4.2.5 Hypothesis

Since the systematic review of the literature (Chapter 2) revealed a high heterogeneity of health care resource utilization and costs between children who accessed PPCP to those under usual care, a hypothesis was made that in the local data comparison, cases and controls would present differences in costs. However, no speculation on the direction of these differences was made, and a 2-sided test was performed in the statistical analyses.

4.2.6 Sample Size

As previously discussed and presented in the utilization analysis, recruiting a larger sample at the time of this study was unfeasible irrespective of any sample size calculation, since we used the entire population of LTC children who died at the hospice. Only 11 pairs were found. While a challenge for the current study, this sample provides information about the variability of the outcomes, which plays an important role in such sample size calculations and may provide useful guidance for the planning of future related studies. Thus a related calculation was performed based on the standard deviations of costs found in this study and is presented after the results. It determines how large the mean difference in costs would have to be, to have a reasonable chance of detecting it with this study of 11 matched pairs. Further, the ideal sample size to achieve a certain power to detect differences in costs between groups was estimated.

4.2.7 Statistical Analyses – 'Combined System'

After applying the final units of costs (per type of admission and facility) to the health care utilization data from the cohort study designed in Chapter 3, costs were displayed in time series graphs to observe the cost trend in both groups. As observed in the health care utilization study

(Chapter 3) the aggregate comparison might be misleading. Therefore, similar to the health care utilization analysis, a comparison between groups before and after the "referral point" was presented. In the control group, the referral period was determined individually by applying the LOP of their respective cases. Changes from pre- to post-referral period were analyzed separately for each group to understand its disease trajectory (accounting for any systematic selection bias in PPCP enrolment).

Mean values and standard deviation of monthly cost were presented for future research purposes, budgeting and economic models. However, the estimates for the differences between groups were tested with pairwise analyses to account for disease and age, based on the median values. Whenever possible, subgroup analysis for cancer patients was presented. An exploratory analysis by health care setting was presented.

A Wilcoxon signed-rank test was applied to account for the limitations discussed in Chapter 3 (mainly related to small sample size). The null hypothesis stated the median difference in costs between pairs is zero. An $\alpha = 0.05$ was chosen to determine significance, a 2-sided test was employed to assess significance in both directions, and 95% confidence intervals (CI) were calculated. All statistical analyses were conducted in R software© (Version 0.98.5072009).

4.2.8 Simulation Exercise – 'Single System'

For the purpose of this research, a scenario where inpatient admissions can only be accessed through the hospital was named the 'single system'. To simulate the cost of providing inpatient care for the cases in a single system, assuming their admissions would have occurred exclusively at the hospital with similar levels of acuity (CPCH Level 4/5 = NICU/PICU =; Ward = Level 1-3), the respective costs from BCCH were applied to their utilization post-referral, replacing the

respective costs at the hospice. The statistical analysis was reapplied as previously described to test changes between groups and within the CPCH group. To simulate the cost of inpatient care in a single system to all children who accessed hospice inpatient care in the same fiscal year, the same procedure was applied to the hospice total capacity in the same period. All inpatient care provided at the hospice were priced with the BCCH equivalent cost, beyond the cohort study subjects, and a cost impact to the publicly funded health care system was presented.

4.3 Results

4.3.1 Final Units of Cost

4.3.1.1 Outpatient and Emergency Visits

As shown in Table 28, in the cancer subgroup, cases presented higher average partial direct care cost per visit than their controls for both outpatient ($362 vs $323) and emergency visits ($111 vs $56). Conversely, in the non-cancer group, cases had lower average costs per outpatient visit than controls ($87 vs $183). The average cost per emergency visit among non-cancer cases was $79, and no data among non-cancer controls from the cohort study was found.

Cancer patients had higher partial direct care cost of outpatient visits than non-cancer patients in both groups. The differences between groups and sub-groups could not be statistically tested due to the lack of standard deviation.

Table 28. Outpatient and Emergency Visits Final Units of Costs – Average Cost per Visit*

Type of Admission	Hospital	
	Cancer Patients	
	CPCH Group	**BCCH Group**
Outpatient	$362	$323
Emergency	$111	$56
	Non-Cancer Patients	
	CPCH Group	**BCCH Group**
Outpatient	$87	$183
Emergency**	$79	NA

* Partial Direct Care Cost, source PHSA; # Data source: PHSA, data from cohort subjects (n= 22); ** No data for Emergency Visits for non-cancer patients from the cohort study was found; NA: not available/not found

4.3.1.2 Inpatient Admissions

Table 29 summarizes the final units of costs for the inpatient admissions per type and facility. CIHI provided the costs for admissions that occurred at the hospital. The average cost per bed day for non-critical admission was $2,912, and for critical admissions was $4,281.[60‡‡] Complete methodology for the cost allocation used by CIHI is described in Appendix F.

CPCH provided the costs for admissions that occurred at the hospice.[61**] The average cost per bed day for non-critical admissions was lower at the hospice (range from $612 to $2,033). Similarly, critical care admissions had lower costs (from $2,750 to $3,888). The difference in costs between settings could not be tested due the lack of standard deviation in both groups.

‡‡ Personal communication reproduced with permission from CIHI CMDB

Table 29. Inpatient Admissions Final Units of Costs (Facility and Type of Admission) – Average Cost per Day per Bed*

Type of Admission	Hospital #	Hospice**
	Inpatient: Non-critical Admission	
Ward	$2,912	-
Level 1	-	$612
Level 2	-	$1,354
Level 3	-	$2,033
	Inpatient: Critical Care Admission	
NICU/PICU	$4,281.00	-
Level 4	-	$2,750
Level 5	-	$3,888

* Direct Cost; # Data source: CIHI, data from general pediatric population cared for in 2011-2012; ** Data source: CPCH cost distribution, data from all LTC inpatient care in 2011-2012 (n= 153).

The details of the hospice operating cost allocation are presented in Table 30. CPCH provided 2192 bed-days of inpatient care in the 2011-2012 fiscal year. According to the weighted acuity, 24% of those beds were critical care admissions (level 4-5) and 76% were non-critical care admissions (level 1-3). Using this weighted system, the operating cost for inpatient care was distributed proportionally to the number of beds, attributing higher costs to higher levels of acuity. On average, there were 6 children per day and 7.8 family members staying at the hospice for inpatient care. The operating costs with inpatient care were allocated to the number of beds provided and the number of family members in the hospice, except for the cost associated with clinical care. This last cost centre was exclusively allocated to the inpatient care beds since they are not related to the care or accommodation provided for families accompanying the patients. The total cost to provide inpatient care in the hospice was $3,888,578 for the children and $1,246,272 for the families (siblings, parents and extended family).

Table 30. Canuck Place Cost Components and Cost Distribution – Direct Cost (assembling CIHI Methodology)

OPERATING COSTS - 2011/12 INPATIENT CARE				CANUCK PLACE ALLOCATION OF COSTS								
				CHILDREN						TOTAL	TOTAL	TOTAL
		WORKLOAD MEASURE		Total	Level 1	Level 2	Level 3	Level 4	Level 5	CHILDREN	FAMILY	PATIENT CARE
		Actual Bed Day Usage		2192	254	689	918	311	20			
	2011/12	Weighted by Acuity Level		5730	254	1378	2754	1244	100			
	ACTUAL COSTS $	OCCUPANCY (weighted acuity)		100%	4%	24%	48%	22%	2%	**6.01** ***per day****	**7.82** ***per day****	
1. DIRECT PATIENT CARE		**ALLOCATION BASIS**										
Clinical Care	2,931,077	57%	*Children - weighted acuity*		117,243	703,459	1,406,917	644,837.00	58,622	2,931,077		**2,931,077**
Physicians (Funded through APP)	-	0%	*Children - weighted acuity*		-	-	-	-	-	-		**-**
Recreation Therapy	95,015	2%	*family/child ratio*		1,651	9,908	19,816	9,082	826	41,282	53,733	**95,015**
Counselling Team	221,118	4%	*family/child ratio*		3,843	23,057	46,115	21,136	1,921	96,072	125,046	**221,118**
Food Services	306,018	6%	*family/child ratio*		5,318	31,910	63,821	29,251	2,659	132,959	173,059	**306,018**
Housekeeping	178,636	3%	*family/child ratio*		3,105	18,627	37,255	17,075	1,552	77,614	101,022	**178,636**
Facilities	284,445	6%	*family/child ratio*		4,943	29,661	59,321	27,189	2,472	123,586	160,859	**284,445**
Total Direct Patient Care	**4,016,310**	**78%**			136,104	816,622	1,633,244	748,570	68,052	3,402,592	613,718	**4,016,310**
2. INDIRECT PATIENT CARE												
Research and Education	113,666	2%	*family/child ratio*		1,975	11,853	23,705	10,865	988	49,386	64,280	**113,666**
Executive	289,769	6%	*family/child ratio*		5,036	30,216	60,432	27,698	2,518	125,899	163,869	**289,769**
Finance	198,758	4%	*family/child ratio*		3,454	20,726	41,451	18,999	1,727	86,357	112,401	**198,758**
Information Systems	197,759	4%	*family/child ratio*		3,437	20,621	41,243	18,903	1,718	85,923	111,836	**197,759**
Volunteer Services	146,598	3%	*family/child ratio*		2,548	15,287	30,573	14,013	1,274	63,694	82,904	**146,598**
Human Resources	171,991	3%	*family/child ratio*		2,989	17,934	35,869	16,440	1,495	74,727	97,264	**171,991**
Amortization	-	0%	*family/child ratio*		-	-	-	-	-	-	-	**-**
Total Indirect Patient Care	**1,118,541**	**22%**			**19,439**	**116,637**	**233,273**	**106,917**	**9,720**	**485,986**	**632,554**	**1,118,541**
TOTAL PATIENT CARE	**5,134,850**	**100%**			**155,543**	**933,259**	**1,866,517**	**855,487**	**77,772**	**3,888,578**	**1,246,272**	**5,134,850**
		Costs per bed day			**$612.37**	**$1,354.51**	**$2,033.24**	**$2,750.76**	**$3,888.58**	**$1,773.99**		

Once the final units of cost per type of admission and facility were found, those units were applied to the utilization observed in the cohort study to obtain the total costs incurred in the observational period, observe the cost trend overtime and compare groups pre- and post-referral to the PPCP.

4.3.2 Cost Comparison – 'Combined System'

In this section, costs for the 'combined system' are assessed. The time series graphs demonstrate the cost trends in each group and the level of enrollment in the PPCP (Figure 17 and 18). The dark and light blue lines represent the total cost per person-month at risk observed over time, in the CPCH and BCCH groups, respectively. The grey bars represent the observed proportion of patients enrolled in the PPCP per person-month at risk, over time.

Children in the CPCH group began enrollment in the PPCP from 12 months before death, with a maximum enrollment rate of 67% even in the last month of life. This finding may be due to children who enrolled only a few days before death or being younger than 30 days old.

Costs of outpatient visits were very steady and similar between groups. Similar to what we observed with the utilization comparison (Chapter 3.3.3), the groups were very similar until approximately 15 months prior to death when a drastic divergence in costs with inpatient admissions were observed between groups (approximately $9,000 person-month), which became even more heightened during the last 6 months of life. Both groups showed an upward trend in inpatient cost towards death, but CPCH patients seemed to be more costly during the last year of life, especially in the last 2 months prior to death due to critical care admissions.

Figure 17. Total Cost per Person-Month at Risk Over Time, by Group: Outpatient and Inpatient Admissions

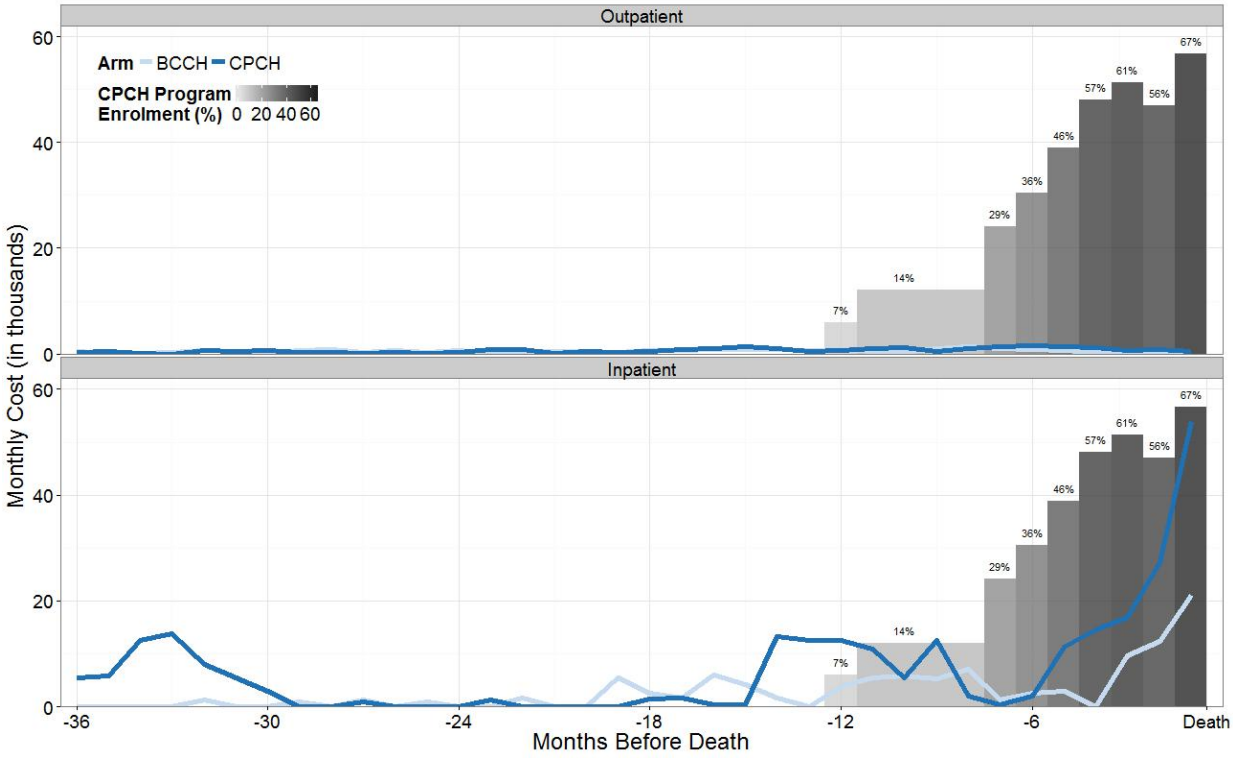
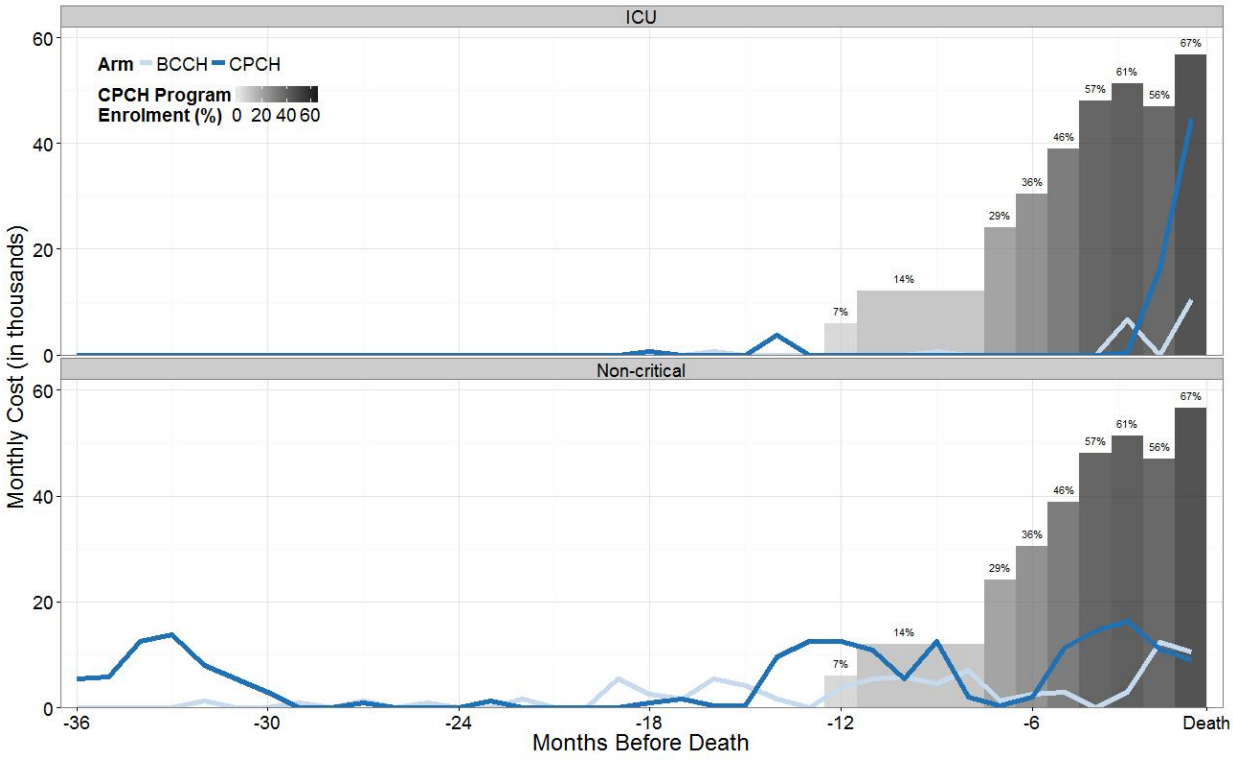


Figure 18. Total Cost per Person-Month at Risk Over Time Over Time by Group: Critical and Non-Critical Admissions



The pairwise analysis also showed a tendency in the same direction, with CPCH children having higher costs both pre- and post-referral compared to their pairs with and increased difference post-referral.

During the **pre-referral period** (Table 31), children in both groups were under usual care only. Overall, the average monthly cost per patient was $38,243 in the CPCH group versus $37,462 in the BCCH group. In the pairwise analyses (controlling for disease and age at death) children in the CPCH tended to have higher overall costs than their matches (median difference between pairs was $2,521), which seems to be driven by higher costs with inpatient care ($2,547), specifically critical care ($939). Conversely, CPCH children presented with lower costs in non-critical admissions ($374) and outpatient visits ($62) than their pairs. Costs with emergency visits were small due to the low number of visits observed in both groups. However, these differences in costs between groups did not reach the significance level, potentially due to the small sample size and consequently the low power of this study to detect true differences if they existed.

Table 31. Monthly Cost per Type of Admission, and Pairwise Test (Wilcoxon) – Pre-Referral [##]

Pre-Referral						
	Cost /Patient/Month - Mean (sd)		Median Pairwise Difference			
				Confidence Interval		
Variable	CPCH	BCCH	Estimate	2.50%	97.50%	*p*
Total	38,243 (56,351)	37,462 (94,778)	2,522	-51,238	40,382	0.275
Outpatient	407 (553)	447 (685)	-62	-472	402	0.813
Emergency**	3.37 (4.13)	0.42 (0.89)	6	4	8	0.059
Inpatient Total	37,833(56,567)	37,014 (94,924)	2,547	-51,089	40,394	0.275
Inpatient: Critical Care*	33,797(58,695)	31,130 (86,804)	939	-64,585	55,856	0.529
Inpatient: General	4,035(7,236)	5,883 (9,495)	-374	-13,201	11,334	0.906

Costs expressed in dollars without decimals; CI: confidence interval; * CI of 95% was not achievable because the lack of data, so R calculated a CI 90% automatically; ** CI of 95% was not achievable because the lack of data, so R calculated a CI 80% automatically; CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); p: p-value; sd: standard deviation.

During the **post-referral period** (Table 32), children in the CPCH group started to access the services from the PPCP while those in the BCCH groups continued under usual care only. Overall, the average monthly cost per patient was $17,769 in the CPCH group versus $21,692 in the BCCH group. Although it could be incorrectly interpreted as CPCH having had lower costs, as outliers affect the averages, the pairwise analyses (controlling for disease and age at death) showed higher cost estimates in the CPCH group. The median difference between pairs was $4,270, higher in the CPCH group. This tendency of higher costs was consistent for all types of admissions compared to their matches, even for non-critical admissions ($8,715) and outpatient visits ($580). Again, it is worthwhile to mention that the study had a very low power to actually detect significant differences if they were present.

Table 32. Monthly Cost per Type of Admission, and Pairwise Test (Wilcoxon) – Post-Referral.

Post-Referral						
Cost /Patient/Month - Mean (sd)			Median Pairwise Difference			
				Confidence Interval		
Variable	CPCH	BCCH	Estimate	2.50%	97.50%	*p*
Total	17,769 (16,917)	21,692 (49,674)	4,270	-49,430	21,454	0.610
Outpatient*	678 (1,183)	347 (635)	580	10	1,266	0.093
Emergency						
Inpatient	17,085 (17,122)	21,341(49,705)	1,909	-54,265	28,899	0.834
Inpatient: Critical Care	9,885 (13,407)	15,984 (42,971)	3,153	-58,357	22,561	0.624
Inpatient: General**	7,200(9,767)	5,357 (10,810)	8,715	-20,667	20,081	0.855

Costs expressed in dollars without decimals; CI: confidence interval; * CI of 95% was not achievable because the lack of data, so R calculated a CI 90% automatically; ** CI of 95% was not achievable because the lack of data, so R calculated a CI 80% automatically; CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); p: p-value; sd: standard deviation.

When **comparing patients to themselves in the pre- and post-referral periods**, similar patterns between groups were found, with a drop in the monthly cost per patient in both groups post-enrollment, however, these were non-significant and in bigger proportions in the CPCH group (Table 33).

Overall, the median decrease observed in the BCCH group was $74. This tendency of lower monthly costs was observed for all types of admissions: outpatient visits $49,61; critical care admissions $2,398.30; and non-critical admissions $1.195.80 (Table 33).

Table 33. BCCH Group: Changes in Monthly Cost per Type of Admission and Pairwise Test (Wilcoxon) – Pre-Post-Referral to PPCP[§]

Changes from Pre- to Post-referral BCCH Group						
Cost /Patient/Month - Mean (sd)			Median Pairwise Difference			
				Confidence Interval		
Variable	Before	After	Estimate	2.50%	97.50%	*p*
Total	37,462 (94,778)	7,165 (12,738)	-74	-141,234	14,222	0.846
Outpatient	447 (685)	382 (659)	-49	-306	214	0.286
Emergency	NA	NA				
Inpatient	37,014 (94,924)	6,778 (12,376)	-1,046	-151,721	18,409	0.834
Inpatient: Critical Care*	31,130 (86,804)	3,312 (9,443)	-2,398	-138,169	14,964	0.529
Inpatient: General	5,883 (9,495)	3,466 (9,282)	-1,195	-14,984	10,277	0.529

Costs expressed in dollars without decimals; CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); CI: confidence interval; p: p-value; ICU: intensive care unit admissions; sd: standard deviation; NA: not available/did not occurred; § Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth; * CI of 95% was not achievable because of lack of data. Software R automatically calculated a CI of 90%.

The median decline in monthly costs was more pronounced in the CPCH group ($8,846) specially in critical care costs ($23,579) which was partially compensated by an increase in the monthly costs of outpatient visits ($253) and non-critical admissions ($2,885) (Table 34).

Table 34. CPCH Group: Changes in Monthly Cost per Type of Admission and Pairwise Test (Wilcoxon) – Pre-Post-Referral to PPCP[§]

Changes from Pre- to Post-referral CPCH Group						
Cost /Patient/Month - Mean (sd)			Median Pairwise Difference			
				Confidence Interval		
Variable	Before	After	Estimate	2.50%	97.50%	*p*
Total	38,243 (56,351)	14,905 (14,659)	-8,846	-62,979	9,431	0.322
Outpatient	407 (553)	746 (1,225)	253	-187	1,531	0.272
Emergency	NA	NA				
Inpatient	37,833 (56,567)	14,152 (14,757)	-9,072	-62,974	9,002	0.275
Inpatient: Critical Care	33,797 (58,695)	8,650 (13,490)	-23,579	-87,239	5,183	0.529
Inpatient: General*	4,035 (7,236)	5,502 (8,402)	2885.20	-9,297	12,882	0.834

Costs expressed in dollars without decimals; CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); CI: confidence interval; p: p-value; ICU: intensive care unit admissions; sd: standard deviation; NA: not available/did not occurred; § Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth; * CI of 95% was not achievable because of lack of data. Software R automatically calculated a CI of 90%.

Table 35 displays the cost with inpatient care pre- and post-referral to PPCP, and shows that the shift in health care setting utilization from hospital to hospice transferred approximately half of the costs with inpatient care to this provider, more noticeably so in the non-cancer subgroup.

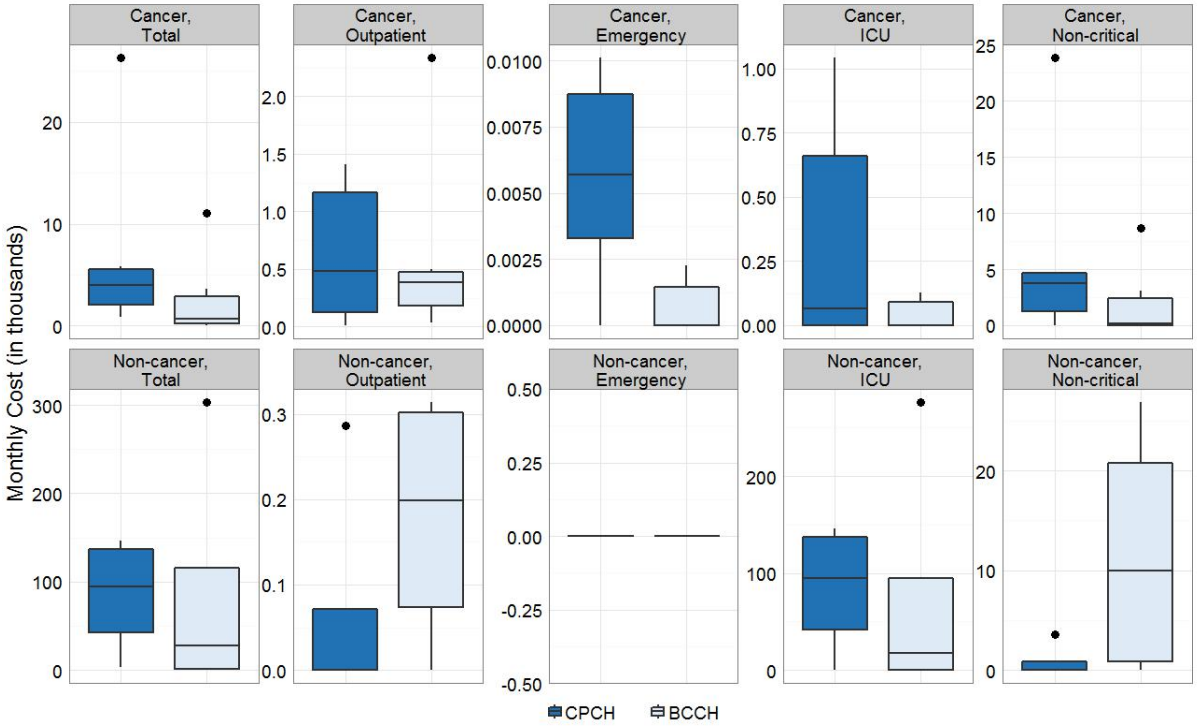
Table 35. Monthly Cost per Facility – Entire Study Period. §

Total Cost/Patient/Month by facility - Mean (sd) ##			
All			
Group	**CPCH Group**		**BCCH Group**
Facility	Hospital	Hospice	Hospital
Pre	38,243 (5,6351)		37,462 (94,778)
Post	11,354 (9,793)	14,497 (13,616)	39,769 (63,815)
Total Cost/Patient/Month by facility - Mean (sd) ##			
Non-cancer Patients			
Group	**CPCH Group**		**BCCH Group**
Facility	Hospital	Hospice	Hospital
Pre	84,956.01 (67,260)		89,626 (14,4467)
Post	23,933 (471)	20,788 (13,543)	68,441 (86,014)
Total Cost/Patient/Month by facility - Mean (sd) ##			
Cancer Patients			
Group	**CPCH Group**		**BCCH Group**
Facility	Hospital	Hospice	Hospital
Pre	7,102 (9597)		2,685 (4,306)
Post	6,323 (5749)	4,013 (4,274)	11,097 (17,780)

Costs expressed in dollars without decimals; CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); sd: standard deviation; § Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth.

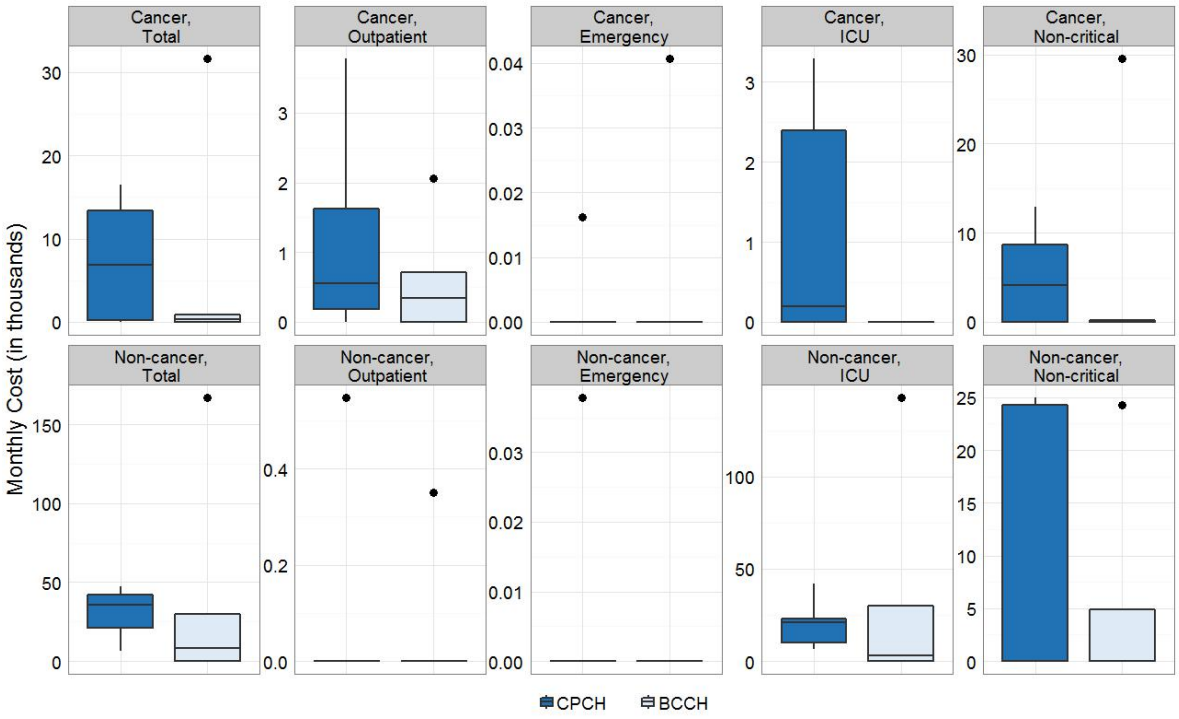
Figures 19-22 show that these results were driven by the costs from the non-cancer patients, who were substantially more costly than cancer patients. The individual pairwise analyses are presented in Appendix G.

Figure 19. Monthly Cost per Type of Admission, by Cancer and Non-cancer subgroup, Pre-Referral to PPCP. §



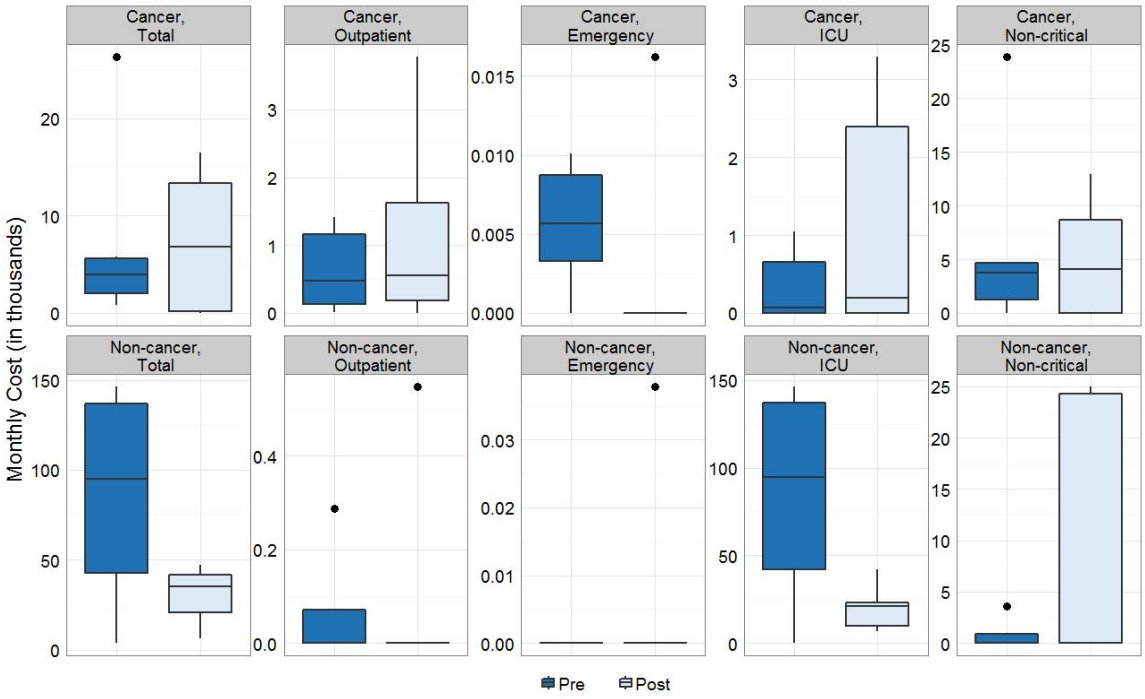
§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth.

Figure 20. Monthly Cost per Type of Admission, by Cancer and Non-cancer subgroup, Post-Referral to PPCP. §



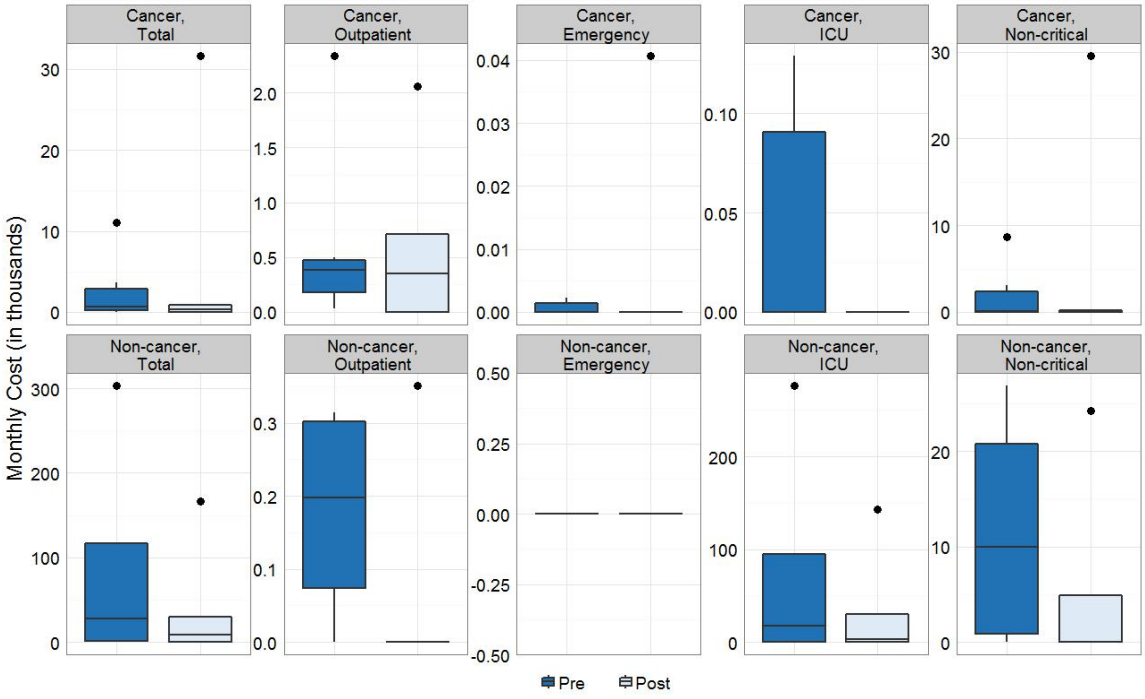
§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth.

Figure 21. CPCH Group - Changes in the Monthly Cost per Type of Admission, by Cancer and Non-cancer subgroup – Pre-Post-Referral to PPCP[§]



§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth.

Figure 22. BCCH Group - Changes in the Monthly Cost per Type of Admission, by Cancer and Non-cancer subgroup – Pre-Post-Referral to PPCP[§]



§ Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth.

In sum, both groups had an upward trend in cost from 15 months prior to death but the PPCP group presented higher costs compared to the control group even before the referral, especially regards to critical care admissions. If one consider the referral point as the landmark for when the treatment goals changed from curative to palliative, children in both groups presented a decrease in costs compared to their own previous period. This drop in costs was more pronounced in the CPCH group where a shift in health care setting to the hospice was observed (costs of critical care dropped and costs of non-critical admissions and outpatient visits increased). However, statistical tests of those differences did not find significance on those results, likely due to the small sample size and low power to detect differences if they were to be true.

The exploratory analyses showed an opposite trend compared to the changes observed in the aggregate data when patients were compared to themselves in the pre- and post-referral periods. It may have occurred due to the limitations of the data discussed in Chapter 3 and referral to the program less than a month before the death occurred.

4.3.3 Sample Size Calculation for Future Research

This study provides information on the variability of the costs among children who died from LTC, from a broader comparative approach to inpatient health care utilization (outpatient and emergency visits, hospital costs, and hospice costs). This is relevant to inform sample size calculations and may provide useful guidance for the planning of future related studies.

Using total inpatient costs after referral as the main outcome, estimates of effect size and power to detect differences between groups is presented, relevant to comparative research in the PPCP field. In the current sample (n=11 pairs), the mean difference between groups in monthly

total inpatient cost was $4,255 lower in the CPCH group, with a standard deviation of $42,904 (Table 36).

Table 36. Difference Between Groups in Total Inpatient Cost Post-Referral: Sample Mean and Variability.

Mean Total Inpatient Cost/Month ($) ##	
BCCH	21,692
CPCH	17,769
Difference	-4,255
sd	42,904

Costs expressed in dollars without decimals; CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); sd: standard deviation.

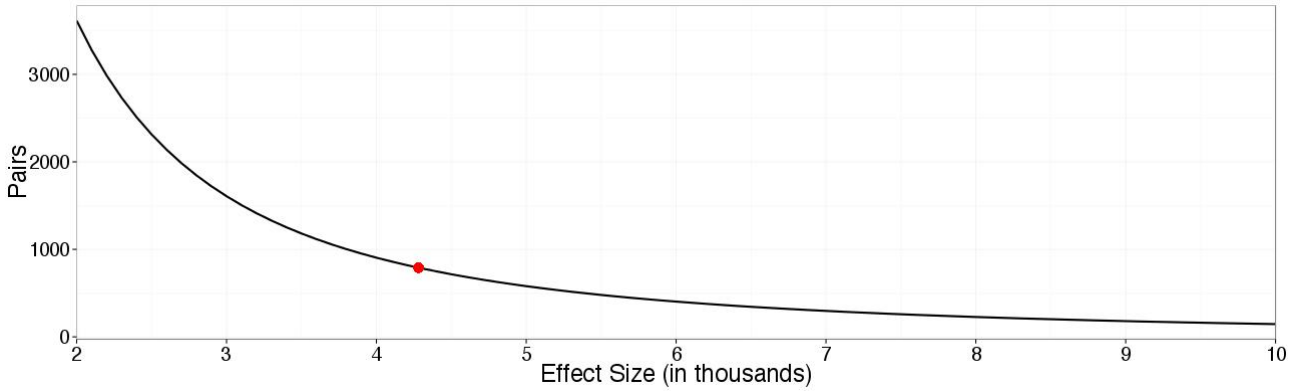
Arguably, from a health care provider perspective, the cost of 1 day in critical care per month could be viewed as the minimum difference needed to impact costs and operational processes. Considering the variance found in this study, to detect a minimum difference of $4,281/month/patient between groups, an ideal comparative cohort study would have to include 791 pairs of children, using a significance level of 5% and a Power of 80%. Therefore, the current study on 11 pairs under the same parameters only had a power of 6% to detect such effect size, with a 5% probability that any difference found occurred by chance (Table 37).

Table 37. Parametric Test's Sample Size Calculation.

Paired t-test Power Calculation (2-sided)	
Pairs Needed to Detect the Minimum Effect Size	Power of this Study to Detect the Minimum Effect Size
n = 791 α = 0.05 Power = 0.8	n = 11 α = 0.05 Power = 0.060
NOTE: n is number of *pairs*	

Figure 23 presents estimates of the different samples sizes required to test other effect sizes, considering the variability found in this study.

Figure 23. Sample Size Estimates for Different Effect Sizes – Total Cost



The only previous study demonstrating some variability in cost from similar perspective was Pascuet et al.[46] This study compared the patients before and after enrollment in a PPCP and found an average decrease in inpatient cost of $-4,252/month/patient and a standard deviation of $3,674.08 (n=66). Compared to that article, the PPCP group from this study (n=11) had a much larger variability with an average decrease in inpatient cost after enrollment of $23,757.94/month and a standard deviation of $49,904.17.

4.3.4 Single System Simulation - What if CPCH had not Provided Inpatient Care?

4.3.4.1 Within the Matched-Pairs Cohort Study

Assuming that in a single system all the admissions would have occurred exclusively at the hospital, the costs of inpatient admissions that occurred at the hospice were replaced by their respective costs from BCCH. The dashed black lines in the time series graphs (Figures 24,25) represent this new cost scenario. No significant changes in the cost trend line can be observed, except for the last month prior to death, where the black line and dark blue line (observed cost) slightly separate, more noticeably in costs of critical care admissions.

Figure 24. Direct Cost per Person-Month at Risk Over Time, by Group: Outpatient and Inpatient Admissions

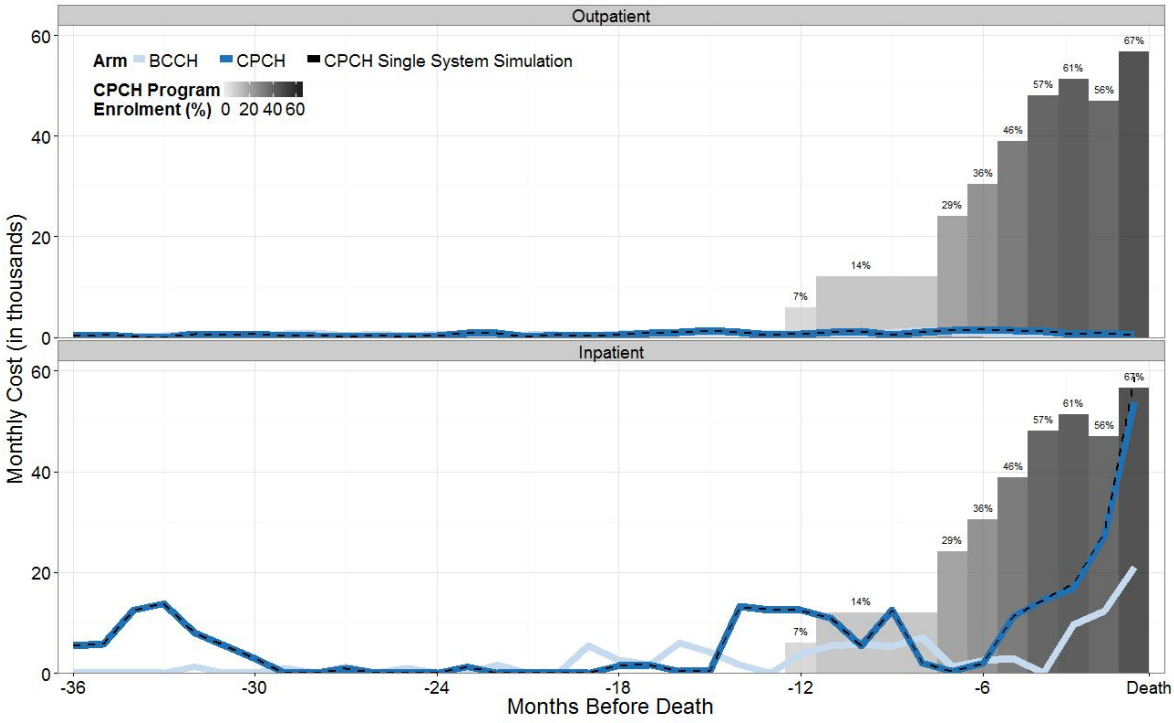
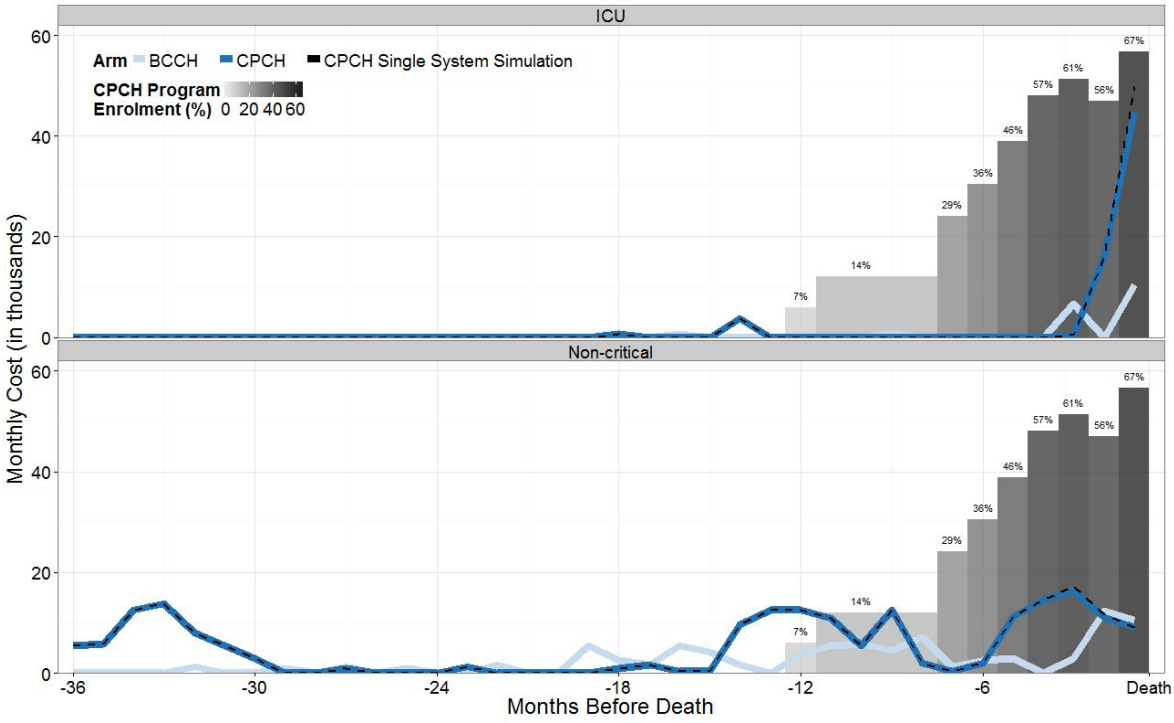


Figure 25. Direct cost per Person-Month at Risk Over Time Over Time by Group: Critical and Non-Critical Admissions



The differences in costs between CPCH and BCCH groups became larger in the pairwise analyses (controlling for disease and age at death), although still not statistically significant. In the **post-referral period**, the higher costs of children in the CPCH group would have been more pronounced in all types of admissions as compared to their matches (Table 38 vs Table 32). Overall, the median difference in monthly costs per patient would have been $7,098 higher in the CPCH group versus the previously observed $4,270. Likewise, the median difference in monthly costs for non-critical admissions would have been $10,206 versus the previous $8,715. Critical care admissions would have been $4,246 versus the previous $3,153.

Table 38. Monthly Cost per Type of Admission Simulating a Single System, and Pairwise Test (Wilcoxon) - Post-Referral Period.

Post-Referral ##						
	Cost /Patient/Month – Mean (sd)		Median Pairwise Difference			
				Confidence Interval		
Variable	CPCH	BCCH	Estimate	2.50%	97.50%	*p*
Total	23,514 (23,386)	21,692 (49,674)	7,098	-37,235	32,843	0.476
Outpatient*	678 (1,183)	347 (635)	579	9	1,265	0.093
Emergency	NA	NA				
Inpatient	22,830 (23,676)	21,341 (49,705)	6,593	-47,079	42,421	0.726
Inpatient: Critical Care	15,211 (20,968)	15,984 (42,971)	4,246	-51,980	35,006	0.529
Inpatient: General**	7,619 (10,198)	5,357 (10,810)	10,206	-20,667	20,922	0.584

Costs expressed in dollars without decimals; * CI of 95% was not achievable because the lack of data, so R calculated a CI 90% automatically; ** CI of 95% was not achievable because the lack of data, so R calculated a CI 80% automatically; CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); CI: confidence interval; p: p-value; sd: standard deviation; NA: not available/did not occurred

The average monthly cost per patient in the CPCH group would have been $23,514.46 versus the previous $17,857.34 (Table 39). This rise in costs in the CPCH group represents an average 32% increase in total costs driven by a 54% rise in critical care costs. The non-parametric test showed the median increase in costs across cases would have been $7,163.06 per patient per month.

Table 39. CPCH Group, Changes in Monthly Cost per Type and Pairwise Test (Wilcoxon) – Simulating a Usual Care Scenario in the Post-Referral Period

"CPCH Group - Changes in the Post-referral Period Simulating Single System ## Cost /Patient/Month - Mean (sd)**						
				Median Pairwise Difference		
					Confidence Interval	
Variable	Combined System	Single System	% Change	Estimate	2.50%	97.50%
Total	17,769 (16,917)	23,514 (23,386)	32%	7,163	1,901	14,805
Inpatient	17,085 (17,122)	22,830 (23,676)	34%	7,110	1,901	14,805
Inpatient: Critical Care	9,885 (13,407)	15,211 (20,968)	54%	7,507	920	18,012

** Costs expressed in dollars without decimals; ## Inpatient: General was not possible to test due to insufficient data. Only 2 pairs had this type of admission at the hospice in the post-period; CPCH: Canuck Place; BCCH: BC Children's Cohort; sd: standard deviation.

The absolute total costs for the admissions that occurred at the hospice would increase 56% had they been at the hospital (Table 40).

Table 40. CPCH Group, Absolute Number of Admissions to the Hospice and their Respective Costs of Similar Admissions at BCCH

CPCH Group - Admissions to the Hospice and the Respective Costs of Similar Admissions at BCCH **									
			Costs at the Hospice			Cost at the Hospital			% Absolute Change in Total cost
	Number of Admissions	Total LOS	Total Cost	Cost/ Admission	Cost/ Day	Total Cost	Cost/ Admission	Cost/ Day	
Inpatient: Critical Care	7	32	88,024	12,574	2,750	136,992	19,570	4,281	56%
Inpatient: General	3	16	32,531	10,843	2,033	46,592	15,530	2,912	43%

** Costs expressed in dollars without decimals; CPCH: Canuck Place; BCCH: BC Children's Cohort; sd: standard deviation; LOS: length of stay;

Moreover, when **comparing patients to themselves in the pre- and post-referral periods**, the median decrease in monthly costs observed in the CPCH group would have decreased to $7,767 versus the previous $8,922 (Table 41 vs Table 34). The decline in monthly critical care costs would have been $21,957 instead of the previous $23,685; and the rise in non-critical care costs would have been $3,298 instead of the previous $2,877.

Table 41. CPCH Group: Changes in Monthly Cost per Type of Admissions under a Single System Scenario, and Pairwise Test (Wilcoxon) – Pre-Post-Referral to PPCP[§]

Changes from Pre to Post-referral CPCH Group Cost /Patient/Month - Mean (sd) **						
	Single System Scenario		Median Pairwise Difference			
				Confidence Interval		
Variable	Before	After	Estimate	2.50%	97.50%	*p*
Total	38,243 (56,351)	19,871 (21,106)	-7,767	-57,248	13,036	0.322
Outpatient	407 (553)	746 (1,225)	253	-187	1,531	0.272
Emergency	NA	NA				
Inpatient	37,833 (56,567)	19,119 (21,320)	-8,274	-57,243	11,939	0.322
Inpatient: Critical Care	33,797 (58,695)	13,164 (20,913)	-21,957	-68,800	7,999	0.529
Inpatient: General*	4,035 (7,236)	5,955 (9,038)	3,298	-9,297	15,971	0.675

** Costs expressed in dollars without decimals; CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls); CI: confidence interval; p: p-value; ICU: intensive care unit admissions; sd: standard deviation; NA: not available/did not occurred; § Pair #78 was excluded from the pre-referral period, as the case was a newborn enrolled in the PPCP from birth; * CI of 95% was not achievable because of lack of data. Software R automatically

When projecting these changes over the entire observational period of the cohort study (3 years) they represented an average rise of 5% in the total costs of the CPCH group (Table 42). Although it seems like a diminished impact, the non-parametric test showed that the median increase in costs across the cases would be a monthly $692. This may be due to later referral to PPCP, as 6 months prior to death the proportion of enrollment in the program was only 37% in the CPCH group.

Table 42. CPCH Group, Changes in Monthly Cost per Type and Pairwise Test (Wilcoxon) – Simulating a Usual Care Scenario in the Entire Observational Period

"CPCH Group - Changes in the Entire Observational Period Simulating Usual Care Cost /Patient/Month - Mean (sd)"						
				Median Pairwise Difference		
					Confidence Interval	
Variable	Combined System	Single System	% Change	Estimate	2.50%	97.50%
Total	35,754 (47,285)	37,410 (48,272)	5%	692	286	6,743
Inpatient	35,321 (47,535)	36,977 (48,531)	5%	692	286	6,743
Inpatient: Critical Care	29,555 (49,470)	31,175 (50,015)	5%	757	276	7,651

Inpatient: General was not possible to test due to insufficient data. Only 2 pairs had this type of admission at the hospice in the post-period; CPCH: Canuck Place; BCCH: BC Children's Cohort; sd: standard deviation.

In a brief, children in the CPCH group who already presented with higher costs compared to their matches, would have a significant rise in costs if inpatient admissions had not been provided in hospice, specially in regards to critical care admissions.

4.3.4.2 Within the Entire Hospice Population

To simulate the impact in costs from all inpatient care provided by the hospice, all the admissions from 2011-2012 fiscal year were priced with the equivalent costs from BCCH. In total 153 distinct children had at least 1 admission in that year, and 2192 bed-days were occupied, of those 331 were critical care beds and 1861 were non-critical care beds. The specific numbers of bed usage by acuity level and the impact in costs are presented in Table 43.

The total cost of inpatient care at the hospice was 3,888,578. Had these admissions happened at BCCH, they would have cost $6,836,243. Potential annual savings in 2012 from this shift of inpatient care to the hospice was $2,947,665, which represented $1,579 for every day in a ward-bed and $961 for every day of critical care avoided at BCCH.

In terms of hospital management processes in the planning and provision of inpatient care, the services provided by the hospice decreased the occupancy at BCCH by 155 ward beds and 28 critical care beds per month by the LTC pediatric population. Considering a 12h-shift for nurses and the minimum requirement of Mandated Nurse-Patient Ratios (MNPR)[62] published in 2011, an equivalent decrease of 156 shifts in wards and 56 shifts in the critical care unit per month at BCCH occurred due to the shift in health care setting. This workload decrease corresponds to

approximately 17 registered nurse positions, and in average, an annual $1,154,156 in nursing salaries (based on the 2010-2012 Collective Agreement[63]).

To account for the uncertainty around the cost units for the hospital admissions, a sensitivity analysis was performed. A range of 20% increase to 20% decrease in the hospital costs was applied, and a scenario where the costs of admissions at the hospital would be equivalent to the most expensive admission at the hospice (Table 44). The sensitivity analysis showed that the potential annual savings from this shift of inpatient care to the hospice would range from $1,182,402 to 4,314,914, which represented $700 to 2,161 for every day in a ward-bed and $569 to $1,818 for every day of critical care avoided at BCCH.

Thus, in the existence of CPCH, the hospital potentially had the opportunity to apply those 6.8 million dollars to care for other pediatric clientele, derived from the inpatient care provided at hospice to the LTC population.

Table 43. Yearly Inpatient Care Provided at CPCH, Fiscal Year 2011/2012 (n= 153 children with LTC)

Annual Inpatient Care Provided at CPCH - Fiscal Year 2011/2012						
	Ward Admissions			Critical Care		
Level of Acuity	Level 1	Level 2	Level 3	Level 4	Level 5	Total
Bed Day Usage	254	689	918	311	20	2192
CPCH Cost of a Bed per Day	612.37	1,354.51	2,033.24	2,750.76	3,888.58	
Total Cost of Inpatient Care at CPCH	155,543	933,259	1,866,517	855,487	77,772	3,888,578
BCCH Cost of a Bed per Day	$2,912	$2,912	$2,912	$4,281	$4,281	
Total Cost of Similar Inpatient Care at BCCH	739,648	2,006,368	2,673,216	1,331,391	85,620	6,836,243
Potential Annual Savings of Shifting Inpatient Care to the Hospice	**584,105**	**1,073,109**	**806,699**	**475,904**	**7,848**	**2,947,665**
Potential Savings of Shifting Inpatient Care to the Hospice per Bed, per Day	2,300	1,557	879	1,530	392	
Average Potential Savings of Shifting Inpatient Care to the Hospice per Bed, per Day		**1,579**		**961**		

Bed-days Avoided at BCCH	**Per day**	**Per week**	**Per month**	**Per year**
Ward bed-days	5	36	155	1861
Critical care bed-days	1	6	28	331
Number of Nurses Required in 24h (12h-shift) - rounded up	**Per day**	**Per week**	**Per month**	**Per year**
Ward (ratio 1:2)	6	36	156	1861
Critical Care (ratio 1:1)	2	13	56	662

CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls).

Table 44. Sensitivity Analysis for the Uncertainty Around the Hospital Cost Units

Annual Inpatient Care Provided at CPCH - Fiscal Year 2011/2012		Sensitivity Analysis Around the Hospital Cost Units				
		20% higher	**10% higher**	**10% lower**	**20% lower**	**Equal to the most costly bed at CPCH**
Bed Day Usage	2,192					
Total Cost of Inpatient Care at CPCH	3,888,578					
Total Cost of Similar Inpatient Care at BCCH	6,836,243	8,203,492	7,519,867	6,152,619	5,468,994	5,070,980
Potential Annual Savings of Shifting Inpatient Care to the Hospice	**2,947,665**	4,314,914	3,631,289	2,264,041	1,580,416	1,182,402
Ward Bed - Average Potential Savings of Shifting Inpatient Care to the Hospice per Bed, per Day	1,579	2,161	1,870	1,287	996	700
Critical Care Bed - Average Potential Savings of Shifting Inpatient Care to the Hospice per Bed, per Day	961	1,818	1,389	533	105	569

CPCH: Canuck Place cohort (cases); BCCH: BC Children's Cohort (controls).

4.4 Discussion

4.4.1 Summary of Results

The units of costs per day for all types of admissions at the hospice were found to be lower when compared to those at the hospital. On average, the daily cost of a non-critical care bed was $2,912 at the hospital and between $612 and $2,033 at the hospice. Whereas, the daily cost of a critical care bed was $4,281 at the hospital and ranged from $2,750 to $3,888 at the hospice. This difference in cost between the final units of cost per type of admission could not be tested statistically. The cost of outpatient visits for cancer patients had a higher average cost than for non-cancer patients in both groups, which is expected due to frequent adjuvant therapy, imaging and laboratory follow-ups as outpatients.

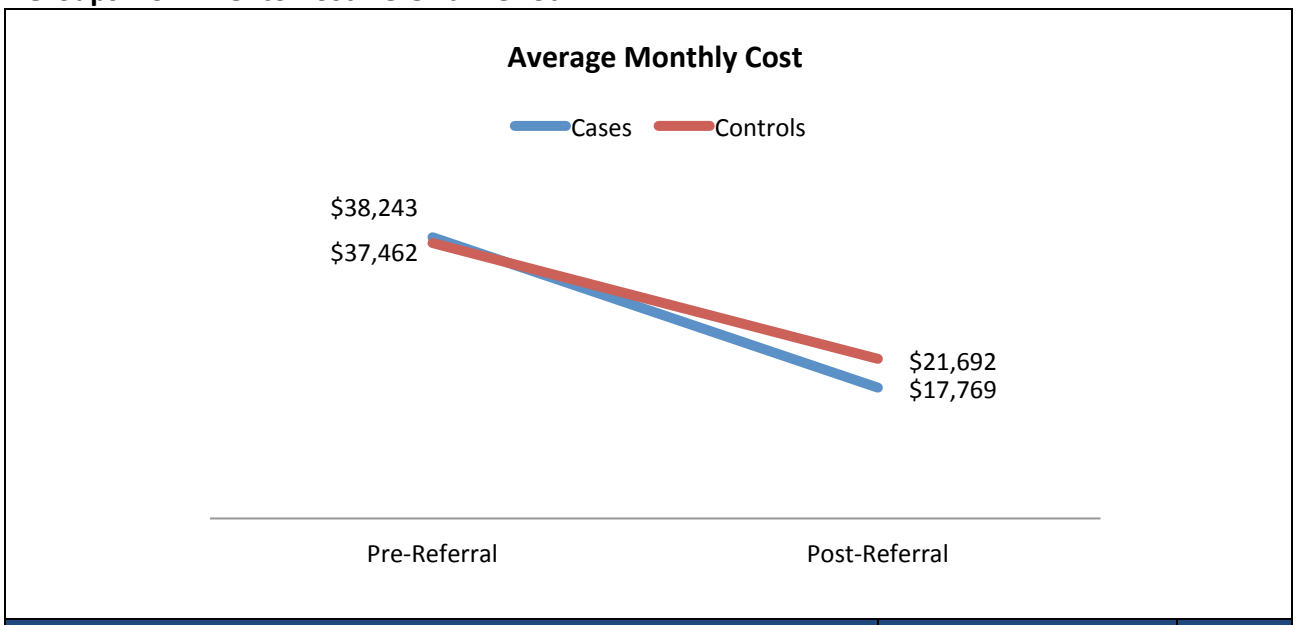
In this combined system with hospital and hospice provider, applying the specific units of cost to patients' utilization, the time series graphs demonstrate that both groups were very similar until approximately 15 months prior to death when they became markedly distinct. Although both groups showed an upward trend in inpatient cost towards death, the CPCH patients seemed to be more costly, particularly in the last 2 months prior to deaths (attributed to critical care admissions). Costs of outpatient visits were very steady and similar between groups over time.

However, Figure 26 summarizes the cost comparison of the aggregate results, which showed no statistically significant differences in costs between groups in any comparison, as well in their changes after the referral point. The lack of significance does not prove lack of difference between groups but might have been due to the low power of this study to detect true differences.

In the **pre-referral period**, children in both groups were under usual care only. Overall, the average monthly cost per patient in the CPCH group was $38,243 versus $37,462 in the BCCH

group. In the **post-referral period**, when the CPCH group started to access PPCP services their average monthly total cost per patient was $17,769 in the CPCH group versus $21,692 in the BCCH group. The average monthly cost estimates could lead to misinterpreting the CPCH group as having lower costs after referral. However, the pairwise analyses (controlling for disease and age at death) showed that children in the CPCH group tended to have higher overall costs compared to their matches both pre- and post-referral, as their median difference in costs were higher and increased in the post-referral period.

Figure 26. Average Monthly Cost: Summary of Comparison Between Groups and Changes within Groups from Pre- to Post-Referral Period **



	Pre-Referral	Post-Referral	Changes Within Group	*p*
Cases	$38,243	$17,769	-$8,846.00	0.322
Controls	$37,462	$21,692	-$74.00	0.846
Median Difference Between Groups	$2,522	$4,270		
p	0.275	0.610		

When **comparing patients to themselves in the pre- and post-referral periods**, both groups showed a drop in monthly cost per patient post-enrollment with distinct magnitude, however, this decrease was not found to be significant. Overall, the median decrease observed in the CPCH group was $8,846 compared to $74 at the BCCH group. In the BCCH group this downward trend was observed for all types of admissions (outpatient $49, critical care $2,398, non-critical admissions $1,195). Whereas in the CPCH group, the drop in monthly costs of critical care admissions ($23,579) was partially offset by a rise in outpatient visits ($253) and non-critical admissions ($2,885). These changes within groups conflict with the exploratory analysis from the time series and might be due to the limitations in the data previously discussed in Chapter 3.4.2 and referral to the program less than a month before the death occurred. Overall, 56% of costs in the CPCH group post-referral were shifted to the hospice post-referral, more so with non-cancer patients who were noticeably more costly than cancer patients.

Given the variability of costs found in this study (n=11 pairs, mean difference in monthly inpatient cost $4,255, sd= $ 42,904), an ideal sample size for a comparative study would be 791 pairs of children to find a minimum difference between groups of $ 4,281/month/patient (equal to the cost of 1 critical care day at BCCH). As a result, under the same parameters (significance level = 5% and power = 80%) this study had a power of 6%. This study presented much larger variability in mean decrease in monthly inpatient cost when compared to a published pre-post study that utilized a similar broad perspective to measure changes in health care utilization after enrollment in a PPCP (Pascuet et al[46] , n=66, $4,252 per patient, sd=$3,674.08 versus CPCH group, n=11, $23,757 per patient, sd = $49,904).

Simulating a single system scenario for children in the CPCH group, with all admissions occurring at the hospital (same level of acuity), did not change the results from the comparison between groups but it was relevant within the CPCH group. In the **post-referral period**, the average monthly cost per patient would have increased from $17,857 to $23,514 in the CPCH group. This 32% rise in total costs was driven by a 54% increase in critical care costs, and represents a monthly increment across cases of $7,163. The absolute total costs for the admissions occurred to the hospice would increase 56% had they happened at the hospital. In the pairwise analyses (controlling for disease and age at death) between CPCH and BCCH groups, the higher costs of children in the CPCH group would have been more pronounced in all types of admissions when compared to their matches, however, still did not reach the significance level.

Moreover, when **comparing patients to themselves in the pre- and post-referral periods**, the median decrease in monthly overall costs observed within the CPCH group would have been less pronounced, along with the decrease in critical care costs. A slight increase in non-critical care costs would have been observed. Projecting this rise in cost over the **entire observational period (3 years**), would have impacted 5% of total costs in the CPCH group with a monthly increment across cases of $692. This may be due to later enrollment to PPCP, which was more intense in the last 4 months of life.

Simulating the same scenario (shifting back the inpatient admissions to the hospital) to all the 2192 bed-days provided by the hospice in 2011-2012 would drive an increase of $2,947,665 in inpatient care costs (ranging from $1,182,402 to 4,314,914 in the sensitivity analysis) for the 153 children admitted in that fiscal year (provided at the hospice $3,888,578 versus at BCCH $6,836,243). The potential annual savings in 2012 from the shift of inpatient care from the hospital

to the hospice were equivalent to $1,579 for each day in a ward bed and $ 961 for each day in critical care avoided at BCCH. This was equivalent to a decrease in the hospital occupancy at BCCH of 155 ward beds and 28 critical care beds per month, an equivalent decrease of 156 nursing shifts in wards and 56 nursing shifts in the critical care unit per month at BCCH (considering 12-hour shifts and MNPR).

These numbers were interpreted as potential savings because, in order to translate to real savings in the health care system, the reallocation of funds that would have been applied to pay for admissions to the hospital should have been transferred from the hospital budget to the hospice funding. A linear application of government funding that has supported the hospice (historically, 26% of the hospice budget) would represent a subsidy of $1,011,028 to inpatient care. No information is available on whether there has been such reallocation from the hospital's annual budget to the hospice, or whether it was an additional expense to the publicly funded health system. Further, no speculation about the feasibility of the decreasing number of hospital beds offered at BCCH due to this shift in inpatient care to the hospice is applicable. There is no information about whether the opened beds at BCCH were occupied by a backlogged demand for hospital beds among other pediatric patients or if the were left empty, although in the current climate of bed occupancy one can surmise the former.

Finally, CPCH is a free-standing hospice that provides a more holistic approach to EOL and therefore, some services are provided for families during the child's admission, free of charge. This in-house support for families (counseling, accommodations, meals, siblings therapy) accounted for $1,246,272 in the same year. These services, in the absence of the hospice care, would either represent an out-of-pocket expense for families (e.g. meals, accommodation, baby-sitting for siblings) or not being provided at all (e.g. memory making, play therapy and counseling for siblings).

4.4.2 Limitations of this Study

Besides the obvious sample size limitation of this study, and others previously discussed in Chapter 3, a number of additional shortcomings should be highlighted. No micro-costing was possible to derive final units of cost for specific types of admissions by facility (measurement where each component of resource use is estimated and assigned a final unit of cost at the patient-level). The final units of cost for outpatient and emergency visits were calculated based on data from a small sample size (n=22), without including overhead costs and without much description of the methodology for data extraction. It raises concerns about the completeness and rigor of the data. Further to this, no standard deviations of the means were available, and as a result any differences between groups could not be tested for significance.

The cost units for the BCCH admissions, extracted from a general pediatric population by CIHI, could potentially be different if extracted exclusively from a LTC population cared for within the hospital. The direction of this bias is unknown as the costs could be diluted with less severe non-LTC patients, or overestimated for being aggregated with costs from more expensive curative treatments. Neither 'disease specific per diem' or 'case-mix group' costs are available from this population to enable a more precise cost comparison.

The cost units for the CPCH admissions, although not from a sophisticated cost distribution methodology (such as step-down allocation with iterations) still offers the most specific cost for inpatient care within a LTC pediatric population. However, the hospice does not have patient-level data on in-house and outreach service consultations by patients and family members, to allow for an even broader cost comparison and societal perspective.

A more complete measure of direct costs from a publicly funded health care system perspective was not feasible due to lack of data on a number of other costs such as community-

based services, rehabilitation, and aids/ appliances provided outside of BCCH and CPCH, disability benefits, social services, patient/family's time spent for travel or receiving care, lost time at unpaid work, out-of-pocket expenses for families, paid caregivers, and productivity loss. A societal perspective would be ideal for future studies especially in regards to those costs since they are known to occur within this population. Finally, the extension of the cost impact beyond the cohort study must be considered with caution as more information is needed on hospital occupancy rates and general population demand for hospital care to draw conclusions on the reallocation of resources or planning changes in capacity and funding.

4.5 Conclusions

Overall an upward trend in inpatient cost towards death was observed in both groups. However, the CPCH group seemed to be more costly than the usual care group both pre- and post-referral to the program, especially for critical care. While this finding may be due to heighted severity of illness in the CPCH group, no statistically significant differences in costs between the groups were found in any comparison, possibly due to the small sample size. Overall, approximately half of the costs of the CPCH group was shifted to the hospice post-referral, with non-cancer patients remarkably driving the results.

Simulating a 'usual care' scenario for children in the CPCH group, where all inpatient care was sought at the hospital instead of the hospice, resulted in greater cost to the system. This simulation demonstrated that the higher costs incurred by the CPCH group would have been even more pronounced had their care remained at the hospital. A striking rise in total costs (32%) especially with critical care admissions (54%) would translate into a relevant median monthly increase of $7,163 across cases. Extending this cost exercise beyond this cohort study to all

inpatient care provided by the hospice in the fiscal year 2011-2012, a potential savings to the health care system ranging from just over $1.18M to just over $4.3M was calculated due to a shift of care from hospital to hospice for patients who had the most expensive care demands.

These results should be interpreted with caution due to the limitations of a small sample size and the lack of more precise cost measurements. Studies on the reallocation of these resources within the publicly funded health care system, along with mapping of the health care providers' capacity and demand for pediatric hospital care is recommended. Future studies with more sophisticated methods on operating costs allocation and costs measurements such as micro-costing, case-mix group or disease specific per diem should be undertaken. Also, a societal perspective would be ideal for further investigation to evaluate the financial impact on families who are caring for children with LTC.

Chapter 5: Final Discussion

5.1 Summary of Results

The effects of PPCP on health care utilization and costs from the published literature and from the BC program were described in this thesis. In particular, inpatient care accessed by this LTC pediatric population in the different health care settings was examined.

Prior to initiating this study, it was hypothesized that health care utilization and costs would be lower among children who accessed PPCP. We hypothesized that this reduction in costs would be especially in health care settings with hospice facilities that can better manage EOL conditions with a holistic approach to care, family training/education, and coordination of care across settings. However, after the literature review, the hypothesis slightly changed. It was still expected to find differences between PPCP users and non-users, but the direction of the results were not anticipated yet tested in both sides.

Chapter 2 presented a systematic review of the published literature that found moderate to low methodological quality of previously published studies investigating health care utilization and costs. The majority of studies used a narrow measurement perspective, focusing mostly on hospital admissions. Only 2 studies utilized broader measurement including hospice use and the results were context dependent. The literature does, however, demonstrate that PPCPs resulted in no increase in hospital resource utilization and suggests a shift to other health care settings. Depending on the health care system this may lead to overall costs increasing; in the Canadian context, there would appear to be cost savings.

Chapter 3 presented a retrospective matched-pair cohort study (n=11 pairs) comparing data from the BC PPCP and usual care. Findings from the aggregate data suggest that children who enrolled in the program presented an increased number of monthly admissions, especially in

critical care nearing death (compared both to their controls and to themselves in the period prior to referral). However, this increase in admissions might still have happened by chance due to the study limitations. When referring to critical care, it includes admission to the hospice classified as acuity level 4-5, which are equivalent to those occurring in NICU and PICU at the hospital, based on nursing workload and patients symptoms. Despite this increase in number of admissions, the LOS was not significantly affected. However it seems that program users spent more days in a health care provider compared to their pairs even before being enrolled in the PPCP. The lack of significance in the analysis does not prove lack of difference between the groups but could be due to the low power of the study to find true differences. The time series showed that, both groups had similar inpatient utilization until approximately 15 months before death, when the group of children who later would be enrolled in the PPCP, started to have consistently more non-critical admissions than the usual care group. In the last 2 months prior to death, these children became more severely ill indicated by more time spent in critical care (despite the controls having died around the same time). This in itself might be the differential reason that led to the referral and enrollment to the PPCP. Regarding the EOL admission, while no significant difference was found between groups, children enrolled in PPCP had shorter LOS and lower absolute number of procedures to prolong life. Despite the need for critical care, a shift in setting of health care utilization from hospital to hospice was observed (more evident in the non-cancer subgroup).

During the cost analysis (Chapter 4), as would be expected, the impact on costs followed similar trends to the health care utilization observed in the matched-cohort study with both groups showing an upward trend in inpatient cost towards death when displayed overtime. In the aggregate data patients enrolled in PPCP seemed more costly than their matches both pre- and post-referral, especially with respect to critical care costs. However, no statistically significant

differences in costs between the groups were found. Overall, post-referral, approximately 50% of their costs shifted to the hospice (remarkably among non-cancer patients).

Simulating a 'usual care' scenario where children under the PPCP would have accessed the same inpatient care exclusively at the hospital (instead of at the hospice) showed they would be even more costly to the health care system with a rise in total costs (32%) especially for critical care admissions (54%). Overall, had the children in the CPCH group received the care they needed solely at the hospital, a median increment across PPCP users of $7,163 per month per child would have occurred. Extending the cost exercise beyond the cohort study to all inpatient care provided by the hospice during the fiscal year 2011-2012 (153 children), potential annual savings to the health care system equivalent to $1.1M to $4.3M was calculated as a result of the shift of care from hospital to hospice. This of course is not a 'real' savings, but does indicate that should CPCH not be available, the costs to the system for this population of patients would potentially higher.

These findings align with the only 2 previously published studies that also measured hospice utilization/cost and showed PPCP users to have longer LOS[46] and higher costs[21] than their controls, but with a decrease in inpatient costs[46] after enrollment to a hospice-based PPCP.

5.2 Contributions to Research

Chapter 2 represents the first systematic review on the effects of PPCP on utilization and costs, and chapters 3 and 4 describe the first matched-pairs cohort study performed in this pediatric population. Our findings demonstrate incomplete measurement in the majority of previous literature, which was limited to the use of hospital admissions to assess the effects of the PPCP on those outcomes. However, when a more comprehensive measurement of health care utilization (including other health care settings) any assumptions that PPCP users have lower

utilization is not supported. Questions remain as to whether selection bias is playing an important role in the results of any study in this field, given the self-selective nature of enrollment in these programs. Perhaps these children end up enrolled in PPCP for already being more severely symptomatic and intense users of health care resources. Also, the challenges in the matching process demonstrate the difficulty of conducting comparative studies in this population. The low number of matches suggests that perhaps children who died under the PPCP are systematically different from those who died without being referred to PPCP in regards to disease type or onset of the condition. While this work on utilization does represent an advance in the literature, clearly any conclusions from this work should be viewed with caution. A further contribution was in providing hard information on the variability of the outcomes, with a standard deviation, which could be considered for sample size calculations when planning future studies. Finally, the hospice management team assumed that inpatient hospice care would be more expensive than hospital care. However, when preparing the cost analyses for this study, a great proportion of outreach services were found not to be included as a final department under the cost distribution of the hospice. These services accounted for approximately 30% of the hospice operating cost, which was historically allocated to the cost of inpatient beds and in-house family services. As a result, the cost of a bed per day at the facility becomes inflated. As such, any cost comparisons using hospice cost data must account for this. More generally in terms of costing, it was perhaps surprising that substantial differences between facilities did not arise, as there would be a common understanding that hospice care is more expensive. This work (cautiously) dispels this misconception and suggests, again, that more work is required in understanding the types of patients presenting at each type of facility and the movement between facilities particularly near end-of-life.

5.3 Limitations of this Study

While this study has been designed to provide local-level data on the current trends in health system utilization and costs between the two pediatric populations who died from LTC, the BCCH and CPCH databases used in this study are secondary data from administrative databases not collected for research purposes. Hence, the databases may have incomplete data for certain contents (clinical outcomes) and services utilized outside of those providers. Data from other providers such as general practitioners (GPs), community hospitals and other facilities were not included in these databases. However, both these databases currently represent the largest and most comprehensive datasets available on this particular population.

In addition, the results should not be generalized beyond the population included in the study as the 11 pairs include represented only a few conditions. This research is limited by the available population (i.e. approximately 210-250 deaths per year [18,32]). Further, the target population included in the matched-cohort comparison did not include approximately 50% of the patients enrolled in the BC PPCP every year[53§§] who died elsewhere. This excluded population might have presented a different pattern with respect to health care resource utilization/costs and may be systematically different from the patients in this study, as EOL did not occur at the hospice. Population level data is needed to enable a comparative study that included these children in order to have a database to match controls from.

Furthermore, there is no established indicator for when the goals of care shift from curative to palliative (i.e. when a child should be "referred" to PPCP). This uncertainty did not allow for a precise "ground zero" for palliative care from which comparisons should begin, leaving "death" as the indicator of any potential benefits received through PPCP among cases, where the same

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beneficial period was approximated among controls from their matches. Likewise, no formal indicator or score for "severity of disease" with which to test for any such systematic differences between groups was available.

Many challenges were faced using the ICD code classification, as it does not entirely cover the range of rare conditions within LTCs, diminishing the specificity of such classification. The age at death criterion further restricted the matching process and resulted in small sample size, thereby limiting the use of parametric tests or more sophisticated regression models, which that would have allowed to better control other confounders.

The power to detect any difference between groups in utilization/costs with only 11 matched-pairs was only 5-6%. The ideal comparative cohort study would need over 800 pairs of children, which would require a multicenter study with other similar settings of a hospice-based PPCP or a database with a longer observational period, as the population of children who die from LTC is naturally small.

The inclusion of newborns may also have overestimated the mean estimates of outcomes due to the 'weighting by length of observational period' technique used to address differences within and across pairs. Additionally, measurement of LOS in the current administrative system did not take into account transfers between units (e.g. ward to critical care) duplicating the count when a transfer occurred instead of assigning proportional LOS. The results of the non-parametric tests should not be influenced by these challenges, as the median is not sensitive to extreme observations. However, the data treatment to allocate the outcomes to pre- or post period, added to later referral to the PPCP in a substantial number of cases, may have affected some results inflating some outcome measurements.

No micro-costing, disease specific per diem or case-mix group costs are available from this population to enable a more precise cost comparison. The 'partial direct care' units of cost were calculated based on the small sample size, without including overhead costs. The reported methodology was also relatively short and at points difficult to interpret despite numerous contacts with administrators within PHSA. The direct costs were more disease specific for the hospice costs, as the hospital costs were calculated using data from the general pediatric population. The direction of the effect on the final units of cost is unknown.

The perspective of the cost analysis was limited by the lack of data within this population on a number of other cost components such as utilization across other settings such as community-based services, rehabilitation, direct costs to publicly funded services other than health care, time costs to patients and their families among others. Moreover, the extension of the cost impact beyond the cohort study needs to be considered with considerable caution as additional data on hospital occupancy and population demand for hospital care is needed to support the reallocation of resources.

As well, the late enrollment in the PPCP observed in this sample may have also affected costs, and could have been more pronounced had inpatient care shifted earlier in the disease trajectory. This assumption may not hold for cancer patients who have a different disease course yet to non-cancer conditions known to be incurable.

The potential savings of shifting inpatient care to the hospice were calculated without accounting for the hospital funding system (global budget). Perhaps in the absence of the hospice provider the hospital would manage to accommodate the demand within its budget without compromising the provision of services to other pediatric populations. However, it would still lack

some components of the program to the patient care and family support keeping the *status quo* of its services.

Finally, this study did not address quality of life in this population and, therefore, cost-utility analyses are still lacking in this field. During the systematic review, the search strategy and combination of terms was built to allow for cost-effectiveness and cost-utility studies to be retrieved, however, no studies found. The cost/quality-adjusted life year (QALY) indicator was discussed within the research team yet chosen not to be among the main outcomes for various reasons such as time constraint and complexity of measuring QALYs at EOL. Numerous challenges for measuring QALYs among pediatric palliative care users range from choosing the appropriate respondent (child, parent(s), care giver, clinician) to the appropriateness of measuring QALYs in palliative care, the need for adjustments in QALY indicators or for new utility measures.[64-66] These issues clearly suggest that QALY measurement in PPCP could be a topic in itself as a separate research project.

5.4 Knowledge Translation

Several strategies will be used to ensure dissemination of the findings of this evaluation to health care professionals, the public, policy makers, and researchers. The three components of this research are expected to result in publications in one or more high-level academic journals, and presented in conferences of various specialties such as palliative care, health services research and health economics. Publication will be sought following the thesis defense for the papers on health care utilization and cost study. The results will also be presented to the Hospice Senior Leadership Team to support strategic planning. Further, policy makers will be informed of these results

through reports, which will be shared with BCCH and the regional health authorities in BC to inform decision-making and priority setting.

5.5 Policy Implications

The results of this study should be relevant to decisions on provision and funding of pediatric palliative care in the province. It might inform initiatives on reallocation of resources within the publicly funded health care system, mapping health care providers' capacity and population demand for hospital and hospice care. In addition, the findings of these studies should be useful to the health care providers and clinicians in BC who may use the results to potentially improve patterns of referral of LTC children to the provincial PPCP. Enhanced timing of shifts to PPCP can consequently optimize resource use within the health system across different settings, as well as offer a more holistic approach to care at EOL for families earlier in the disease trajectory, and relieve hospital resources to meet other demands within the pediatric population.

5.6 Future Research

This project has left important questions unanswered but we hope it will strengthen the foundation of research in pediatric palliative care in Canada and provides the basis for a long-term study that examines the cost-effectiveness of providing pediatric palliative care. In the future, it will be essential to address utility measures for quality of EOL in the pediatric palliative care population in conjunction with assessment of costs. This research area is obviously very complex but the development of cost-utility studies to allow comparisons across different health care programs within the publicly funded health system can enhance priority setting and appropriate reallocation

of resources. Further, prospective studies that report indicators of severity of disease and a record of the 'starting point' from which each child should be referred to palliative care are essential to provide clarifications on the systematic differences between children who enroll in PPCP before death and those who die under usual care, detangling possible confounding factors.

A societal perspective would be ideal for future studies especially with respect to costs borne by social services, income transfer payment aids, time costs to patients and family caring for these children, and out-of-pockets payments. Further, loss in productivity, which is known to occur often within this population, is essential when measuring the burden of the diseases on families and society.

Standardization of outcome measures will enhance comparability and pooling of future research to increase power, given the nature of this small population. Also, more transparency across providers on the methodology used to reach the final units of cost should be pursued to increase comparability across settings and avoid potential misallocation of resources. Finally, future studies with more sophisticated methods on operating cost allocation and cost measurements such as micro-costing, case-mix group or disease specific per diem should be developed.

5.7 Conclusion

This thesis represents an important step towards a more comprehensive understanding of the impact of PPCP on resource utilization across various health care settings. The findings suggest that the LTC pediatric population referred to PPCP might potentially have systematic differences compared to those who died without using these programs, presenting with a more intense utilization of the health care system resources and consequently more costly. Misconceptions about higher costs of providing hospice care compared to the hospital were discredited in the local

context, and potential savings to the health system of this shift in health care setting was calculated. Any conclusions from this work should be viewed with caution given the small sample size available for comparative research and the challenges to overcome selection bias given the nature of the enrollment into the program. Future research in the field should aim for indicators of severity of the diseases and starting point for palliative care, measurements of QALY in this population, standardization of measurements of utilization for beyond the hospital setting, and broadening of the economic outlook to a societal perspective. Further, more transparency and sophisticated methods to calculate final units of cost for services provided across different settings are imperative. Overall, this is an advance in the literature within the pediatric palliative care field and a valuable insight to administrators to inform measurement techniques and resource allocation.

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Appendices

Appendix A . Search Strategy

A.1 Medline Search Strategy

Ovid MEDLINE(R) In-Process & Other Non-Indexed Citations and Ovid MEDLINE(R)

1 adolescent/ or exp child/ or exp infant/
2 exp Pediatrics/
3 Minors/
4 or/1-3 [children]
5 *Palliative Care/ec, mt, st, sn, td, ut [Economics, Methods, Standards, Statistics & Numerical Data, Trends, Utilization]
6 *Hospice Care/
7 *Terminal Care/ec, mt, st, sn, td, ut [Economics, Methods, Standards, Statistics & Numerical Data, Trends, Utilization]
8 *Respite Care/
9 or/5-8 [Palliative Care narrow main topic]
10 4 and 9 [children and palliative care]
11 limit 10 to "review articles"
12 limit 10 to systematic reviews
13 limit 10 to meta analysis
14 systematic review?.mp.
15 Cochrane database of systematic reviews.jn.
16 or/14-15
17 10 and 16
18 or/11-13,17 [children and palliative care reviews] (
19 limit 18 to yr="1974 -Current" [reviews after 1974]
20 Adrenoleukodystrophy/
21 Alagille Syndrome/
22 Alexander Disease/
23 alpha-N-Acetylgalactosaminidase/df [Deficiency]
24 Argininosuccinic Aciduria/
25 Aspartylglucosaminuria/
26 Bartter Syndrome/
27 Canavan Disease/
28 Carbamoyl-Phosphate Synthase I Deficiency Disease/
29 "Congenital Disorders of Glycosylation"/
30 Carnitine O-Palmitoyltransferase/df [Deficiency]
31 Cholesterol Ester Storage Disease/
32 Citrullinemia/
33 Costello Syndrome/
34 Cri-du-Chat Syndrome/
35 De Lange Syndrome/

36 "Diffuse Cerebral Sclerosis of Schilder"/
37 DiGeorge Syndrome/
38 Farber Lipogranulomatosis/
39 Fucosidosis/
40 Gangliosidoses/
41 Gangliosidoses, GM2/
42 Glycogen Storage Disease Type II/
43 Glycogen Storage Disease Type IIb/
44 Glycogen Storage Disease Type IV/
45 Hereditary Central Nervous System Demyelinating Diseases/
46 Histiocytosis, Langerhans-Cell/
47 Huntington Disease/
48 Hyperglycinemia, Nonketotic/
49 Incontinentia Pigmenti/
50 Jacobsen Distal 11q Deletion Syndrome/
51 Kearns-Sayre Syndrome/
52 Klippel-Trenaunay-Weber Syndrome/
53 Lafora Disease/
54 Leigh Disease/
55 Lesch-Nyhan Syndrome/
56 Leukodystrophy, Globoid Cell/
57 Leukodystrophy, Metachromatic/
58 Lipidoses/
59 MELAS Syndrome/
60 Menkes Kinky Hair Syndrome/
61 MERRF Syndrome/
62 Methylmalonyl-CoA Mutase/df [Deficiency]
63 Mevalonate Kinase Deficiency/
64 Mitochondrial Encephalomyopathies/
65 Mitochondrial Myopathies/
66 Mucolipidoses/
67 Mucopolysaccharidosis I/
68 Mucopolysaccharidosis II/
69 Mucopolysaccharidosis III/
70 Mucopolysaccharidosis VII/
71 Multiple Acyl Coenzyme A Dehydrogenase Deficiency/
72 Multiple Sulfatase Deficiency Disease/
73 Myoclonic Epilepsies, Progressive/
74 Neuroaxonal Dystrophies/cn [Congenital]
75 Neuronal Ceroid-Lipofuscinoses/
76 niemann-pick disease, type a/ or niemann-pick disease, type b/ or niemann-pick disease, type c/
77 Oculocerebrorenal Syndrome/
78 Olivopontocerebellar Atrophies/
79 Ophthalmoplegia, Chronic Progressive External/

80 Ornithine Carbamoyltransferase Deficiency Disease/
81 Pelizaeus-Merzbacher Disease/
82 Peroxisomal Disorders/
83 Propionic Acidemia/
84 Pyruvate Carboxylase Deficiency Disease/
85 Pyruvate Dehydrogenase Complex Deficiency Disease/
86 Pyruvate Metabolism, Inborn Errors/
87 Refsum Disease/
88 Refsum Disease, Infantile/
89 Rett Syndrome/
90 Rubinstein-Taybi Syndrome/
91 Sandhoff Disease/
92 Sea-Blue Histiocyte Syndrome/
93 Sialic Acid Storage Disease/
94 Smith-Lemli-Opitz Syndrome/
95 Spasms, Infantile/
96 Sphingolipidoses/
97 sulfatidosis/
98 Unverricht-Lundborg Syndrome/
99 von Hippel-Lindau Disease/
100 Wolf-Hirschhorn Syndrome/
101 Wolman Disease/
102 Zellweger Syndrome/
103 Aicardi Syndrome/
104 Aicardi? Syndrome.mp.
105 alpha-N-Acetylgalactosaminidase/df [Deficiency]
106 alpha-NAGA deficiency.mp.
107 alpha-Mannosidosis/
108 Argininosuccinic Aciduria/
109 Argininosuccinicaciduria.mp.
110 Argininosuccinate lyase deficiency.mp.
111 ASAuria.mp.
112 ASL deficiency.mp.
113 ARSACS.mp.
114 Autosomal Recessive Spastic Ataxia of Charlevoix-Saguenay.mp.
115 beta-Mannosidosis/
116 Charlevoix-saguenay spastic ataxia.mp.
117 Dubowitz.mp.
118 Escobar Syndrome.mp.
119 Galactosidases/
120 Glutaric Acidemia Type I.mp.
121 Glutaric Aciduria Type I.mp.
122 Glutathione/df [Deficiency]
123 Infantile Neuroaxonal Dystrophy.mp. or Neuroaxonal Dystrophies/
124 Seitelberger's Disease.mp.

125 Jeune Thoracic Dystrophy.mp.
126 Asphyxiating Thoracic Dystrophy.mp.
127 Kanzaki Disease.mp.
128 Lennox-Gastaut Syndrome.mp.
129 3-methylcrotonyl-CoA carboxylase deficiency.mp.
130 Methylcrotonyl-CoA carboxylase deficiency.mp.
131 3-MCC deficiency.mp.
132 3MCC.mp.
133 MCC deficiency.mp.
134 Muscular Dystrophy, Duchenne/
135 exp Leukemia, Lymphoid/
136 Cerebral Palsy/
137 Neuroblastoma/
138 Muscular Atrophy, Spinal/
139 Neuroectodermal Tumors, Primitive/
140 Leukemia, Myeloid, Acute/
141 Mitochondrial Diseases/
142 Mitochondrial Diseases/
143 Friedreich Ataxia/
144 Osteosarcoma/
145 Trisomy 18.mp.
146 Medulloblastoma/
147 "Spinal Muscular Atrophies of Childhood"/ or SMA Type II.mp.
148 Pontine Glioma.mp.
149 Rett Syndrome/
150 Rhabdomyosarcoma/
151 Sarcoma, Ewing/
152 Brain Neoplasms/
153 Cystic Fibrosis/
154 Hypoxia-Ischemia, Brain/
155 Trisomy 13.mp.
156 Batten's Disease.mp.
157 Brain stem glioma.mp.
158 San Filippo Syndrome.mp.
159 Brain Stem Neoplasms/
160 CHARGE Syndrome/
161 Ependymoma/
162 Carcinoma, Hepatocellular/
163 exp HIV Infections/
164 Microcephaly/
165 Mitochondrial Diseases/
166 Adrenoleukodystrophy/
167 Biliary Atresia/
168 Glioblastoma/
169 Hodgkin Disease/

170 Hurler's syndrome.mp.
171 Leigh Disease/
172 Epilepsy/
173 Arthrogryposis/
174 Astrocytoma/
175 Atypical Teratoid Rhabdoid Tumour.mp.
176 Burkitt Lymphoma/
177 Chromosome Aberrations/
178 Down Syndrome/
179 Dravet Syndrome.mp.
180 Glioblastoma/
181 Glutaric aciduria.mp.
182 Hydranencephaly/
183 Hypoplastic Left Heart Syndrome/
184 Krabbe Disease.mp.
185 Muscular Dystrophies, Limb-Girdle/
186 Metabolism, Inborn Errors/ or Metabolic Diseases/
187 Leukodystrophy, Metachromatic/
188 Mitochondrial enzyme complex IV.mp.
189 Mitochondrial Myopathies/
190 Enterocolitis, Necrotizing/
191 NYD.mp.
192 Pallister-Killian Syndrome.mp.
193 Pelizaeus-Merbacher Syndrome.mp.
194 Polymicrogyria.mp.
195 Propionic Acidemia/
196 Hypertension, Pulmonary/
197 Severe brain injury.mp.
198 Tay-Sachs Disease/
199 Wilms Tumor/
200 Chromosome Inversion/
201 Adams-Oliver Syndrome.mp.
202 Aicardi-Goutieres Syndrome.mp.
203 Alpers Syndrome.mp. or "Diffuse Cerebral Sclerosis of Schilder"/
204 Aminoacid decarboxylase deficiency.mp.
205 Anaplastic Astrocytoma.mp.
206 Brain/ab [Abnormalities]
207 Adrenoleukodystrophy/
208 Anomalous left coronary artery from pulmonary artery.mp.
209 Anterior Horn Cell Disease.mp.
210 Askin's Tumour.mp.
211 ATRT.mp.
212 Atypical Di George Syndrome.mp.
213 Menkes Kinky Hair Syndrome/ or Atypical Menkes.mp.
214 Lymphoma, B-Cell/

215 Rassmussen's Encephalitis.mp.
216 Ependymoma/
217 Burkitt Lymphoma/ or Burkett Lymphoma.mp.
218 Heart Neoplasms/
219 Cardiomyopathies/
220 Cerebral AV Malformation.mp.
221 Renal Insufficiency, Chronic/
222 Renal Insufficiency/
223 Chronic lung disease.mp.
224 Central Nervous System/ab [Abnormalities]
225 Heart Defects, Congenital/
226 "Tetralogy of Fallot"/ or Complex Tetralogy.mp.
227 De Lange Syndrome/
228 Dandy-Walker Syndrome/
229 severe neurological impairment.mp.
230 Failure to Thrive/
231 Neoplasms/
232 or/20-231
233 adolescent/ or exp child/ or exp infant/
234 exp Pediatrics/
235 Minors/
236 or/233-235
237 232 and 236
238 terminal care/ or hospice care/ or resuscitation orders/
239 *Palliative Care/
240 *Terminally Ill/
241 *Respite Care/
242 *Progressive Patient Care/
243 *Long-Term Care/
244 or/238-243
245 237 and 244
246 limit 245 to "review articles"
247 limit 245 to systematic reviews
248 systematic review?.mp.
249 Cochrane database of systematic reviews.jn.
250 or/248-249
251 245 and 250
252 limit 245 to meta analysis
253 or/246-247,251-252 [reviews in LLC and palliative care]
254 limit 253 to yr="1974 -Current" [reviews after 1974]
255 19 or 254 [LLC and/or Children and Palliative care reviews after 1974]
256 comment/ or editorial/ or letter/ or news/
257 10 or 245
258 257 not (255 or 256) [LLC and/or Children and Palliative care - primary articles total]

259 limit 258 to yr="2000 -Current" [LLC and/or Children and Palliative care - primary articles after 2000]

A.2 EMBASE Search Strategy

Ovid EMBASE

1 adrenoleukodystrophy/
2 Alagille syndrome/
3 Alexander disease/
4 Fabry disease/
5 argininosuccinic aciduria/
6 aspartylglycosaminuria/
7 Bartter syndrome/
8 Canavan disease/
9 carbamoyl phosphate synthetase I deficiency/
10 "congenital disorder of glycosylation"/
11 Carnitine O-Palmitoyltransferase Deficiency.mp.
12 cholesterol ester storage disease/
13 citrullinemia/
14 Costello syndrome/
15 cat cry syndrome/
16 de Lange syndrome/
17 Schilder disease/
18 DiGeorge syndrome/
19 Farber disease/
20 fucosidosis/
21 gangliosidosis/
22 GM2 gangliosidosis/
23 glycogen storage disease type 2/
24 Danon disease/
25 glycogen storage disease type 4/
26 demyelinating disease/
27 Langerhans cell histiocytosis/
28 Huntington chorea/
29 hyperglycinemia/
30 incontinentia pigmenti/
31 Jacobsen syndrome/
32 Kearns Sayre syndrome/
33 angioosteohypertrophy syndrome/
34 myoclonus epilepsy/
35 Leigh disease/
36 Lesch Nyhan syndrome/
37 globoid cell leukodystrophy/

38 metachromatic leukodystrophy/
39 lipidosis/
40 MELAS syndrome/
41 Menkes syndrome/
42 MERRF syndrome/
43 Methylmalonyl-CoA Mutase Deficiency.mp.
44 mevalonate kinase deficiency/
45 mitochondrial encephalomyopathy/
46 mitochondrial myopathy/
47 mucolipidosis/
48 Hurler syndrome/
49 Sanfilippo syndrome/
50 mucopolysaccharidosis type 7/
51 multiple acyl CoA dehydrogenase deficiency/
52 multiple sulfatase deficiency/
53 myoclonus epilepsy/
54 neuroaxonal dystrophy/cn [Congenital Disorder]
55 neuronal ceroid lipofuscinosis/
56 Niemann Pick disease/
57 Lowe syndrome/
58 olivopontocerebellar atrophy/
59 chronic progressive external ophthalmoplegia/
60 ornithine transcarbamylase deficiency/
61 Pelizaeus Merzbacher disease/
62 "disorders of peroxisomal functions"/
63 propionic acidemia/
64 pyruvate carboxylase deficiency/
65 pyruvate dehydrogenase complex deficiency/
66 "disorders of carboxylic acid metabolism"/
67 Refsum disease/
68 infantile Refsum disease/
69 Rett syndrome/
70 Rubinstein syndrome/
71 Sandhoff disease/
72 histiocytosis/
73 sialic acid storage disease/
74 Smith Lemli Opitz syndrome/
75 infantile spasm/
76 lipidosis/
77 metachromatic leukodystrophy/
78 myoclonus epilepsy/
79 von Hippel Lindau disease/
80 Wolf Hirschhorn syndrome/
81 Wolman disease/
82 Zellweger syndrome/

83 Aicardi syndrome/
84 Aicardi? Syndrome.mp.
85 alpha-N-Acetylgalactosaminidase Deficiency.mp.
86 alpha-NAGA deficiency.mp.
87 mannosidosis/
88 argininosuccinic aciduria/
89 Autosomal Recessive Spastic Ataxia of Charlevoix-Saguenay.mp.
90 beta mannosidosis/
91 Dubowitz syndrome/
92 webbed neck/ or Escobar Syndrome.mp.
93 galactosidase/
94 Glutaric Acidemia Type I.mp.
95 Glutaric Aciduria Type I.mp.
96 Glutathione Deficiency.mp.
97 neuroaxonal dystrophy/
98 Jeune Thoracic Dystrophy.mp.
99 Asphyxiating Thoracic Dystrophy.mp.
100 Kanzaki Disease.mp.
101 Lennox Gastaut syndrome/
102 3-methylcrotonyl-CoA carboxylase deficiency.mp.
103 Methylcrotonyl-CoA carboxylase deficiency.mp.
104 3-MCC deficiency.mp.
105 3MCC.mp.
106 MCC deficiency.mp.
107 Duchenne muscular dystrophy/
108 exp lymphatic leukemia/
109 cerebral palsy/
110 neuroblastoma/
111 spinal muscular atrophy/
112 neuroectoderm tumor/
113 acute granulocytic leukemia/
114 "disorders of mitochondrial functions"/
115 Friedreich ataxia/
116 osteosarcoma/
117 trisomy 18/
118 medulloblastoma/
119 hereditary spinal muscular atrophy/
120 pontine glioma/
121 rhabdomyosarcoma/
122 Ewing sarcoma/
123 brain tumor/
124 cystic fibrosis/
125 hypoxic ischemic encephalopathy/
126 trisomy 13/
127 neuronal ceroid lipofuscinosis/

128 Brain stem glioma.mp.
129 brain stem tumor/
130 syndrome CHARGE/
131 ependymoma/
132 liver cell carcinoma/
133 Human immunodeficiency virus infection/
134 microcephaly/
135 "disorders of mitochondrial functions"/
136 adrenoleukodystrophy/
137 bile duct atresia/
138 glioblastoma/
139 Hodgkin disease/
140 Leigh disease/
141 epilepsy/
142 arthrogryposis/
143 astrocytoma/
144 rhabdoid tumor/
145 Burkitt lymphoma/
146 chromosome aberration/
147 Down syndrome/
148 severe myoclonic epilepsy in infancy/
149 glioblastoma/
150 Glutaric aciduria.mp.
151 hydranencephaly/
152 hypoplastic left heart syndrome/
153 globoid cell leukodystrophy/
154 limb girdle muscular dystrophy/
155 "inborn error of metabolism"/
156 metabolic disorder/
157 metachromatic leukodystrophy/
158 Mitochondrial enzyme complex IV.mp.
159 mitochondrial myopathy/
160 necrotizing enterocolitis/
161 NYD.mp.
162 Pallister Killian syndrome/
163 Pelizaeus Merzbacher disease/
164 microgyria/
165 propionic acidemia/
166 pulmonary hypertension/
167 brain injury/
168 Tay Sachs disease/
169 nephroblastoma/
170 chromosome inversion/
171 Adams Oliver syndrome/
172 Aicardi Goutieres syndrome/

173 Alpers disease/
174 Schilder disease/
175 Aminoacid decarboxylase deficiency.mp.
176 glioblastoma/
177 brain malformation/
178 adrenoleukodystrophy/
179 coronary artery anomaly/
180 anterior horn cell disease/
181 Askin's Tumour.mp.
182 ATRT.mp.
183 DiGeorge syndrome/
184 Menkes syndrome/
185 B cell lymphoma/
186 Rassmussen's Encephalitis.mp. (4)
187 ependymoma/
188 Burkitt lymphoma/
189 heart tumor/
190 cardiomyopathy/
191 brain arteriovenous malformation/
192 chronic kidney failure/
193 kidney failure/
194 chronic lung disease/
195 central nervous system malformation/
196 congenital heart malformation/
197 Fallot tetralogy/
198 de Lange syndrome/
199 Dandy Walker syndrome/
200 severe neurological impairment.mp.
201 failure to thrive/
202 exp *neoplasm/
203 or/1-202 [LLC]
204 adolescent/
205 exp child/
206 exp infant/
207 child*.ti,ot,sh,hw,kw.
208 infant?.ti,ot,sh,hw,kw.
209 adolescent?.ti,ot,sh,hw,kw.
210 exp pediatrics/
211 P?ediatric*.ti,ot,sh,hw,kw.
212 exp juvenile/
213 minor?.ti,ot,sh,hw,kw.
214 youth?.ti,ot,sh,hw,kw.
215 teen?.ti,ot,sh,hw,kw.
216 or/204-215 [children broad strategy]
217 203 and 216 [LLC and Children]

218 hospice care/
219 terminal care/
220 *palliative therapy/
221 *terminally ill patient/
222 *hospice patient/
223 *respite care/
224 *progressive patient care/
225 *long term care/
226 (care adj3 (terminal or Palliative or hospice or respite or bereavement or end-of-life or terminally ill or dying)).mp. [mp=title, abstract, subject headings, heading word, drug trade name, original title, device manufacturer, drug manufacturer, device trade name, keyword]
227 (palliative adj3 (treatment* or medicine or therap* or care)).mp. [mp=title, abstract, subject headings, heading word, drug trade name, original title, device manufacturer, drug manufacturer, device trade name, keyword]
228 or/218-227 [palliative care broad search]
229 217 and 228 [Palliative care in children with LLC]
230 limit 229 to "systematic review"
231 limit 229 to meta analysis
232 Cochrane database of systematic reviews.mp.
233 systematic review?.mp.
234 232 or 233
235 229 and 234
236 limit 229 to evidence based medicine
237 or/230-231,235-236 [reviews EMBASE for palliative care in child with LLC]
238 limit 237 to yr="1974 -Current"
239 or/204-206,210-212 [Children specific search strategy]
240 or/218-223 [palliative care more specific strategy]
241 239 and 240 [children and palliative care]
242 limit 241 to "systematic review"
243 limit 241 to meta analysis
244 241 and 234
245 limit 241 to evidence based medicine
246 or/242-245 [reviews EMBASE for palliative care in child - more specific no key words]
247 limit 246 to yr="1974 -Current"
248 238 or 247
249 241 or 229 [Palliativa care in children and/or LLC total]
250 comment/ or editorial/ or letter/ or news/
251 249 not (248 or 250) [Palliativa care in children and/or LLC primary studies no reviews or comments]
252 limit 251 to yr="2000 -Current" [LLC and/or Children and Palliative care - primary articles after 2000]

A.3 CINAHL Search Strategy

Search	Search Options	Actions
S11	(((MH "Child") OR (MH "Infant") OR (MH "Minors (Legal)") OR (MH "Adolescence") OR (MH "Pediatric Units+") OR (MH "Pediatric Care")) AND ((MH "Terminal Care") OR (MH "Hospice Care") OR (MH "Palliative Care") OR (MH "Resuscitation Orders"))) NOT (S1 OR S2 OR S3 OR S4 OR S5 OR S6 OR S7)	**Limiters** - Published Date: 20000101-20131231 **Search modes** - Boolean/Phrase
S10	(((MH "Child") OR (MH "Infant") OR (MH "Minors (Legal)") OR (MH "Adolescence") OR (MH "Pediatric Units+") OR (MH "Pediatric Care")) AND ((MH "Terminal Care") OR (MH "Hospice Care") OR (MH "Palliative Care") OR (MH "Resuscitation Orders"))) NOT (S1 OR S2 OR S3 OR S4 OR S5 OR S6 OR S7)	**Limiters** - Published Date: 19900101-20131231 **Search modes** - Boolean/Phrase
S9	(((MH "Child") OR (MH "Infant") OR (MH "Minors (Legal)") OR (MH "Adolescence") OR (MH "Pediatric Units+") OR (MH "Pediatric Care")) AND ((MH "Terminal Care") OR (MH "Hospice Care") OR (MH "Palliative Care") OR (MH "Resuscitation Orders"))) NOT (S1 OR S2 OR S3 OR S4 OR S5 OR S6 OR S7)	**Search modes** - Boolean/Phrase
S8	(((MH "Child") OR (MH "Infant") OR (MH "Minors (Legal)") OR (MH "Adolescence") OR (MH "Pediatric Units+") OR (MH "Pediatric Care")) AND ((MH "Terminal Care") OR (MH "Hospice Care") OR (MH "Palliative Care") OR (MH "Resuscitation Orders"))) AND (S1 OR S2 OR S3 OR S4 OR S5 OR S6 OR S7)	**Search modes** - Boolean/Phrase
S7	((MH "Child") OR (MH "Infant") OR (MH "Minors (Legal)") OR (MH "Adolescence") OR (MH "Pediatric Units+") OR (MH "Pediatric Care")) AND ((MH "Terminal Care") OR (MH "Hospice Care") OR (MH "Palliative Care") OR (MH "Resuscitation Orders"))	**Limiters** - Publication Type: Statistics **Search modes** - Boolean/Phrase
S6	((MH "Child") OR (MH "Infant") OR (MH "Minors (Legal)") OR (MH "Adolescence") OR (MH "Pediatric Units+") OR (MH "Pediatric Care")) AND ((MH "Terminal Care") OR (MH "Hospice Care") OR (MH "Palliative Care") OR (MH "Resuscitation Orders"))	**Limiters** - Publication Type: Review **Search modes** - Boolean/Phrase
S5	((MH "Child") OR (MH "Infant") OR (MH "Minors (Legal)") OR (MH "Adolescence") OR (MH "Pediatric Units+") OR (MH "Pediatric Care")) AND ((MH "Terminal Care") OR (MH "Hospice Care") OR (MH "Palliative Care") OR (MH "Resuscitation Orders"))	**Limiters** - Publication Type: Meta Synthesis **Search modes** - SmartText Searching
S4	((MH "Child") OR (MH "Infant") OR (MH "Minors (Legal)") OR (MH "Adolescence") OR (MH "Pediatric Units+") OR (MH "Pediatric Care")) AND ((MH "Terminal Care") OR (MH "Hospice Care") OR (MH "Palliative Care") OR (MH "Resuscitation Orders"))	**Limiters** - Publication Type: Meta Synthesis **Search modes** - Boolean/Phrase
S3	((MH "Child") OR (MH "Infant") OR (MH "Minors (Legal)") OR (MH "Adolescence") OR (MH "Pediatric Units+") OR (MH "Pediatric Care")) AND ((MH "Terminal Care") OR (MH "Hospice Care") OR (MH "Palliative Care") OR (MH "Resuscitation Orders"))	**Limiters** - Publication Type: Meta Analysis **Search modes** - SmartText Searching
S2	((MH "Child") OR (MH "Infant") OR (MH "Minors (Legal)") OR (MH "Adolescence") OR (MH "Pediatric Units+") OR (MH "Pediatric Care")) AND ((MH "Terminal Care") OR (MH "Hospice Care") OR (MH "Palliative Care") OR (MH "Resuscitation Orders"))	**Limiters** - Publication Type: Masters Thesis **Search modes** - SmartText Searching
S1	((MH "Child") OR (MH "Infant") OR (MH "Minors (Legal)") OR (MH "Adolescence") OR (MH "Pediatric Units+") OR (MH "Pediatric Care")) AND ((MH "Terminal Care") OR (MH "Hospice Care") OR (MH "Palliative Care") OR (MH "Resuscitation Orders"))	**Limiters** - Publication Type: Systematic Review **Search modes** - Boolean/Phrase

A.4 LILACS Search Strategy

"cuidado PALIATIVO" or "tratamento PALIATIVO" or "cuidado PALIATIVO a doentes terminais" or "cuidados PALIATIVOs" or "programas de cuidados PALIATIVOs" or "cuidados PALIATIVOs na terminalidade da vida" or "cuidados INTERMITENTES" or "programas de cuidados INTERMITENTES" [Descritor de assunto] and "hospitais pediatricos" or "PEDIATRIA" or "crianca" or "cuidado da crianca" or "saude da crianca" or "servicos de saude da crianca" or "crianca pos-termo" or "crianca pre-escolar" or "criancas" or "criancas pre-escolares" or "ADOLESCENTE" or "ADOLESCENTEs" [Descritor de assunto]

A.5 Grey Literature Search Strategy

Organization/Conference	Website
American Academy of Hospice and Palliative Medicine (AAHPM)	http://www.aahpm.org/resources/
American Academy of Pediatrics (AAP)	http://www.aap.org/
American Cancer Society	www.cancer.org
Association for Children with LifeThreatening or Terminal Conditions and their Families (ACT)	http://www.act.org.uk
Center to Advance Palliative Care	www.capc.org
Children's Hospice and Palliative Care Organization	http://www.childrenshospice.org
Children's Hospice International (CHI)	http://www.chionline.org
Children's Oncology Group	www.childrensoncologygroup.org
ChiPPS of the National Hospice and Palliative Care Organization Children's Project on Palliative/Hospice Services (ChiPPS)	http://www.nhpco.org/resources/pediatric-hospice-and-palliative-care
City of Hope Pain & Palliative Care Resource Center (COHPPRC)	http://www.cityofhope.org/PRC/
Education in Palliative and End-of-Life Care (EPEC)	http://www.epec.net/
End of Life Nursing Education Curriculum	www.aacn.nche.edu/ELNEC
End-of-Life/Palliative Education Resource Center (EPERC)	http://www.eperc.mcw.edu/
Hospice and Palliative Nurses Association	www.hpna.org
National Alliance for Children with Life-Threatening Conditions	http://www.nacwltc.org
National Consensus Project on Quality Palliative Care	http://www.nationalconsensusproject.org
The Children's Room	www.childrensroom.org
The Initiative forPPC (IPPC)	http://www.ippcweb.org/
CAPC National Seminar 2012 - posters	http://www.capc.org/capc-resources/capc-poster-sessions/
Canadian Hospice Palliative Care Association	http://www.chpca.net/
International Congress in Palliative Care	http://www.palliativecare.ca/en/index.html
Canadian Network of Palliative Care for Children	http://cnpcc.ca/

Appendix B . Study CKaracteristics – Full Details

Article	Fraser et al 2013	Keele et al 2013	Dussel et al 2009
Objective	Assessed the impact of specialist pediatric palliative care services (SPPCSs) carried by a pediatric hospice	Compared demographic and clinic characteristics of patients who received PC consultations to those who did not	• Determined association of modifiable clinical factors with parental planning of local of death (LOD) • Explored whether planning of the child's LOD had any impact on patterns of care and the parent's experience with child's EOL
Participants	Children who died from cancer (0-19 years), diagnosed between 1996 to 2009, and who died before Sep 2011	Children (<18 years of age), who died at hospital >5 days after admission, from all causes of death with complete administrative data on charges and hospital admissions between 2001-2011 (patients discharged < 5 days under hospice care were not included)	Children who died from cancer from 2 tertiary centres whose physicians authorized the researchers to contact the family. Deaths occurred between 1990 and 1999. Families were interviews between 1997 and 2001
Study design	Cohort comparison	• Retrospective administrative database analysis • Pediatric Health Information System (PHIS) database developed by collaboration of >40 children's hospitals across the states	• Retrospective cross-sectional survey of bereaved parents • Retrospective Chart review
Observation period	Referral to death	Last admission before death	Last month of life
n	497	24342	140
Data sources	• Secondary data base analysis • Linked data from SPPCSs, Register of Cancer, NHS Hospital episode statistics	Secondary analysis of the Pediatric Health Information System including > 40 hospitals across the country	• Parental survey: 390 questions, partially validated, carried over the phone or in person • Patient charts
Intervention Group	**Hospice Group (n=132)** Patients referred to a specialist palliative care service carried by a pediatric hospice	**PC group (n=919)** PC consultation in the last admission (measured by billing code for ICD9 - PC V66.7)	**Planned LOD (n=88)**
Comparator	**Control group (n = 311)** Patients not referred to hospice services	**No PC (n=23423)** No PC consultation in the last admission (no billing code)	**Did not plan LOD (n=52)**
Setting	Residents in the Yorkshire Health Authority, UK	Children who died across > 40 US Children's Hospitals part of the Children's Hospital Association (USA) database	Dana-Farber Cancer Institute/Children's Hospital Boston, and Children's Hospitals and Clinics of Minnesota, USA
Outcomes	• Primary: Total number of hospital admissions • Secondary: Number of planned hospital admissions; Number of emergency hospital admissions	Age, gender, LOS, major group category diagnostic, medications, procedures in the last admission	EOL planning, EOL support from physicians, use of home care, hospital resources utilization, place of death
Funding	Not disclosed	No external funding was received. The authors disclosed no conflict of interest	No conflict of interest was disclosed. Different sources of funding supported the authors (Agency for Health Research and Quality, National Cancer Institute, Child Health Research Grant from the Charles H. Hood Foundation, Pine Tree Apple Tennis Classic Oncology Research Fund)

Article	Knapp et al 2009	Arland et al 2013	Postier et al 2014
Objective	• Described demographic characteristics, cause and location of death, and expenditure patterns of hospice users and nonusers • Investigated hospice expenditure variations and characteristics of children	Investigated relationship between changes in outcomes and an EOL program	Explored health care service utilization by children prior/after enrollment in home-based PPCP/hospice program carried by a tertiary care provider
Participants	Children who died in and were residents of Florida state (1-21 years) between Jul 2003 - Jun 2006 and were enrolled in the Medicaid program	Children who died of brain tumors (1 month - 19 years), with documented location of death and reason for hospital admission	Children enrolled in the home PPCP/ hospice program (1 to 21 years old) for at least 1 day between 2000- 2010 (excluded children < 1 year old)
Study design	Retrospective administrative data analysis	Pre-post observational study - both periods included hospice care as part of the EOL care	Pre-post observational study
Observation period	Last year of life	• Before standardization: 5 years • After standardization: 10 years	• Before enrollment: 12 months • After enrollment: 12 months
n	1527	114	425
Data sources/ measurement	Medicaid claims, encounter and enrollment files, death certificate	Retrospective chart review	• Retrospective secondary data analysis • Electronic medical records and accounting system for billed charges
Intervention Group	Hospice use (n= 85)	After group (n= 92 / 1996-2005) Standardized EOL care program coordinated by a hospital (comprehensive EOL discussions, medications for symptom control, primary family liaison, home visits)	Pre-PPCP
Comparator	Non-hospice use (n= 848)	Before Group (n= 22 /1990-1995) Non standardized EOL care managed by individual hospices in the geographic area (not specialized in pediatric palliative care)	Post-PPCP
Setting	Florida, USA	Children's Hospital Colorado, Colorado, USA (program implemented in 1995).	Children's Hospitals and Clinics of Minnesota's (CHC) Homecare, Pain Medicine, Palliative Care & Integrative Medicine Programs, Minnesota, USA
Outcomes measured	• Hospice use • Hospice expenditures	• Symptoms • Hospitalizations (number and LOS) • Location of death	Change in number of hospitalizations, LOS, and total billed charges for hospital/ER stays
Funding	No conflict of interest was disclosed. Source of funding not disclosed	Did not state funding. Authors reported no conflict of interest but some of them occupied positions in Children's Hospital Colorado.	No funding was received for the research. Authors disclosed no conflict of interest. However, 4 authors were employees in the Department of Pain Medicine, Palliative Care & Integrative Medicine, Children's Hospitals and Clinics of Minnesota.

Article	Gans et al 2012	Pascuet et al 2010	Smith et al 2013
Objective	Demonstrated shift in health care resource use and cost with the implementation of a community palliative care program	Measured differences in hospital utilization and costs with the use of respite services at a pediatric hospice	Evaluated PPCP utilization among the most costly hospitalized patients Examined factors associated with receipt of PPCP and inpatient costs.
Participants	Children living with life-threatening conditions (0 to 20 years old), enrolled in a community based pediatric palliative care program (implemented in 2010)	Children with life-limiting illnesses (age range not defined) who used 'Respite' at the pediatric Hospice at least once from May 2005 to Feb 2009	The 10% most costly patients, in 2010, among all patients discharged from Primary Children's Medical Center (PCMC)
Study design	• Pre-post assessment of health care utilization and expenditures • Brief report	Pre-Post observational study	Cohort comparison between who received PPCP and those who did not Pre-post assessment in the PPCP cohort before/after the initial PC consultation
Observation period	• Before: 12 months? (2009, first and last months unclear) • After: 18 months (January 2010 to September 2011)	• Before 1st respite: 12 months • After 1st respite: 12 months	Cohort comparison: up to 2 years Pre-post: undisclosed
n	123	66	1001
Data sources/ measurement	• Secondary analysis of claims databases (MIS/DSS claims, MEDS and CMS Net) • Family quality of life and satisfaction survey	• Retrospective chart review • Non-randomized	Undisclosed
Intervention Group	**After PPCP** • Included coordination of care and community resources, massage, art, play and music therapy • Family education and training in devices operation • Family counseling and bereavement, pain and symptom management, respite out of home, hospice facilities (not necessarily specialized in pediatric population)	**Before respite**	**PPCP Group** (n=81): patients who used the program
Comparator	**Before PPCP**	**After respite**	**Control Group** (n= 920): patients who did not use the program
Setting	• 11 counties in California, USA • Program included several health care providers (home care providers, hospices and contract agencies who voluntarily participated in the program	• Roger's House Pediatric Hospice (RH), Ontario, Canada • Children's Hospital of Eastern Ontario (CHEO), Ontario, Canada	Primary Children's Medical Center (PCMC), Salt Lake City, Utah, USA
Outcomes measured	• LOS • Medical Expenditures • Family's quality of life and satisfaction	• LOS • ER and Outpatient visits • Overall Cost in hospital/hospice admission	• Cost • Demographics • Use of technology
Funding	Policy brief supported by Children's Hospice & Palliative Care Coalition (CHPCC) All authors belonged to UCLA University.	Funded by the Hospice	Did not state funding. First author is employed by the hospital where the research was conducted

Article	Ward-Smith et al 2008	Belasco et al 2000
Objective	Compared inpatient hospital costs associated with PPCP carried by a tertiary provider	Compared cost of care at home versus at the hospital
Participants	• Children enrolled in the PPCP within 6 months prior to death (age range not specified) • Cases: identified within 18 months, 2 years after PPCP became fully implemented • Controls: criteria for matching not stated (potentially by diagnosis), period not specified • Exclusion criteria: children in the neonatal intensive care unit; those who died within 72 hours of initial admission; patients with incomplete medical records; and patients who enrolled in the PPCP program less than 30 days to death.	• Children referred to a home based pediatric palliative care program between 1988-1992 (age bracket not specified) carried by a tertiary care provider • Applied costs from 1995 and 1996. • Of 154 patients enrolled in the PPCP during the study period, some were selected to reflect medically complicated patients whose level of care at home was comparable to being at the hospital and differed only in palliative intent rather than intent to cure.
Study design	Retrospective matched case-control	Case series
Observation period	6 months prior to death	1 day
n	18	3
Data sources/ measurement	Hospital-based charges	Retrospective chart review
Intervention Group	**PPCP group (n=9)** Enrolled in the Pediatric Palliative Care Program	**Home care**
Comparator	**Non PPCP (n=9)** Not enrolled in the Pediatric Palliative Care Program	**Hospital care**
Setting	Children's Mercy Hospital, Kansas, USA	Children's Hospital Philadelphia, Pennsylvania, USA
Outcomes measured	• Total hospital costs • LOS • Differences in types of procedures	• Type of interventions delivered. • Place of death • Comparison of charges of care
Funding	No funding was disclosed. Authors were employees of Children's Mercy Hospital	No funding was disclosed. Authors were employees of Children's Hospital Pennsylvania.

Appendix C . Study Design Features for Non-Observational Studies

Article		Fraser et al 2013
Allocation		Individual level
Study design		RCS
Study design features		**Support for judgment**
Was there a comparison:		
Between 2 or more groups of clusters receiving different interventions?	Y	Children referred to a hospice service compared to those not referred within a health authority involving all the hospitals in the area. Regression model also allowed for comparison within group over time
Within the same group of clusters over time?	Y	
Were participant/clusters allocated to groups by:		
Concealed randomization?	N	Record of acceptance onto the program from the Pediatric Hospice. It's unknow wheter in the same period only 1/3 of the cohort was referred to the hospice program (family preferences, stigma, distance to the hospice, etc)
Quasi-randomization?	N	
By other action of researchers?	N	
Time differences?	N	
Location differences?	U	
Policy/public health decisions?	Y	
Cluster preferences?	U	
Some other process? (specify)	U	
Which parts of the study were prospective:		
Identification of participating clusters?	N	Administrative database analysis - entirely retrospective
Assessment of baseline and allocation to intervention?	N	
Assessment of outcomes?	N	
Generation of hypotheses?	U	
On what variables was comparability between groups assessed:		
Potential confounders?	Y	"- The regression model controlled for confounders including the covariates: age at diagnosis, disease category, gender and deprivation category.
Baseline assessment of outcome variables?	N	
Other potential sources of bias/confouding/limitations/comments		
-There was no comparison of the outcome variable before the "referral" point between the groups to check for baseline differences. - Whether the patients where still in disease-directed treatment in both groups was not measured and/or controlled, and could be an explanatory factor for decrease in planned admissions. - The authors didn't include days spent in hospice for the referred group to complement the total number of admissions for that group. It might conceal some shifting in resource utilization important to be measured in terms of health care resources consumption. - In the hospice group, median time from the diagnosis date to referral was calculated by cancer category and then applied to the same category in the control group, to create a point for comparison before/after referral. Interquartile range for time to referral varied widely between categories from 85 to over 1100 days. - Negative binomial regression modeling was used including each person's post-referral observation period time in the model as an exposure term. - The patients who did not linked to the NHS hospital admission system (10.1%) differed from the patients included in this analysis and tended to be male, diagnosed under age of 5, and diagnosed towards the beggining of the stydy period. - Among the patients included in the analysis, the groups did differ in some demographics such as smaller % of patients between 15-19 referred to the hospice services, and disease category of Central Nervous System being the largest group disease among those referred to the hospice.		

Article	**Keele et al 2013**	
Allocation	Individual level	
Stydy design	RCS	
Study design features	**Support for judgment**	
Was there a comparison:		
Between 2 or more groups of clusters receiving different interventions?	Y	Database from >40 hospital acrros USA. Didn't compare caractheristics over time other than proportion of patients accessing PC services
Within the same group of clusters over time?	N	
Were participant/clusters allocated to groups by:		
Concealed randomization?	N	
Quasi-randomization?	N	
By other action of researchers?	N	
Time differences?	N	
Location differences?	N	
Policy/public health decisions?	Y	
Cluster preferences?	U	Based on billing code, which changed over time according to guidelines for PC
Some other process? (specify)	U	
Which parts of the study were prospective:		
Identification of participating clusters?	N	
Assessment of baseline and allocation to intervention?	N	
Assessment of outcomes?	N	
Generation of hypotheses?	N	Administrative database analysis - entirely retrospective
On what variables was comparability between groups assessed:		
Potential confounders?	Y	LOS and Cost were not adjusted for other possible confounders, other than geography. Differences in the distributions of some caractheristics were presented (age, health insurance, race, diagnosis)
Baseline assessment of outcome variables?	N	
Other potential sources of bias/confouding/limitations/comments		
- Children who received PC consultations in the last admission before death were different in some characteristics such as older age, race distributions with less access by blacks, having more private insurance, and increase access along the years. - Diseases categories varied significantly. In a subgroup analysis of complex chronic conditions(CCCs) patients (85% of the entire cohort) compared to those not having CCCs, patients with CCCs were more likely to have had a PC consultation (RR 2.2; 95% CI 1.7–2.8). - Comparison included all causes of death, no subgroup analysis for CCC group were presented on the differences in demographics and clinical characteristics. - The authors discussed limitations of the study regards to exclusion of patients discharged under hospice program and admissions < 5 days which may have underestimated the total numbers. - Changes in coding practices and maturation of PC services also represent a potential bias because it cannot be measured.		

Article	**Dussel et al 2009**	
Allocation	Individual level	
Stydy design	RCS	
Study design features		**Support for judgment**
Was there a comparison:		
Between 2 or more groups of clusters receiving different interventions?	Y	Cross-sectional survey with retrospective chart review, that originate a retrospective cohort comparison.
Within the same group of clusters over time?	Y	
Were participant/clusters allocated to groups by:		
Concealed randomization?	N	Children from 2 clusters were separate in 2 groups (had or had not planned the LOD by their parents) based in the survey response
Quasi-randomization?	N	
By other action of researchers?	Y	
Time differences?	N	
Location differences?	N	
Policy/public health decisions?	Y	
Cluster preferences?	U	
Some other process? (specify)	U	
Which parts of the study were prospective:		
Identification of participating clusters?	N	Retrospective chart review Cross-sectional survey
Assessment of baseline and allocation to intervention?	N	
Assessment of outcomes?	N	
Generation of hypotheses?	U	
On what variables was comparability between groups assessed:		
Potential confounders?	Y	For the determinants of having or not planned LOD there was some control for confounders. For the health resource utilization no confounding was addressed.
Baseline assessment of outcome variables?	N	The impact of LOD planning on health care resources was a secondary outcome and was not controlled for any confounder or further explored.
Other potential sources of bias/confouding/limitations/comments		
- Eligibility of the families depended upon physician's consent, which was declined for 19 families. It might introduce some selection bias. - Only 1 parent was interview which might have introduced some non-response bias. - Some Interviews were done long time after the fact which might represent somo recall bias (median 3 years). - Response rate 64%. The non-respondents were similar at child's age at death and diagnosis. - The study used regression with stepwise approach to study the determinants of planning LOD and control for confounders. The authors run sensitivity analysis for missing data and by physicians cluster. No differences in the results were shown. - Children with hematological cancer, those who died from treatment related complications, those families who were very religious were less likely to have planned LOD. - Children who had private insurance, families who had experience previous losses, those who reported that oncologist clearly explained treatment options and those who access home care were more likely to have planned LOD.		

Article	**Knapp et al 2009**	
Allocation	Individual level	
Stydy design	RCS	
Study design features		**Support for judgment**
Was there a comparison:		
Between 2 or more groups of clusters receiving different interventions?	Y	Included children from several hospitals and hospice catchment areas within province
Within the same group of clusters over time?	Y	
Were participant/clusters allocated to groups by:		
Concealed randomization?	N	The authors allocated the 2 groups based on claims for hospice services. It has its limitations regarded to unbilled and unpaid services, which was observed since 5 patients in the non hospice users group had died in hospice.
Quasi-randomization?	N	
By other action of researchers?	Y	
Time differences?	N	
Location differences?	N	
Policy/public health decisions?	Y	
Cluster preferences?	U	
Some other process? (specify)	U	
Which parts of the study were prospective:		
Identification of participating clusters?	N	Administrative database entirely retrospective
Assessment of baseline and allocation to intervention?	N	
Assessment of outcomes?	N	
Generation of hypotheses?	U	
On what variables was comparability between groups assessed:		
Potential confounders?	N	There was soubgroup analysis per diagnosis group but no regression was carried, controling for other covariates sucs as gender, race, time enrolled in the insurance to determine the differences between groups in health care expenditures. No statistical test was applied to differences between group in health care expenditures.
Baseline assessment of outcome variables?	N	
Other potential sources of bias/confouding/limitations/comments		
Although the authours found some patients caractheristics to be associated to more or less hospice use, when analysing the expenditures, only subgroup analysis by diagnostic category were presented. No other factor was control as confounder (gender, race and time enrolled in the Medicaid program, place of death). The authors discussed the limitations of the study such as the limited generalizability for children with private insurance or uninsured, which represents 2/3 of the pediatric population dying in the province.		

Article	**Arland et al 2013**	
Allocation	Group Level	
Stydy design	ChBA	
Study design features	**Support for judgment**	
Was there a comparison:		
Between 2 or more groups of clusters receiving different interventions?	N	Children with brain tumour from a pediatric-oncology in a single hospital that implemented a EOL program
Within the same group of clusters over time?	Y	
Were participant/clusters allocated to groups by:		
Concealed randomization?	N	Study Before/after the implementation of a standardized EOL program carried by a hospital
Quasi-randomization?	N	
By other action of researchers?	N	
Time differences?	Y	
Location differences?	N	
Policy/public health decisions?	Y	
Cluster preferences?	na	
Some other process? (specify)	na	

Which parts of the study were prospective:		
Identification of participating clusters?	N	Chart review entirely prospective
Assessment of baseline and allocation to intervention?	N	
Assessment of outcomes?	N	
Generation of hypotheses?	U	

On what variables was comparability between groups assessed:		
Potential confounders?	N	Authors disclosed not having addressed any potential confounders and dificulties such as missing data (demographics), unclear EOL period before the program was implemented and changes in treatment course/disease management
Baseline assessment of outcome variables?	na	

Other potential sources of bias/confouding/limitations/comments

The groups had different criteria to determine EOL period with several individuals in the historical control having that determine by based on radiology reports of the disease progression. It doesn't mean they had been treated as EOL patients. The intervention group had a date for EOL discussion, referral to hospice or complete DNR order.
The historical control cohort period was reduced because there was no formal onco-pediatric program previous to this date compromising the quality of data quality.
Authors explain exclusion of only 22/52 patients excluded from the initial cohort of 166 patients.
The authors aimed to measure symptoms but didn't present any data on that other than hospitalizations.
In the discussion session authors stated fewer complication after the implementation of the program but didn't show data.
No demographic data comparison was presented. No ethics approval was mentioned.
Although the authors extensively stated the limitations for the study such as temporality, demographics information missing, no symptom measurement scale available, maturation of the disease management and EOL care, changes in health insurance policies, no statistical analysis were applied to some outcomes presented.

Article	**Postier et al 2014**		
Allocation	Individual level		
Stydy design	ChBA		
Study design features		**Support for judgment**	
Was there a comparison:			
Between 2 or more groups of clusters receiving different interventions?	N	Children enrolled in the PPC program carried by a tertiary provider Pre/Post cost and hospital admissions comparison	
Within the same group of clusters over time?	Y		
Were participant/clusters allocated to groups by:			
Concealed randomization?	N	Authors classified the pre/post period based on the first day to the PPC/hospice program utilization	
Quasi-randomization?	N		
By other action of researchers?	Y		
Time differences?	N		
Location differences?	N		
Policy/public health decisions?	N		
Cluster preferences?	U		
Some other process? (specify)	U		
Which parts of the study were prospective:			
Identification of participating clusters?	N	Administrative database entirely retrospective	
Assessment of baseline and allocation to intervention?	N		
Assessment of outcomes?	N		
Generation of hypotheses?	N		
On what variables was comparability between groups assessed:			
Potential confounders?	Y	Multivariate regression accounting for exposure to the program, disease group and study period	
Baseline assessment of outcome variables?	Y		
Other potential sources of bias/confouding/limitations/comments			
As any other pre/post design without a control group for comparison, if the decrease in LOS and charges observed are due to the PPC program or a natural trend among those type of patients. Its not clear the proportion of patients who died at the hospital/home, which would deeply affect charges closer to death. Selection bias regardless to the referral to the program is always present in this type of program. Charges with home care were not accounted for. Non-parametric test applied to compare the outcomes pre/post doesn't take into account the different time exposed to the program or time/per person/in the post period of the study which may overestimated the diferences pre/post. Authors do not report the estimates from the regressions.			

Article	**Gans et al 2012**	
Allocation	Individual level	
Stydy design	ChBA	
Study design features	**Support for judgment**	
Was there a comparison:		
Between 2 or more groups of clusters receiving different interventions?	Y	Children enrolled in the community palliative care program in California, using several health care providers in the different counties
Within the same group of clusters over time?	Y	
Were participant/clusters allocated to groups by:		
Concealed randomization?	N	
Quasi-randomization?	N	
By other action of researchers?	N	
Time differences?	Y	
Location differences?	N	
Policy/public health decisions?	Y	
Cluster preferences?	U	Before-after enrollment in the program criteria not clearly stated. It seems to be a registry for the enrollees.
Some other process? (specify)	U	
Which parts of the study were prospective:		
Identification of participating clusters?	N	
Assessment of baseline and allocation to intervention?	N	
Assessment of outcomes?	N	
Generation of hypotheses?	N	Administrative database entirely retrospective
On what variables was comparability between groups assessed:		
Potential confounders?	N	Authors did not address confounders that could influence the outcomes such as diagnosis type, cities, age, availability of services, proximity to death, etc
Baseline assessment of outcome variables?	N	
Other potential sources of bias/confouding/limitations/comments		
The enrollment in the program depended on financial criteira to be covered by MediCal. Which included life-threatning conditions and were expanded to all conditions expected to consume more than 30days/year of hospital admissions. Not clear if all the patients enrolled in the same point in time, and if the before and after expenditures were flagged as such, independent of how long they were under the program. Unbilled or unpaied claims were excluded from the data, possibily overestimating cost savings. Survey used a likert scale of 4 points the author's called quality of life. No validation mentioned. No control group was used to compare natural trends in shift of health care resources utilization. The authors briefly mention certain limitations of the study and the need to use full administrative data with control, to better estimate the differences suggested by this report on the shift of health care resource allocation.		

Article	**Pascuet et al 2010**	
Allocation	Individual level	
Stydy design	ChBA	
Study design features		**Support for judgment**
Was there a comparison:		
Between 2 or more groups of clusters receiving different interventions?	N	Children who used the respite admission at least once, had their total hospital/hospice admissions measured before and after the access of the first respite
Within the same group of clusters over time?	Y	
Were participant/clusters allocated to groups by:		
Concealed randomization?	N	It is not clear whether the groups were determine by the date of hospice opening, or the date of first utilization of respite services from a pediatric hospice
Quasi-randomization?	N	
By other action of researchers?	N	
Time differences?	N	
Location differences?	N	
Policy/public health decisions?	Y	
Cluster preferences?	U	
Some other process? (specify)	U	
Which parts of the study were prospective:		
Identification of participating clusters?	N	Administrative database entirely retrospective
Assessment of baseline and allocation to intervention?	N	
Assessment of outcomes?	N	
Generation of hypotheses?	N	
On what variables was comparability between groups assessed:		
Potential confounders?	N	Authors did not address confounders that could influence the outcomes such different types of inpatient utilization, diseases categories age or proximity to services.
Baseline assessment of outcome variables?	N	
Other potential sources of bias/confouding/limitations/comments		
The authors stated that the cost for inpatient admissions at the hospital had a fixed cost per day (based on 2007 cost), based on the interprovincial billing rate (including direct health care cost and overhead costs). Costs were not differentiated per type of admission - general, critical care. Not clear if costs included emergency and outpatients visits, and how their cost were addressed. Cost for hospice care was calculated by average cost per day , being the anual hospice budget /number of beds per year. It seems that hospice only provided respite care. Not clear if all patients included had 24 months of observation period. Not clear, in case of shorter observation period, if the outcomes were weighted by time in the study. The authors recognize the limitations of the different cost analysis in each institution.		

Article	Smith et al 2013	
Allocation	Individual level	
Stydy design	ChBA/RCS	
Study design features		**Support for judgment**
Was there a comparison:		
Between 2 or more groups of clusters receiving different interventions?	N	Children discharged from a single tertiary care provider
Within the same group of clusters over time?	Y	
Were participant/clusters allocated to groups by:		
Concealed randomization?	N	Authors classified the groups based on utilization of PPC program consultation
Quasi-randomization?	N	
By other action of researchers?	N	
Time differences?	Y	
Location differences?	N	
Policy/public health decisions?	Y	
Cluster preferences?	na	
Some other process? (specify)	na	
Which parts of the study were prospective:		
Identification of participating clusters?	na	Abstract doesn't bring enough information on the methods
Assessment of baseline and allocation to intervention?	na	
Assessment of outcomes?	na	
Generation of hypotheses?	na	
On what variables was comparability between groups assessed:		
Potential confounders?	N	Authors did not controll for any confounders
Baseline assessment of outcome variables?	N	
Other potential sources of bias/confouding/limitations/comments		
Abstract presented at a conference. It doesn't bring enough information about the methods applied in this research. We are unable to evaluate risk of bias, selection and identification of participants, intervention definition. The authors didn't control for differences in the population found in the research such as gender, comorbidities, technology dependence.		

Article	Ward-Smith et al	
Allocation	Group Level	
Stydy design	CC	
Study design features		**Support for judgment**
Was there a comparison:		
Between 2 or more groups of clusters receiving different interventions?	Y	Cases and controls at 1 hospital who carried the PPC program
Within the same group of clusters over time?	Y	
Were participant/clusters allocated to groups by:		
Concealed randomization?	N	The authors chose the cases and controls, not randomly but made to provide a range of diagnostics and enrollment in the PPCP within 6 months before death.
Quasi-randomization?	N	
By other action of researchers?	Y	
Time differences?	U	
Location differences?	N	
Policy/public health decisions?	N	
Cluster preferences?	U	
Some other process? (specify)	U	
Which parts of the study were prospective:		
Identification of participating clusters?	N	Administrative database entirely retrospective
Assessment of baseline and allocation to intervention?	N	
Assessment of outcomes?	N	
Generation of hypotheses?	N	
On what variables was comparability between groups assessed:		
Potential confounders?	N	None
Baseline assessment of outcome variables?	N	
Other potential sources of bias/confouding/limitations/comments		
Although the authors named the study as case-control, it is technically a cohort comparison, where the cohorts were distinct by the intervention – received services from the PPCP. Among the 133 possible cases identified under the inclusion criteria, 9 were chosen by the authors. This choice was not random but made by the authors to provide a range of diagnostics and because they had being enrolled in the PPCP within 6 months before death. Do not state the matching criteria and if it was randomly selected or, as the cases, chosen by nurses. Not clear if the controls were contemporary to the cases or if they were selected from the period before the implementation of the program. Controls were slightly different in gender, and race. It doesn't specify if the cost was adjusted to reflect the inflation, or if they incurred in the same period for cases and controls.		

Article	**Belasco et al**	
Allocation		
Stydy design	CR/CS	
Study design features		**Support for judgment**
Was there a comparison:		
Between 2 or more groups of clusters receiving different interventions?	na	
Within the same group of clusters over time?	na	Case series with 3 patients
Were participant/clusters allocated to groups by:		
Concealed randomization?	N	Out of the 154 patients enrolled in the PPCP during the period, some were selected by the author to reflect medically complicated patients whose level of care at home approximatelly equal that in the hospital and differed only in palliative intent rather than intent to cure.
Quasi-randomization?	N	
By other action of researchers?	Y	
Time differences?	N	
Location differences?	N	
Policy/public health decisions?	N	
Cluster preferences?	U	
Some other process? (specify)	U	
Which parts of the study were prospective:		
Identification of participating clusters?	N	
Assessment of baseline and allocation to intervention?	N	
Assessment of outcomes?	N	
Generation of hypotheses?	N	Administrative database entirely retrospective
On what variables was comparability between groups assessed:		
Potential confounders?	na	
Baseline assessment of outcome variables?	na	None
Other potential sources of bias/confouding/limitations/comments		
Do not state how the patients were selected. Do no describe how the number and types of procedures for charges comparison were measured and the comparison was created. It's not clear if the type of prcoedures were compared to a control or if it was estimated to adapt to the home care model for the same patient, or if it was measured from the same patient in both settings. The authors stated that for home care, because the way the insurances operate locally, charges per day did not included physicians home visit, social worker, coordinator of care, skilled nurse visits longer than 2 hours. ALso, visits and procedures not authorized by insurance were not included, which may represent part of the out-of-pocket expenses for families, and not reflected in this comparison. Charges do not appropriately reflect costs introducing important measurement bias.		

Appendix D . Newcastle-Ottawa Scale

Assessment of quality of a cohort study – Newcastle Ottawa Scale	**Retrospective Cohort Studies**						**Cohort Before-and-after studies**				**Case-Series**
Selection (tick 1 box in each section)	Keele L et at	Fraser et al	Smith et al	Dussel et al	Knapp et al	* Ward-Smith et al	Postier et al	Arland et al	Gans et al	Pascuet et al	Belasco et al
1. Representativeness of the intervention cohort											
a) truly representative of the average, children, recipient of palliative care ★	☐	☐	☐	☐	☐	☐	★	☐	☐	★	☐
b) somewhat representative of the average, children, recipient of palliative care (only 1 disease category e.g. cancer) ★	★	★	☐	★	★	☐	☐	★	☐	☐	☐
c) selected group of patients, e.g. certain insurance coverage, age specific	☐	☐	☑	☐	☐	★	☐	☐	☑	☐	☑
d) no description of the derivation of the cohort	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
2. Selection of the non intervention cohort											
a) drawn from the same community as the intervention cohort ★	★	★	★	★	★	☐	☐	★	☐	☐	☐
b) drawn from a different source	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
c) no description of the derivation of the non intervention cohort, or no controls	☐	☐	☐	☐	☐	☑	☑	☐	☑	☑	☑
3. Ascertainment of intervention											
a) secure record (eg health care record, claims/billing system) ★	★	★	★	☐	★	★	★	★	★	★	★
b) structured interview ★	☐	☐	☐	★	☐	☐	☐	☐	☐	☐	☐
c) written self report	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
d) other / no description	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
4. Demonstration that outcome of interest was not present at start of study											
a) yes ★	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
b) no	☑	☑	☑	☑	☑	☑	☑	☑	☑	☑	☑
Comparability (tick 1 or both boxes, as appropriate)											
1. Comparability of cohorts on the basis of the design or analysis											
a) study controls for age, sex, exposure to the program (survival), disease ★	☐	★	☐	☐	☐	☐	★	☐	★	☐	☐
b) study controls for any additional factors (e.g. socio-economic status, education, geography) ★	★	☐	☐	☐	★	☐	☐	☐	☐	☐	☐

If the article meets a criterion followed by a ★, the box will appear as a ★. If the article meets a criterion that is not followed by a ★, then the box will appear ticked ☑. If the article does not meet any criteria in the checklist the boxes will not appear ticked ☐. References and manual on how to use the scale from the Ottawa Hospital Research Institute available at http://www.ohri.ca/programs/clinical_epidemiology/oxford.asp

Assessment of quality of a cohort study – Newcastle Ottawa Scale	**Retrospective Cohort Studies**						**Cohort Before-and-after studies**				**Case-Series**
Outcome (tick 1 box in each section)	Keele L et at	Fraser et al	Smith et al	Dussel et al	Knapp et al	* Ward-Smith et al	Postier et al	Arland et al	Gans et al	Pascuet et al	Belasco et al
1. Assessment of outcome											
a) independent blind assessment ★	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
b) record linkage ★	★	★	★	★	★	★	★	★	★	★	★
c) self report	☐	☐	☐	☑	☐	☐	☐	☐	☐	☐	☐
d) other / no description	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
2. Was follow up long enough for outcomes to occur ##											
a) yes, if median duration of follow-up >= 2 months ★	☐	★	★	☐	★	★	★	★	☐	☐	☐
b) no, if median duration of follow-up < 2 months, or unclear ★	☑	☐	☐	☑	☐	☐	☐	☐	☑	☑	☑
3. Adequacy of follow up of cohorts											
a) complete follow up: all subjects accounted for length of exposure to PPCP (survival bias) ★	☐	☐	☐	☐	☐	☐	★	☐	☐	☐	☐
b) subjects lost to follow up unlikely to introduce bias: number lost <= 20%, all ages included, all diseases, or description of those lost suggesting no different from those followed ★	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐
c) follow up rate < 80% (select an adequate %) and no description of those lost, or description suggesting differences from those followed	☑	☑	☑	☑	☑	☑	☐	☑	☑	☑	☑
d) no statement	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐	☐

If the article meets a criterion followed by a ★, the box will appear as a ★. If the article meets a criterion that is not followed by a ★, then the box will appear ticked ☑. If the article does not meet any criteria in the checklist the boxes will not appear ticked ☐. References and manual on how to use the scale from the Ottawa Hospital Research Institute available at http://www.ohri.ca/programs/clinical_epidemiology/oxford.asp

Appendix E . Pairwise Comparison of Admissions and LOS

Figure 27. Pairwise Comparison of Monthly Number of Admissions – Entire Observational Period.

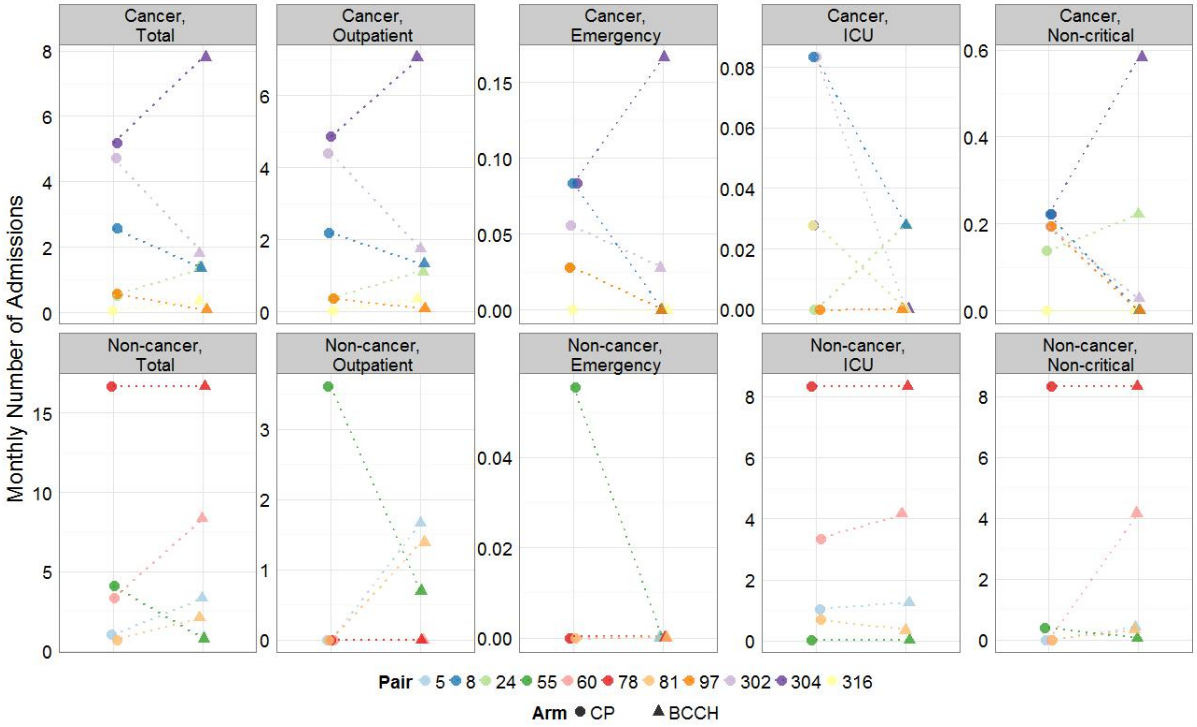


Figure 28. Pairwise Comparison of Monthly Number of Admissions – Pre-Referral Period.

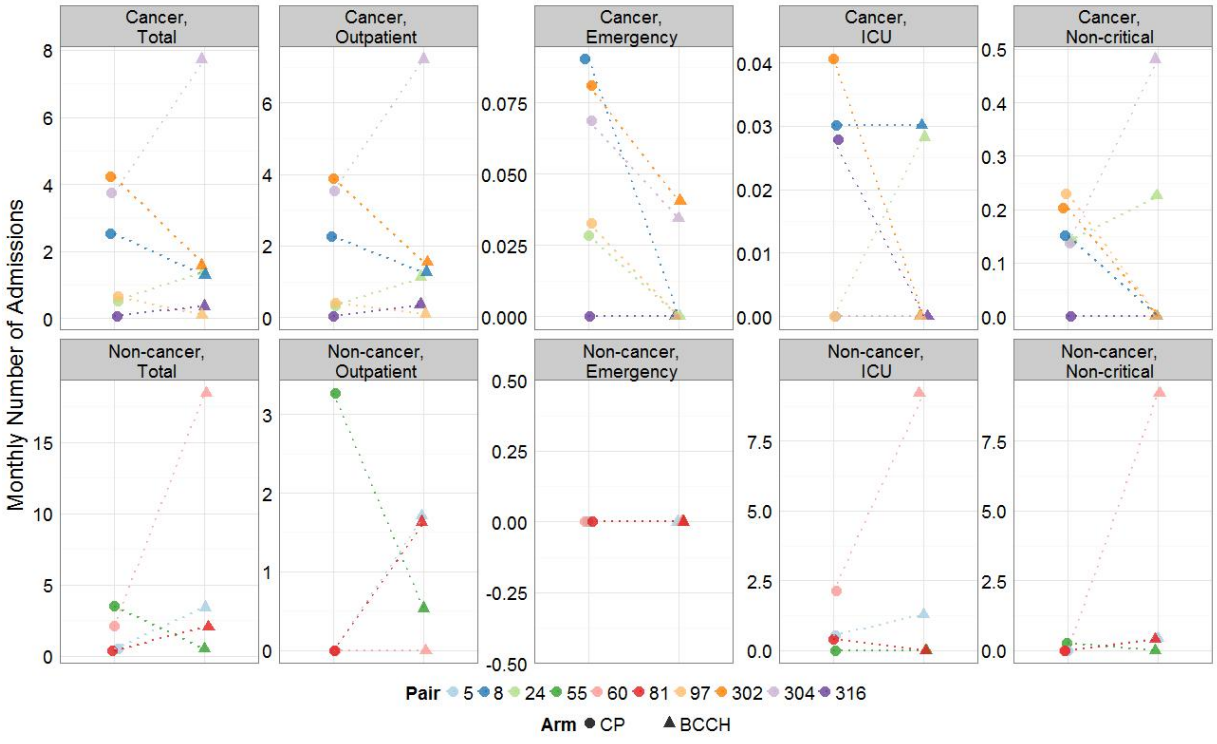


Figure 29. Pairwise Comparison of Monthly Number of Admissions – Post-Referral Period.

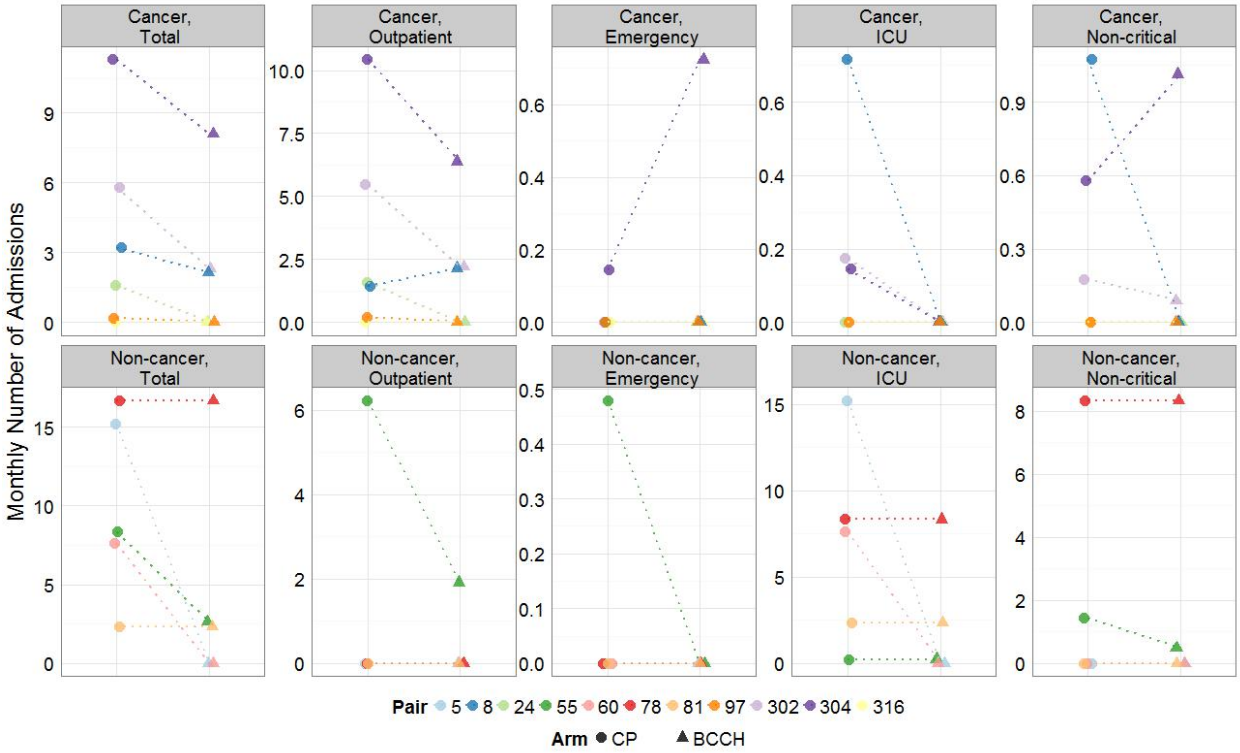


Figure 30. CPCH Group – Changes in Monthly Number of Admissions – Pre- to Post-Referral Period.

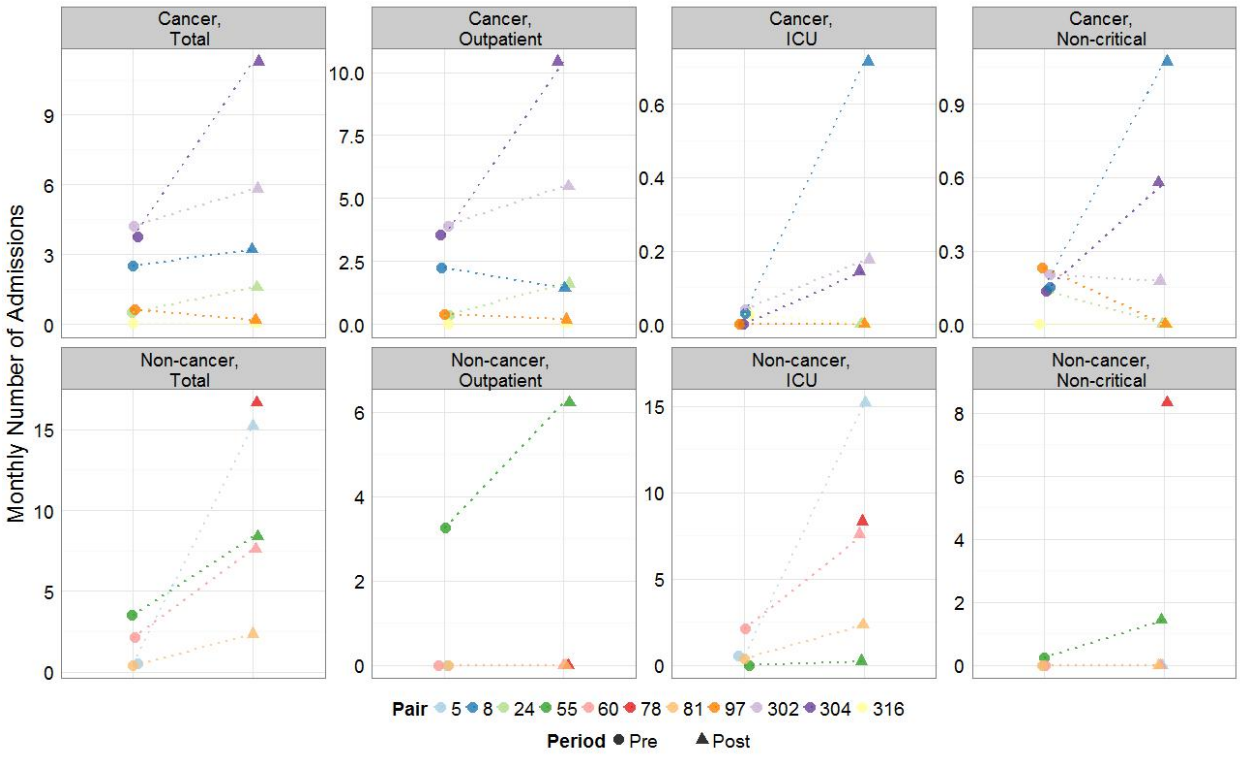


Figure 31. BCCH Group – Changes in Monthly Number of Admissions – Pre- to Post-Referral Period.

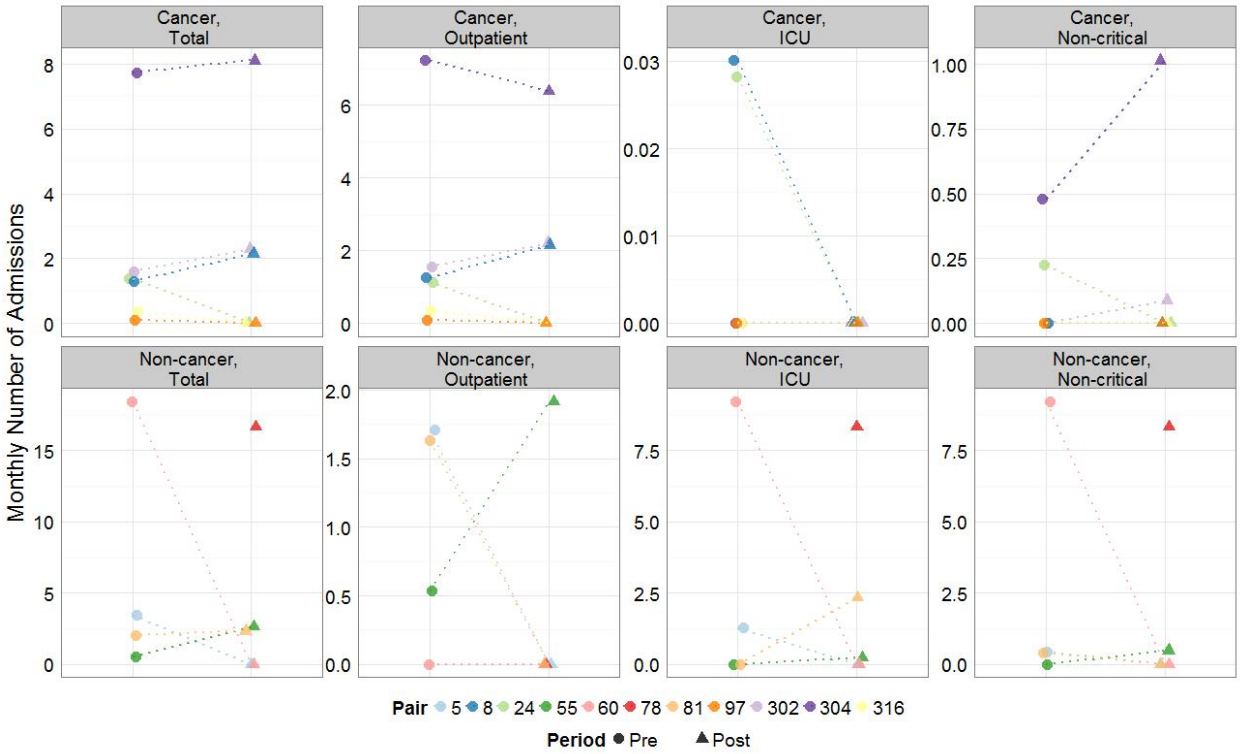


Figure 32. Pairwise Comparison of Monthly LOS – Entire Observational Period.

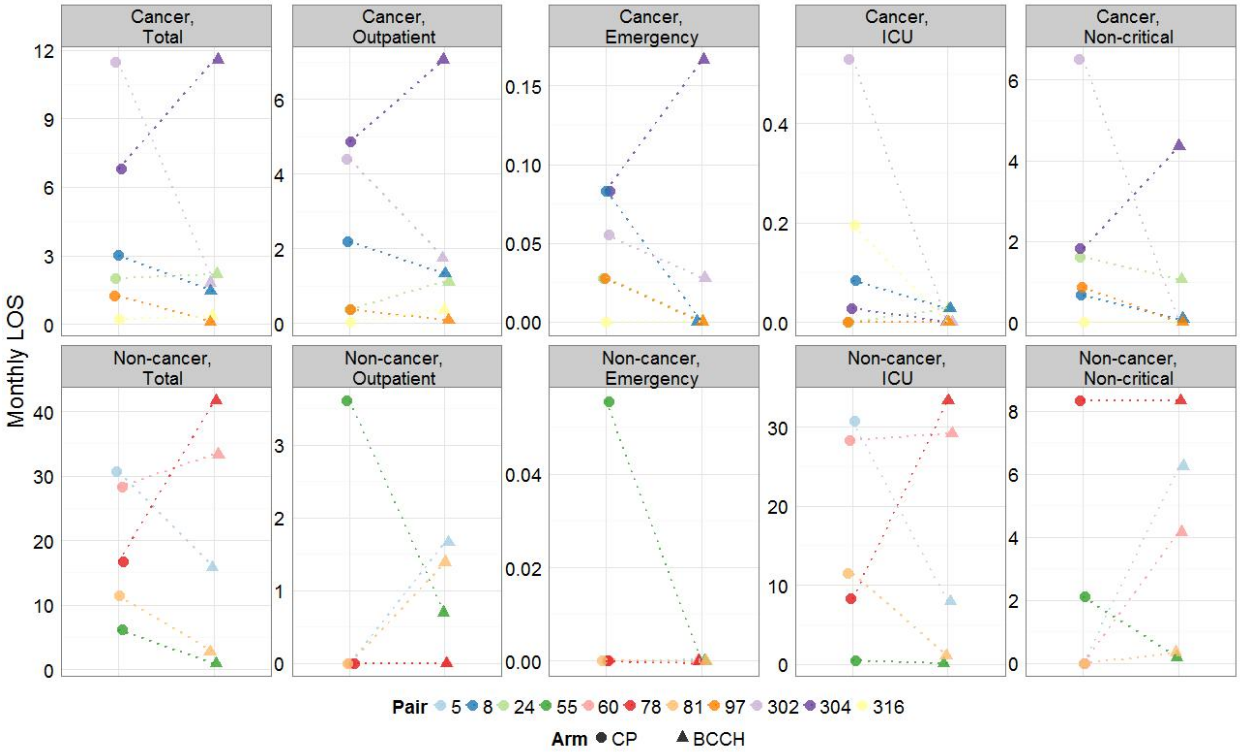


Figure 33. Pairwise Comparison of Monthly LOS – Pre-Referral Period.

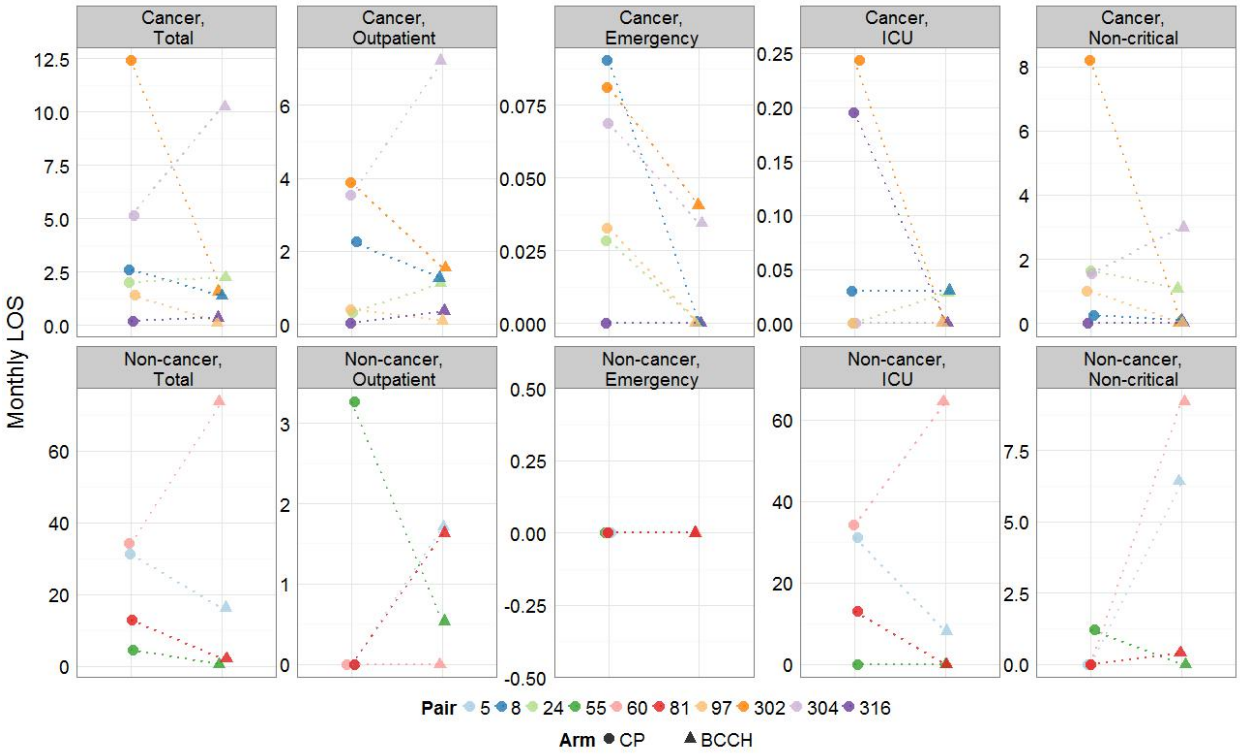


Figure 34. Pairwise Comparison of Monthly LOS – Post-Referral Period.

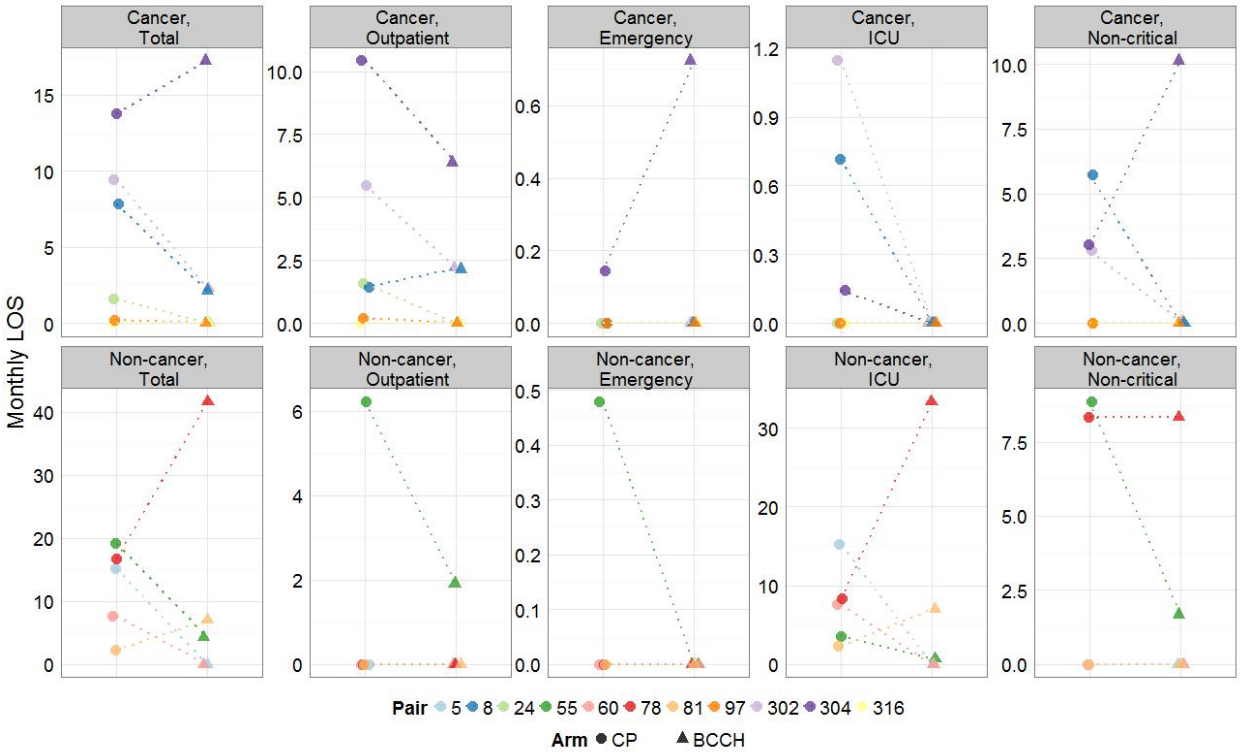


Figure 35. CPCH Group – Changes in Monthly LOS – Pre- to Post-Referral Period.

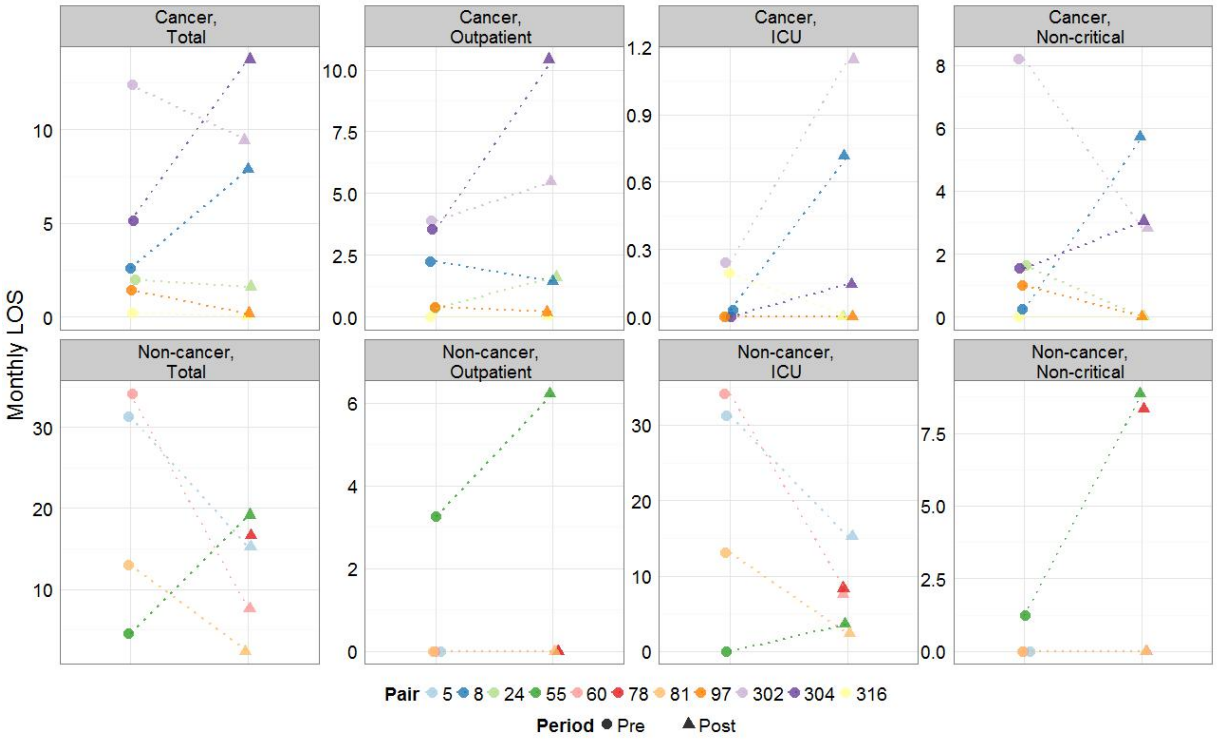
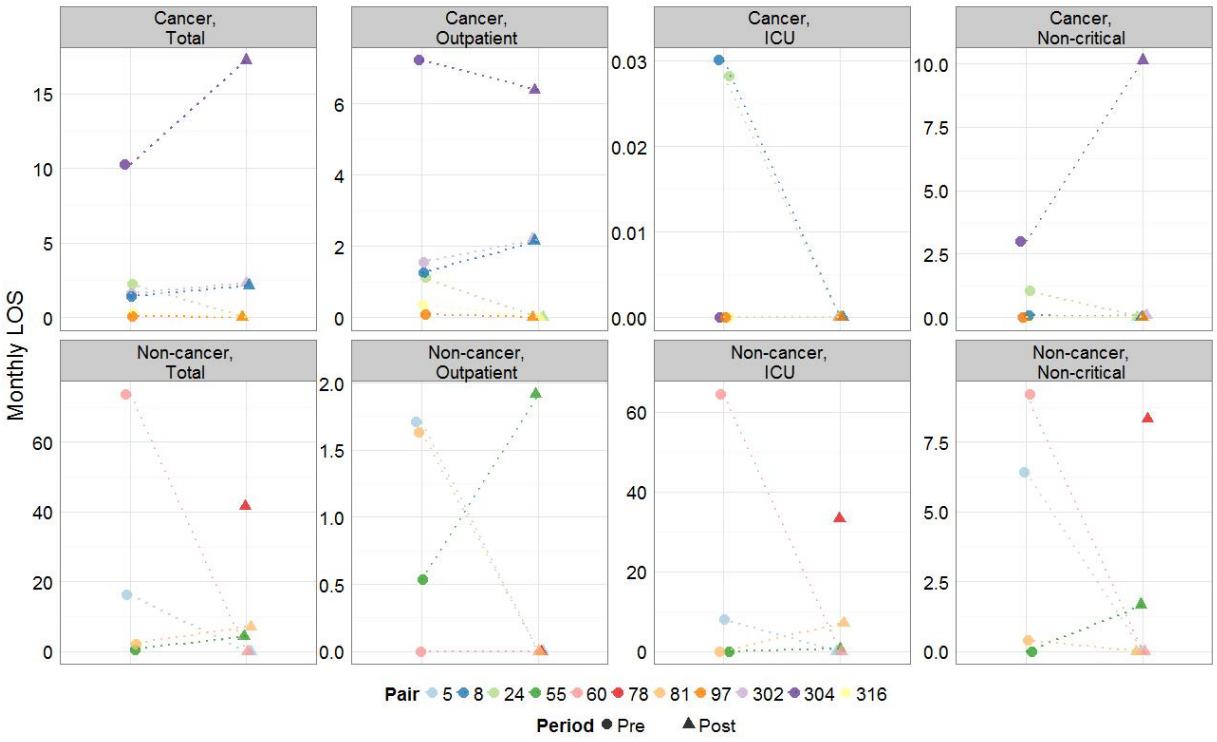


Figure 36. BCCH Group – Changes in Monthly LOS – Pre- to Post-Referral Period.



Appendix F . CIHI Methodology for Calculation of Inpatient Ward and ICU Hospital Per Diem Rates Using the 2011-2012 Canadian MIS Database[59]

Introduction

This document outlines the methodology for the calculation of Intensive Care Unit (ICU) day rates and non-ICU day (WARD) rates for use in reciprocal billing.

Use of the MIS Standards

Data from the Canadian MIS Database (CMDB) has been standardized to the MIS national chart of accounts. Where provincial, regional, or facility specific accounts are used, they are mapped to a corresponding valid account under the MIS national standard where possible. Compliance of financial and statistical data from the jurisdictions to the MIS Standards is critical to the calculation of comparable inter jurisdictional rates.

Per Diem Logic

The method is based on obtaining the full cost of inpatient services, then dividing by the qualifying inpatient days reported by each hospital. This means that, outside of the specified expense and revenue exclusions, all costs are allocated to patient care services - there are no *residual* costs. The full cost of inpatient services also includes expenses associated with regional health authorities (RHA's), such as diagnostic/laboratory services and/or administration/support expenses. The details of this method are outlined in this document.

1. (a) Clean the clearing accounts

The first step in the calculation is to clean the clearing accounts which represent hospital expenses that pertain to a number of functional centres. The MIS Standards require that these accounts be cleared to zero before data is reported to CIHI. In the event that clearing accounts are not cleared, the methodology in Appendix C will be employed in order to allocate these expenses.

1. (b) Revenues Netted, Expenses Removed

The next step in the calculation is to net recovery revenues with valid expenses in each functional centre and removes certain designated expenses. In determining net expenses, all fund types are included[***]. The secondary codes used to derive net expenses are:

Revenues

Secondary Account Description	**Secondary Accounts**
All Recovery Revenues	12*

Expenses: All expenses except the following:

Secondary Account Description	**Secondary Accounts**
Undistributed Amortization[†††] – Grounds, Buildings and Building Service Equipment	95020*, 95040*, 95060*
Interest on Major Equipment Loans	755*
Interest on Long-Term Liabilities	955*

Functional Centre Exclusions[‡‡‡]

The formula proportionally allocates all overhead and other qualifying expenses to inpatient services. Functional centers containing these expenses are:

Primary Account Description	**Primary Accounts**	**Secondary Accounts**
Administration	711*	ALL
Education	718*	ALL
Undistributed and Accounting Centres	719*/819*	ALL

Expenses in the following functional centres attract overhead/qualifying expense allocations, but are not attributed to inpatient services.

Primary Account Description	**Primary Accounts**	**Secondary Accounts**
Long-Term Care	71292*	ALL
Research	717*	ALL

Community functional centres (715*) also attract overhead/qualifying expense allocations; however, if inpatient visits are reported in community functional centres, then expenses associated with those visits are removed from the community functional centre and placed in the inpatient cost pool (see page 6 for more details).

[***] For the purposes of the methodology, the fund type for all functional centers has been converted to '1'.
[†††] Balances reported by hospitals/regions at the roll-up level for undistributed amortization (ie. F9500000) are multiplied by the generally accepted percentage of (30%) undistributed amortization allocated to equipment amortization.
[‡‡‡] The expenses in these functional centres are not excluded until all allocations have been made.

Cost Pools

Primary Account Description	Primary Accounts	Secondary Accounts
WARD	712* (excluding 71240*, 71260*, 71262*, 71265*, and 71292*)	All
ICU	71240*	All
Outpatient	713*	All
Community	715*	All

Expenses reported under Nursing Inpatient Services (primary account 712*, not including 71292* (Long Term Care), 71260* (Operating Room) and 71265* (Post-Anasthetic Recovery Room)) and 71262* (Combined Operating Room and Post-Anasthetic Recovery Room) were separated at the start of the algorithm into Intensive care unit (ICU) expenses (reported under primary account 71240*) and WARD expenses (reported under primary accounts 712*, excluding 71240* as well as the other exceptions listed previously). This separation will allow for the calculation of an "ICU per diem" and a "WARD per diem".

Nursing workload is the primary cost allocation base for the Inpatient and Ambulatory Care functional centres. However, if there exists a priori knowledge that workload in a given hospital is of poor quality or is unreliable, then it is not used.

Expenses reported in the OR, PARR, Combined OR/PARR and Diagnostic and Therapeutic areas are later allocated to the WARD, ICU, Outpatient and Community cost pools as applicable.

As inpatient units are sometimes the location of treatment for outpatients, and outpatient clinics sometimes the location of the treatment for inpatients, the data is inspected so as to determine whether allocations from these areas are required. All of these allocations are described in #2. For 71340* Specialty Day/night Care and 71350* Specialty Clinics, where level 4 accounts are reported they are used; otherwise, these two functional centres are rolled up to level 3.

2. (a) Inpatient activity in the outpatient departments

Outpatient expenses that are associated with an inpatient stay and inpatient days spent in Ambulatory Care Services functional centres need to be adjusted for. Therefore the following adjustments are required:

For 713* (excluding 71360 Day Surgery Operating Room, 71362 Day Surgery Combined Operating and Post-Anesthetic Recovery Room, 71365 Day Surgery Post-Anesthetic Recovery Room, 71367 Day Surgery Pre- and Post-Operative Care, 71369 Day Surgery Combined Operating and Post-Anesthetic Recovery Room) and 71310 Emergency, where secondary statistical code 4 03 Inpatient Days and/or 4 50 10 Visits – Face-to-Face - Inpatient are reported in corresponding functional centres, a portion of direct expense will be allocated to the inpatient cost pool according to the following rules (in descending order of priority):

Allocation Base	Secondary Code
Use Service Recipient Workload Units:	Total Service Recipient Workload 1* excluding 19* Inpatient Service Recipient Workload 1**1, excluding 19* Outpatient Service Recipient Workload 1**2 and 1**3 , excluding 19*
2(a). If service recipient workload is not reported, then calculate the average inpatient cost per day in all 712 Nursing Inpatient/Resident Services (except 712 40 Intensive Care Nursing Unit, 712 50 Obstetrics Nursing Unit, 71 2 60 Operating Room, 71265 Post-Anesthetic Recovery Room, 71275 Psychiatry/Addiction Nursing Unit, 71280 Psychiatric Long-Term Care) and then use this rate multiplied by the number of inpatient days reported in 713 Ambulatory Care and 71310 Emergency.	Inpatient Days 403*
2(b). If service recipient workload is not reported, then after the allocation of cost per inpatient day in corresponding functional centres, the remainder of the expenses are allocated based in Visits – Face-to-Face	Total Visits - Face-to-Face 450* Inpatient Visits - Face-to-Face 4501* Outpatient Visits - Face-to-Face 4502* and 4503*
3. If service recipient workload is not reported and no inpatient days are reported, then a calculation of inpatient expenses is derived using Visits – Face to Face.	Total Visits - Face-to-Face 450* Inpatient Visits - Face-to-Face 4501* Outpatient Visits - Face-to-Face 4502* and 4503*
4. If neither inpatient days nor inpatient visits are reported then 100% of expenses are allocated to the outpatient cost pool.	

2. (b) Outpatient activity in inpatient departments

Where secondary statistical codes 4502* and 4503* Visits—Face-to-Face—Client are reported in a 712 Nursing Inpatient/Resident Services functional centre (excluding 712 60 Operating Room, 71262 Combined Operating and Post-Anesthetic Recovery Room 71265 Post-Anesthetic Recovery

Room and 71292 Long-Term Care Nursing Unit), expenses will be allocated to the Outpatient Cost Pool according to the following rules in descending order of priority:

Allocation Base	**Secondary Code**
1. Service Recipient Workload Units (if available)	Total Service Recipient Workload 1* excluding 19* Inpatient Service Recipient Workload 1**1, excluding 19* Outpatient Service Recipient Workload 1**2 and 1**3 , excluding 19*
2. If service recipient workload is not reported then use the national average (by type of hospital)[§§§] cost[****] per client visit calculated for all hospitals who reported workload and client visits.	Total Service Recipient Workload 1* excluding 19* Inpatient Service Recipient Workload 1**1, excluding 19* Outpatient Service Recipient Workload 1**2 and 1**3 , excluding 19* Outpatient Visits - Face-to-Face 4502* and 4503*
3. If no outpatient visits are reported, all expenses will be assigned to the inpatient cost pool.	

2. (c) Inpatient Visits Reported in Community Services

Where secondary statistical codes 4501* - Inpatient visits are reported in a 715 Community Services functional centre, expenses will be allocated to the Inpatient Cost Pool as follows:

Allocation Base	**Secondary Code**
A national average cost per visit by type[5] of hospital will be calculated for all hospitals who reported inpatient visits. All visits (inpatient, client, etc.) will be used as the denominator. This national average cost per visit will be calculated at level 4 of the 715 functional centre. Once calculated, it will be multiplied against the number of inpatient visits reported in each hospital to produce the inpatient cost of community services.	Total Visits - Face-to-Face - 450* Inpatient Visits - Face-to-Face - 4501*
If no inpatient visits are reported in Community Services functional centres, then all expenses will remain as Community Services expenses.	

[§§§]. Hospitals are categorized as (i) Small (less than 50 beds), (ii) Non-Teaching and (iii) Teaching.

[****]. Costs are determined by multiplying the direct cost of the Nursing Inpatient/Resident functional centre by the percentage of Client Service Recipient Workload to Total Service Recipient Workload reported in the functional centre.

3. Allocation Methodology - Diagnostic/Therapeutic Services (D&T)

The working group recommended that the preferred method for allocating D&T expenses (i.e. qualifying expenses reported in primary account 714*) to inpatient services is via workload measurement data. All service recipient activity workload is used to derive an inpatient/outpatient ratio.[††††] NOTE: non-service recipient activity workload is excluded, but the expenses associated with non-service recipient activity are allocated using the inpatient/outpatient ratio. Therefore the following formula is used to obtain the inpatient workload ratio:

Inpatient Workload
All Service-Recipient Workload

Service-recipient workload accounts will be used to allocate expenses in Diagnostic and Therapeutic functional centres. The service-recipient categories and the cost pools that they are allocated to in the inpatient per diem model are as follows:

Service Recipient	**Costing Pool**
Inpatient	Inpatient (P712*)
Client	Outpatient (P713*)
Referred-In	Outpatient (P713*)
Resident	Long Term Care (P71292*)
Facility/Organization	Community (715*)
Environmental	Community (715*)
Service Recipient Not Uniquely Identified	Community (715*)

Expenses in Diagnostic and Therapeutic functional centres are allocated based on mandatory MIS reporting requirement level either level4 or level 3. If hospital didn't meet the level 4 MIS minimum reporting requirement, D&T expenses are allocated at level 3.Workload will be used to obtain the ratio for each level 3 or level 4 D&T functional centre. Relevant workload accounts will be used in the appropriate functional centre as described in the table below. Note that although MIS secondary statistical account 115** is no longer valid effective April 1,2010, most jurisdictions are still using this code and it has been included with 118**.

[††††] Negative statistics reported to the CMDB are not used in the allocation of D&T costs. Where negative statistics are reported, they are set to zero.

Diagnostic and Therapeutic Functional Centres

Functional Centre	Inpatient Workload Accounts	Outpatient Workload Accounts	Long-Term Care Workload Accounts	Community Workload Accounts
71 4 05Diagnostic and Therapeutic Nursing 7140515 – Medical Imaging Nursing	1021*	1022*, 1023*	1024*	1025*, 1026*, 1028*, 1029*
71 4 10Clinical Laboratory 7141020 - Specimen Procurement, Receipt and Dispatch 7141025 – Clinical Chemistry 7141030 – Hematology 7141035 – Transfusion Services 7141040 – Anatomical Pathology 7141045 – Microbiology 7141050 – Immunology 7141055 – Cytogenetics 7141060 – Tissue Typing 7141065 – Stat Laboratory 7141075 – Molecular Diagnostics	1151*, 1181*	1152*, 1153*, 1154*, 1182*, 1183*	1184*	1155*, 1185*, 1188*, 1189*
71 4 15Diagnostic Imaging 7141518 – Radiography 7141520 – Mammography 7141523 – Interventional/Angiography Studies 7141525 – Computed Tomography 7141530 – Ultrasound 7141540 – Nuclear Medicine (In Vivo) 7141555 – Cardiac Catheterization Diagnostic Services 7141570 – Magnetic Resonance Imaging	1071*	1072*, 1073*	1074*	1078*
71420 – Radiation Oncology	1161*	1162*, 1163*	1164*	1168*
71425 – Electrodiagnostic Laboratories	1071*	1072*, 1073*	1074*	1078*
71430 – Non-Invasive Cardiology and Vascular Laboratories	1071*	1072*, 1073*	1074*	1078*

Functional Centre	Inpatient Workload Accounts	Outpatient Workload Accounts	Long-Term Care Workload Accounts	Community Workload Accounts
71435 – Respiratory Services	1081*	1082*, 1083*	1084*	1085*, 1088*, 1089*
71 4 40Pharmacy 7144060 – Drug Information 7144070 – Drug Procurement and Distribution	1021*, 1031*	1032*, 1023*, 1022*	1024*, 1034*	1025*, 1026*, 1028*, 1029*, 1038*, 1039*
71445 – Clinical Nutrition	1021*	1022*, 1023*	1024*	1025*, 1026* 1028*, 1029*
71449 – Rehabilitation Services - Administration	Aggregate facility-specific workload in 71450, 71455, 71460 & 71465 functional centres and apply ratios	Aggregate facility-specific workload in 71450, 71455, 71460 & 71465 functional centres and apply ratios	Aggregate facility-specific workload in 71450, 71455, 71460 & 71465 functional centres and apply ratios	Aggregate facility-specific workload in 71450, 71455, 71460 & 71465 functional centres and apply ratios
71450 – Physiotherapy	1021*	1022*, 1023*	1024*	1025*, 1026* 1028*, 1029*
71455 – Occupational Therapy	1021*	1022*, 1023*	1024*	1025*, 1026*
71460 – Audiology and Speech Language/Pathology	1021*	1022*, 1023*	1024*	1025*, 1026*
71465 – Rehabilitation Engineering	1161*	1162*, 1163*	1164*	1168*
71470 – Social Work	1021*	1022*, 1023*	1024*	1025*, 1026*
71475 – Psychology	1021*	1022*, 1023*	1024*	1025*, 1026*
71476 – Genetic Counselling	1021*	1022*, 1023*	1024*	1025*, 1026*

Functional Centre	Inpatient Workload Accounts	Outpatient Workload Accounts	Long-Term Care Workload Accounts	Community Workload Accounts
71480 – Pastoral Care	1021*	1022*, 1023*	1024*	1025*, 1026*
71485 - Recreation	1021*	1022*, 1023*	1024*	1025*, 1026*
71490 – Child Life	1021*	1022*, 1023*	1024*	1025*, 1026*

Where workload is not reported, service activity statistics are used – either visits, exams, procedures or attendance days depending on the functional centre in question. The secondary codes associated with these statistics are:

Functional Centre	Inpatient	Outpatient	Resident	Other
71 4 05Diagnostic and Therapeutic Nursing 7140515 – Medical Imaging Nursing	4501*,8331*,8341*	4502*, 4503*, 8332*,8342*	4504*,8334*, 8344*	4505*, 4506*, 4508*, 4509*, 8338*, 8339*, 8348*, 8349*
71 4 10Clinical Laboratory 7141020 - Specimen Procurement, Receipt and Dispatch 7141025 – Clinical Chemistry 7141030 – Hematology 7141035 – Transfusion Services 7141040 – Anatomical Pathology 7141045 – Microbiology 7141050 – Immunology 7141055 – Cytogenetics 7141060 – Tissue Typing 7141065 – Stat Laboratory 7141075 – Molecular Diagnostics	4581*, 8351*,4631*, 8381*	4582*, 4583*, 4584*, 8352*, 8353*, 8354*, 4632*,4633*, 8382*,8383*	4634*,8384*	4585*, 8355*, 4635*, 4638*, 4639*, 8355* 8385*, 8388*, 8389*

Functional Centre	Inpatient	Outpatient	Resident	Other
71 4 15Diagnostic Imaging 7141518 – Radiography 7141520 – Mammography 7141523 – Interventional/Angiography Studies 7141525 – Computed Tomography 7141530 – Ultrasound 7141540 – Nuclear Medicine (In Vivo) 7141555 – Cardiac Catheterization Diagnostic Services 7141570 – Magnetic Resonance Imaging	4571*, 8361*	4572*, 4573*, 8362*, 8363*	4574*, 8364*	4578*, 8368*
71420 – Radiation Oncology	4591*,8351*	4592*, 4593*, 8352*,8353*	4594*,8354*	4598*, 8358*, 8359*
71425 – Electrodiagnostic Laboratories	4571*, 8361*	4572*, 4573*, 8362*, 8363*	4574*, 8364*	4578*, 8368*
71430 – Non-Invasive Cardiology and Vascular Laboratories	4571*, 8361*	4572*, 4573*, 8362*, 8363*	4574*, 8364*	4578*, 8368*
71435 – Respiratory Services	4681*, 8351*	4682*, 4683*, 8352*, 8353*	4684*, 8354*	4688*, 4689*, 8358*, 8359*
71 4 40Pharmacy 7144060 – Drug Information 7144070 – Drug Procurement and Distribution	4831*,8341*	4832*, 8342*	4834*,8344*	4835*, 4836*, 4838*, 4839*, 8348*, 8349*
71445 – Clinical Nutrition	4831*,8331*, 8341*	4832*, 4833*, 8342*	4834*,8334*, 8344*	4835*, 4836*, 4838*, 4839* 8338*, 8339*, 8348*, 8349*

Functional Centre	Inpatient	Outpatient	Resident	Other
71450 – Physiotherapy	4831*, 8341*	4832*, 4833*, 8342*, 8343*	4834*, 8344*	4835*, 4836*, 4838*, 4839* 8348* 8349*
71455 – Occupational Therapy	4831*, 8341*	4832*, 4833*, 8342*, 8343*	4834*, 8344*	4835*, 4836*, 4838*, 4839* 8348* 8349*
71460 – Audiology and Speech Language/Pathology	4831*, 8341*	4832*, 4833*, 8342*, 8343*	4834*, 8344*	4835*, 4836*, 4838*, 4839* 8348* 8349*
71465 – Rehabilitation Engineering	4591*,8351*	4592*, 4593*, 8352*,8353*	4594*,8354*	4598*, 8358* 8359*
71470 – Social Work	4831*, 8341*	4832*, 4833*, 8342*, 8343*	4834*, 8344*	4835*, 4836*, 4838*, 4839* 8348* 8349*
71475 – Psychology	4831*, 8341*	4832*, 4833*, 8342*, 8343*	4834*, 8344*	4835*, 4836*, 4838*, 4839* 8348* 8349*
71476 – Genetic Counselling	4831*, 8341*	4832*, 4833*, 8342*, 8343*	4834*, 8344*	4835*, 4836*, 4838*, 4839* 8348* 8349*

Functional Centre	Inpatient	Outpatient	Resident	Other
71480 – Pastoral Care	4831*, 8341*	4832*, 4833*, 8342*, 8343*	4834*, 8344*	4835*, 4836*, 4838*, 4839* 8348* 8349*
71485 - Recreation	4831*, 8341*	4832*, 4833*, 8342*, 8343*	4834*, 8344*	4835*, 4836*, 4838*, 4839* 8348* 8349*
71490 – Child Life	4831*, 8341*	4832*, 4833*, 8342*, 8343*	4834*, 8344*	4835*, 4836*, 4838*, 4839* 8348* 8349*

Statistical Hierarchy

Since it is inappropriate to either use in-house service receipt workload or in-house service activity to allocate contract-out expenses as a part of whole D&T expenses, Contract-out and in-house expenses are allocated separately.

Statistics used to break-down D&T in-house expenses are employed in the following order:

Rule	**Statistic Used to Obtain Inpatient D&T Portion**
If workload data are reported	Workload
If no workload data are reported	In-House Exams/Procedures for: 71410*-71435* (Lab, DI and Respiratory Therapy) 71465* (Rehabilitation Engineering) In-House Attendance Days for: 71440*-71490* excluding 71465* (Therapies) In-House visits for: 71405* (Diagnostic and Therapeutic Nursing)
If workload, procedures/exams or attendance days are not reported	Use the national workload average reported for the given level 3 or level 4 account
If a national workload average is not available for the given account	Use the national workload average across all level 3 or level 4 accounts

Statistics used to break-down D&T contracted-out expenses are employed in the following order:

Rule	**Statistic Used to Obtain Inpatient D&T Portion**
If service activity data are reported	Service activity: Contracted-Out Exams/Procedures for: 71410*-71435* (Lab, DI and Respiratory Therapy) 71465* (Rehabilitation Engineering) Contracted-Out Attendance Days for: 71440*-71490* excluding 71465* (Therapies) Contracted-Out Visits for: 71405* (Diagnostic and Therapeutic Nursing)
If contracted out service activity data is not reported	Use the national workload average for contracted out service activity at the given level 3 or level 4 functional centre account
If a national workload average for contracted out service activity for the given functional centre is not available	Use the national workload average for contracted out service activity across all level 3 or level 4 accounts
If a national workload average for contracted out service activity across all level 3 or level 4 accounts is not available	Apply methodology for allocating in-house D & T expenses (see above)

Special Cases – Stand Alone D&T Centres

Manitoba and B.C. operate stand-alone D&T centres. In B.C., these facilities are non-acute sites that do not provide services to the acute/hospital sector – expenses for these centres are not allocated to hospitals. They are located in small, rural communities and provide lab, radiology, emergency room services and possibly physiotherapy. It is possible that a physician is on site or situated near by. There are no inpatient services/ acute beds located at these centres, as they provide outpatient services. With regionalization, the health authorities report some of the administrative & support expenses for these centres at the regional level. These centres should be allocated a portion of the regional administrative/support expenses. In Manitoba, Westman Labs operate in the Brandon Health Authority and service both acute and non-acute sites. These expenses are allocated to individual hospitals based on additional information provided by the Manitoba Ministry of Health. For more information, please see *Appendix A – Allocation of Regional Expenses in Alberta, British Columbia, Newfoundland and Labrador, Saskatchewan and Manitoba.*

4. Allocation Methodology - Operating Room/Post-Anesthetic Recovery Room and Combined Operating and Post-Anesthetic Recovery Room (*Primary Accounts, 71260*, 71262*, 71265*, 71360, 71362*, 71365*, 71367*, 71369

4. (a) Nursing Inpatient Operating Room, Post-Anesthetic Recovery Room and Combined Operating and Post-Anesthetic Recovery Room (*Primary Accounts, 71260*, 71262*, 71265**)*
Many hospitals use their main inpatient operating, Post-Anesthetic Recovery and Combined Operating and Post-Anesthetic Recovery suite to treat both inpatient and outpatient surgical cases. Ideally nursing workload should be used to break out the inpatient/outpatient split in these functional centres. Lack of reporting of nursing workload prohibits this. Instead, surgical cases and Post-Anesthetic Recovery Room visits (PARR visits) are used instead:

Account	**Allocation Base**	**Secondary Code**
71260-Operating Room	1. Surgical visits—inpatient to client ratio 3:1‡‡‡‡ 2. If surgical visits are not reported, hospital type level average will be used to allocate expenses 3. if hospital type level average is not available, national average will be used to allocate expenses	Total Surgical Visits - 437* Inpatient Surgical Visits - 4371* Client Surgical Visits - 4372*

‡‡‡‡. The working group recommended that, while surgical cases and Post-Anesthetic Recovery Room cases could be used to obtain the inpatient/outpatient split, the difference in the resource intensity of an inpatient case to an outpatient case needed to be reflected. Inpatient cases were therefore weighted 3 to every 1 outpatient case For example, If there are 100 inpatient surgical visits and 50 client surgical visits, the total weighted surgical visits would be 300 (100 x a weighting of 3) for inpatients + 50 for clients = 350.

Account	Allocation Base	Secondary Code
71262- Combined Operating and Post-Anesthetic Recovery Room	1. Calculate a national average cost per OR visit and a national average cost per PARR visit (using data from 71 2 60 and 71 2 65), then apply these average costs to the volume of OR and PARR visits in 71 2 65, respectively (with the inpatient visits volume weighted by a factor of 3) 2. If surgical visits are not reported, hospital type level proportions of inpatient visits to total visits and client visits to total visits reported in71262 will be used to allocate expenses 3. if hospital type level proportion is not available, national proportions of inpatient visits to total visits and client visits to total visits reported in71262 will be used to allocate expenses	Total Surgical Visits - 437* Inpatient Surgical Visits - 4371* Client Surgical Visits - 4372* Total Post-Anesthetic Recovery Room Visits - 439* Inpatient Post-Anesthetic Recovery Room Visits - 4391* Client Post-Anesthetic Recovery Room - 4392*
71265- Post-Anesthetic Recovery Room	1. PARR visits—inpatient to client ratio 3:1 2. If PARR visits are not reported, hospital type level average will be used to allocate expenses 3. if hospital type level average is not available, national average will be used to allocate expenses	Total PARR Visits - 439* Inpatient PARR Visits - 4391* Client PARR Visits - 4392*

The working group recommended that, while surgical cases could be used to obtain the inpatient/outpatient split, the difference in the resource intensity of an inpatient case to an outpatient case needed to be reflected. Inpatient cases were therefore weighted 3 to every 1 outpatient case. Where surgical cases are not reported, a national average by hospital type[4] was used to allocate expenses.

4. (b) Day Surgery Operating Room, Combined Operation Room, Post-Anesthetic Recovery Room, Pre- and Post-Operative Care and Combined Operating and Post-Anesthetic Recovery Room (*Primary Accounts, 71360* 71362*, 71365*, 71367*, 71369*)*

Some hospitals also treated both inpatient and outpatient surgical cases using Day Surgery operative and Post-Anesthetic Recovery related services. Similar to methodology applied to Nursing Inpatient Services, surgical cases and Post-Anesthetic Recovery Room visits (PARR visits) are used to allocate expenses as well:

Account	**Allocation Base**	**Secondary Code**
71360- Day Surgery Operating Room	1. Surgical visits— inpatient to client ratio 3:1 2. If surgical visits are not reported, hospital type level average will be used to allocate expenses 3. if hospital type level average is not available, national average will be used to allocate expenses	Total Surgical Visits - 437* Inpatient Surgical Visits - 4371* Client Surgical Visits - 4372*

Account	Allocation Base	Secondary Code
71362- Day Surgery Combined Operating and Post-Anesthetic Recovery Room	1. Calculate a national average cost per OR visit and a national average cost per PARR visit (using data from 71 3 60 and 71 3 65), then apply these average costs to the volume of OR and PARR visits in 71 3 62, respectively (with the inpatient visits volume weighted by a factor of 3) 2. If surgical visits are not reported, hospital type level proportions of inpatient visits to total visits and client visits to total visits reported in71362 will be used to allocate expenses 3. if hospital type level proportion is not available, national proportions of inpatient visits to total visits and client visits to total visits reported in71362 will be used to allocate expenses	Total Surgical Visits - 437* Inpatient Surgical Visits - 4371* Client Surgical Visits - 4372* Total Post-Anesthetic Recovery Room Visits - 439* Inpatient Post-Anesthetic Recovery Room Visits - 4391* Client Post-Anesthetic Recovery Room - 4392*
71365- Day Surgery Post-Anesthetic Recovery Room	1. PARR visits—inpatient to client ratio 3:1 2. If PARR visits are not reported, hospital type level average will be used to allocate expenses 3. if hospital type level average is not available, national average will be used to allocate expenses	Total PARR Visits - 439* Inpatient PARR Visits - 4391* Client PARR Visits - 4392*

Account	Allocation Base	Secondary Code
71367- Day Surgery Pre- and Post-Operative Care	1. Visits - Face-to-Face 2. If visits are not reported, hospital type level average will be used to allocate expenses 3. if hospital type level average is not available, national average will be used to allocate expenses	Total Visits - Face-to-Face – 450* Inpatient Visits-4501* Client Visits- 4502*, 4503*
71369- Day Surgery Combined Operating and Post-Anesthetic Recovery Room and Pre- and Post-Operative Care	1. Calculate a national average cost per OR visit and a national average cost per PARR visit (using data from 71 3 60 and 71 3 65), then apply these average costs to the volume of OR and PARR visits in 71 3 62, respectively (with the inpatient visits volume weighted by a factor of 3) 2. If surgical visits are not reported, hospital type level proportions of inpatient visits to total visits and client visits to total visits reported in71369 will be used to allocate expenses 3. if hospital type level proportion is not available, national proportions of inpatient visits to total visits and client visits to total visits reported in71369 will be used to allocate expenses	Total Surgical Visits - 437* Inpatient Surgical Visits - 4371* Client Surgical Visits - 4372* Total Post-Anesthetic Recovery Room Visits - 439* Inpatient Post-Anesthetic Recovery Room Visits - 4391* Client Post-Anesthetic Recovery Room - 4392*

For 71362 Day Surgery Combined Operating Room and Post-Anesthetic Recovery Room: First use service recipient workload units. Calculate a national average cost per OR visit and a national average cost per PARR visit (using data from 71 3 60 and 71365), then apply these average costs to the volume of OR and PARR visits in 71362, respectively. If workload or surgical visits are not reported, use national proportions of inpatient visits to total visits and client visits to total visits reported in 71362

For 71367 Day Surgery Pre- and Post-Operative Care: First use Service-recipient workload units. If none, then use visits – face-to face.

For 71369 Day Surgery Combined Operating and Post-Anesthetic Recovery Room and Pre- and Post-Operative Care: First use service-recipient workload units. Calculate a national average cost per OR visit and a national average cost per PARR visit (using data from 71360 and 71365), then apply these average costs to the volume of OR and PARR visits in 71369, respectively. If workload or surgical visits are not reported, use national proportions of inpatient visits to total visits and client visits to total visits reported in 71369.

5. Allocation for Regional Expenses

Additional allocations must be made to hospitals that are under the control of regional health authorities. In order to do this, the portions of regional expense that are applicable to the hospitals in each region must first be separated from the portion pertaining to non-hospitals. A hospital/non-hospital ratio is obtained through the use of the non-hospital information supplied to CIHI by the provinces. Based on this information, a ratio representing the hospital sector net expenses to the net expenses of the total region is calculated:

$$\frac{\text{Total Hospital Net Expenses for the Region}}{\text{Total Net Expenses for the Region (hospital+non-hospital)}}$$

Once the hospital portion of regional expenses is obtained, it is allocated according to the proportion of each hospital's total expense to the total hospital expense for that region. This ratio is symbolized by the following formula:

$$\frac{\text{Hospital}_n \text{ Net Expense}}{\text{Net Expense of all Hospitals in the Region}}$$

Regional expenses are rolled up to level 2 and are added to the level 2 categories[§§§§] in each hospital.

[§§§§] Long-Term/Chronic Care and ICU accounts are an exception and are not rolled up to level 2 so they can absorb allocated expenses from other functional centres (eg. D&T, Administration/Support etc.).

6. Allocating Accounting Centres and Undistributed Functional Centres[*****]

If any (net) expenses or revenues remain in the undistributed functional centre or in the accounting centres they must be distributed. A ratio is calculated based on the total facility expense across each level 2 functional centre[†††††], excluding Undistributed and the Accounting Centres. The following formula is used:

$$\frac{F/C_n \text{ Expenses}}{\text{Total Expenses}^{\ddagger\ddagger\ddagger\ddagger\ddagger}(711+712_{+D\&T}+71240_{+D\&T}+713_{+D\&T}+71292+715+717+718)}$$

Where - F/C_n is each of the functional centres identified in the denominator
-D&T is the portion of D&T expenses associated with either inpatient/outpatient services.

7. Allocating Administration & Support Services

Administration & Support Services are allocated using following formula, where administration/support services are excluded from the denominator:

$$\frac{F/C_n \text{ Expenses}}{\text{Total Expenses}^{13}(712_{+D\&T}+71240_{+D\&T}+713_{+D\&T}+71292+715+717+718)}$$

8. Allocating Education

Education is allocated using the following formula, where education and research are excluded from the denominator:

$$\frac{F/C_n \text{ Expenses}}{\text{Total Expenses}^{13}(712_{+D\&T}+71240_{+D\&T}+713_{+D\&T}+71292)}$$

9. High Cost Procedure Adjustment

Certain high cost procedures are funded using a flat rate. The cost and associated days must be removed from the ward rates to avoid double counting/funding of these cases. Clinical Data from FY 2011/2012 were used to obtain the volume of cases and days associated with a given procedure.

[*****] For the purposes of this allocation, the expenses in Accounting Centres and Undistributed Functional Centres are combined.
[†††††] The only exceptions to this rule are the ICU functional centres (71240) and long-term care (71292) which are maintained at level 3 for this allocation.
[‡‡‡‡‡] If any of the functional centre groupings contain a negative cost, they are set to zero so as to permit proper allocations to the other functional centre groupings.

Procedures were counted only if they were completed in the facility reporting the procedure. Costs and days are to be removed based on an agreed upon schedule (below).

High Cost Procedure List*

High Cost Procedures	CCI Definitions	Cost
Heart Transplant	1HZ85LAXXK, 1HZ85LAXXL	$111,084
Heart and Lung Transplant	1HY85LAXXK	$156,894
Lung Transplant	1GT85LAXXJ, 1GT85LAXXK, 1GR85VCXXK, 1GR85VCXXJ, 1GR85LAXXK, 1GR85LAXXJ	$179,408
Liver Transplant	1OA85LAXXK, 1OA85VCXXK, 1OA85WLXXJ, 1OA85WLXXK	$113,809
Kidney Transplant	1PC85LAXXJ 1PC85LAXXK	$30,945
Kidney and Pancreas Transplant	1OK85XUXXK, 1OK85XVXXK	$38,126
Bone Marrow Transplant**	1LZ19HHU7A, 1LZ19HHU7J, 1LZ19HHU8A, 1LZ19HHU8J, 1WY19HHXXA, 1WY19HHXXI, 1WY19HHXXM	Varies see Appendix B)

*Established February 1998 and modified July 2012.
**Costs for Bone Marrow Transplants are also dependent on the type of bone marrow transplant, the age of the patient, and the length of stay of the patient.
Procedure counts and days are excluded from the High Cost Procedure for the case that was performed out of hospital during the patient's hospitalization (out of hospital indicator='Y') or was abandoned after onset (Intervention status attributes='A').

Costs and days will be removed at the level of agreed on payment reimbursement. This will be either the agreed upon listing provided previously or a newly calculated scheduled used to set high cost procedure cases during this recalculation activity. Alternatively a new calculation may determine that the ICU/Ward days split plus an amount for acquisition may be acceptable for reimbursement. The high cost adjustments may require only an acquisition cost adjustment.

10. Per Diem Calculation

Inpatient Days – Adjustment for Newborns

The working group recommended that newborn costs be excluded from the per diem formula, with the exception of newborn days reported in Neonatal Intensive Care functional centres (NICU)[§§§§§]. Costs for non-NICU newborns are removed at a constant rate of $373/day.

All adult/child inpatient days reported in Nursing Inpatient/Resident Services (712* functional centres excluding Long-Term/Chronic Care), Ambulatory Care Services (713* functional centres) and NICU inpatient days (71240*) are included and form the denominator for the per diem calculation[******]:

$$\frac{\text{Full Inpatient Cost-((S4034- } \textit{excluding P71240*)*388)}}{\text{Total Inpatient Days (}\textit{S4031* + (P713* S4031) + (P71240* S4034))}}$$

The denominator of the original (i.e. 2004 release) per diem rates has to be allocated into ICU and non-ICU (WARD) components. Working under the assumption that the numerators and denominators of the ICU and WARD per diems should add up to the numerator and denominator of the original per diem, the numerator and denominator of the ICU per diem and WARD per diem are to be calculated as follows:

ICU Per Diem numerator = Full ICU Inpatient Cost
ICU Per Diem denominator = Total Inpatient Days (Adult/Child (S4031) and Newborn (S4034)) reported in the ICU (P71240*)

WARD Per Diem numerator = Full WARD Inpatient Cost – (373*S4034 in 712*, excluding 71240* and 71292*)
WARD Per Diem denominator = Total Inpatient Days (Adult/Child (S4031)) in 712 * and 713*, excluding 71240* and 71292*

Trimming

Once per diem rates were calculated, the pool of hospital values was trimmed based on the peer group and the hospital status the hospital belonged to respectively.

The three peer groups are:

< 50 Beds Staffed in Operation

[§§§§§] 712 40* Intensive Care Unit is used to capture all neonatal newborn days since 712 40 50 Neonatal Intensive Care account is the minimum reporting requirements for large hospital. Reporting from a small hospital will be accepted at Level 3 (712 40*) if that is the only data available.

[******] A small number of hospitals reported their inpatient days in the accounting centres. These have been included in the denominator.

> 50 and <= 150 Beds Staffed in Operation
> 150 Beds Staffed in Operation

The four status categories are:

Non-teaching hospitals
Teaching hospitals
Pediatric hospitals
Rehabilitation hospitals

The number of beds a facility has is determined by the Beds Staffed and In Operation Statistic (secondary statistical account 825*). This number excludes beds in functional centre 71292 (Long Term Care)

The trim was based on a popular statistical rule involving the interquartile range of the data distribution. The value was trimmed if it fell outside of the following range

(Q1 – 1.5*(Q3-Q1), Q3 + 1.5*(Q3 –Q1))

where Q1 = the 25th percentile of the data range
and Q3 = the 75^{th} percentile of the data range.

Trim points were produced for each peer group. The trim was applied at a national level.

Averages

Once the trim was performed, national and provincial averages were calculated by peer group†††††† and by status.

The averages calculated are weighted averages (i.e. the sum of the numerators divided by the sum of the denominators), not arithmetic averages.
Psychiatric Facilities:

Psychiatric Nursing Unit expenses are recorded in functional centres 71275 (Psychiatry/Addiction Nursing/Resident Units) and 71276 (Mental Health Long-Term Care Nursing Unit) 71292 should not be used for Psychiatric Facilities.

Appendix A – Allocation of Regional Expenses in Alberta, British Columbia, Newfoundland and Labrador, Saskatchewan and Manitoba

Regional expenses allocation is dependent not only on those expenses reported in hospitals and non-hospitals, but under certain sector codes as well.

The details of this appendix will change based on provincial input.

For the purposes of the calculation of this indicator, a hospital expense is defined as any expense reported in a hospital or any expense reported at the Regional Health Authority (RHA) level under national sector codes 1*. Regional expenses are defined as being all expenses reported under national sector codes 001 in the RHA. Non-hospital expenses are all expenses reported in non-hospitals.

In addition, regional expenses reported under national sector codes 1* in the RHA are consider as "direct-to-hospital" expenses – that is, regional expenses for which there are no non-hospital "piece"., and are allocated only to hospitals in the given region.

In Manitoba, expenses reported under Westman Laboratories are allocated to hospitals that use its D&T services. Allocations are based on test volumes by referring facility in a file provided to CIHI directly from Westman Laboratories.

The term "expense" refers to the net expenses as specified in the Inpatient Ward Rate Methodology.

No regional allocation was made for New Brunswick, Nova Scotia, Yukon Territory, Prince Edward Island and Northwest Territories since the regional expenses have already been distributed by the provinces.

Appendix B – Costs for Bone Marrow Transplants

(Effective for discharges on or after **April 1, 2011)**

Service Code	**Service Category**	**Maximum Length of Stay (MLOS)**	**Basic Block Rate**	**Add-on Standard High Cost Per Diem over MLOS**
600	Acquisition costs (outside Canada) includes Monoclonal Antibody	--	Invoice Cost	Invoice Cost
601	Adult Autologous <72 hour discharge	--	$24,158	--
602	Paediatric Autologous <72 hour discharge	--	$28,988	--
603	Adult Autologous >72 hour	16 days	$54,355	$2,013
604	Paediatric Autologous >72 hour	13 days	$72,475	$3,623
605	Adult Allogeneic	25 days	$125,087	$2,149
606	Paediatric Allogeneic	25 days	$154,882	$3,893

For the purposes of the per diem rates, the CCI interventions were classified as follows.

Transplant Type	**CCI Codes**
Allogeneic	1LZ19HHU7J,1LZ19HHU8J,1WY19HHXXI,1WY19HHXXM
Autologous	1LZ19HHU7A,1LZ19HHU8A,1WY19HHXXA

Appendix C – Methodology for Allocation of Clearing Accounts

In all cases, clearing accounts will be allocated to an absorbing functional centre based on its direct expenses as a percentage of all direct expenses in the group of accounts receiving the allocation. If a facility reported clearing accounts under fund types other than fund type '1'(Operating), the clearing accounts will be allocated to corresponding absorbing accounts that have the same fund type.

Clearing accounts that need to be cleared are presented below:

Administrative and Support Services

The following clearing account:

7* 1 53	Plant Administration

is allocated to the 7* 1 55 (Plant Operation), 7* 1 60 (Plant Security) and 7* 1 65 (Plant Maintenance) functional centers based on their direct expenses as a percentage of the total direct expenses in 7* 1 55, 7* 1 60 and 7* 1 65 combined.

Nursing Inpatient/Resident Services

The following clearing accounts:

7* 2 05	Nursing Inpatient/Resident Administration
7* 2 05 10	Nursing Inpatient/Resident Administration
7* 2 05 20	Clinical Resources Nursing Inpatient/Resident
7* 2 05 20 20	IV Therapy
7* 2 05 20 30	Palliative Care Team
7* 2 05 20 40	Enterostomal Therapy (Centralized Service)
7* 2 05 20 60	Transplant Coordination/Organ Procurement
7* 2 07	Nursing Inpatient/Resident Medical Resources

are allocated to each 7* 2 Nursing Inpatient/Resident Services functional centre based on their direct expenses as a percentage of the total direct expenses in all 7* 2 accounts.

The following clearing account:

7* 2 20 09	Preoperative Services Nursing Unit

is allocated to each 7* 2 20 Surgical Nursing Unit functional center based on their direct expenses as a percentage of the total direct expenses in all 7* 2 20 accounts.

Ambulatory Care Services

The following clearing accounts:

7* 3 05	Ambulatory Care Administration
7* 3 07	Ambulatory Care Medical Resources

are allocated to each 7* 3 Ambulatory Care Services functional center based on its direct expenses as a percentage of the total direct expenses in all 7* 3 accounts.

The following clearing account:

7* 3 50 05	Specialty Clinic Administration

is allocated to each 7* 3 50 Specialty Clinics functional center based on its direct expenses as a percentage of the total direct expenses in all 7* 3 50 accounts.

Diagnostic and Therapeutic Services

The following clearing accounts:

7* 4 10 10	Clinical Laboratory Administration
7* 4 10 15	Clinical Laboratory Centralized Support Services

are allocated to each 7*4 10 (Clinical Laboratory) functional centre based on its direct expenses as a percentage of the total direct expenses in all 7* 4 10 accounts.

The following clearing account:

7* 4 15 10	Diagnostic Imaging Administration

is allocated to each 7* 4 15 (Diagnostic Imaging) functional centre based on its direct expenses as a percentage of the total direct expenses in all 7* 4 15 accounts.

The following clearing account:

7* 4 35 10	Respiratory Services – Administration

is allocated to each 7* 4 35 (Respiratory Services) functional centre based on its direct expenses as a percentage of the total direct expenses in all 7* 4 35 accounts.

The following clearing account:

7* 4 40 10 Pharmacy – Administration

is allocated to each 7* 4 40 (Pharmacy) functional centre based on its direct expenses as a percentage of the total direct expenses in all 7* 4 40 accounts.

The following clearing account:

7* 4 49 Rehabilitation Services - Administration

Is allocated to the 7* 4 50 (Physiotherapy), 7* 4 55 (Occupational Therapy), 7* 4 60 (Audiology and Speech/Language Pathology), 7* 4 65 (Rehabilitation Engineering), 7* 4 70 (Social Work), 7* 4 75 (Psychology), 7* 4 76 (Genetic Counseling), 7* 4 80 (Pastoral Care), 7* 4 85 (Recreation) and 7* 4 90 (Child Life) functional centres based on their direct expenses as a percentage of the total direct expenses in 7* 4 50, 7* 4 55, 7* 4 60, 7* 4 65, 7* 4 70, 7* 4 75, 7* 4 76, 7* 4 80, 7* 4 85 and 7* 4 90 combined.

Community and Social Services

The following clearing accounts:

7* 5 05 Community and Social Services Administration
7* 5 07 Community Medical Resources

are allocated to each 7* 5 (Community) functional centre based on its direct expenses as a percentage of the total direct expenses in all 7* 5 accounts.

Undistributed Food Services

The following clearing accounts:

8* 9 10Food Services Clearing Account
8* 9 10 05 Food Services Administration
8* 9 10 20 Food Services Production
8* 9 10 30 Food Services Tray Assembly and Distribution
8* 9 10 40 Food Services Warewashing

are allocated to 7* 1 95(Patient/Resident/Client Food Services) and 7* 9 10(Non-Service Recipient Food Services) functional centres based on their direct expenses as a percentage of the total direct expenses in 7* 1 95 and 7* 9 10 combined.

Appendix G . Pairwise Comparison of Costs

Figure 37. Pairwise Comparison of Monthly Costs– Entire Observational Period

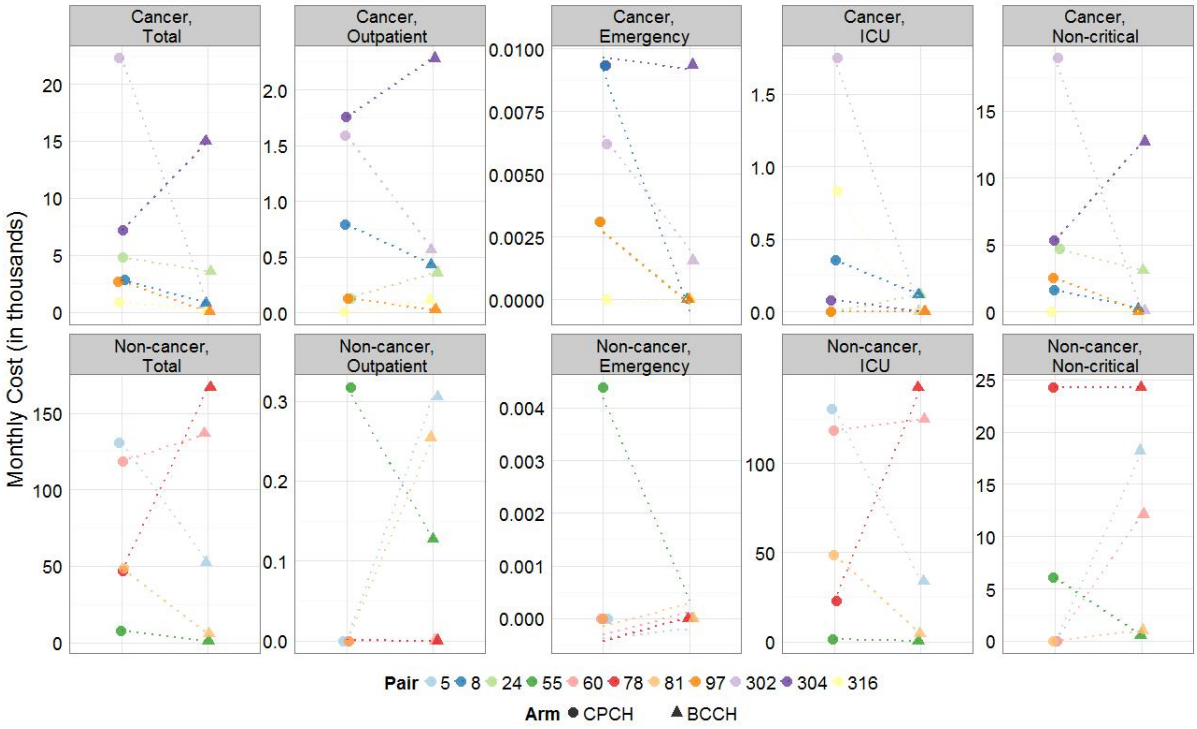


Figure 38. Pairwise Comparison of Monthly Costs – Pre-Referral Period.

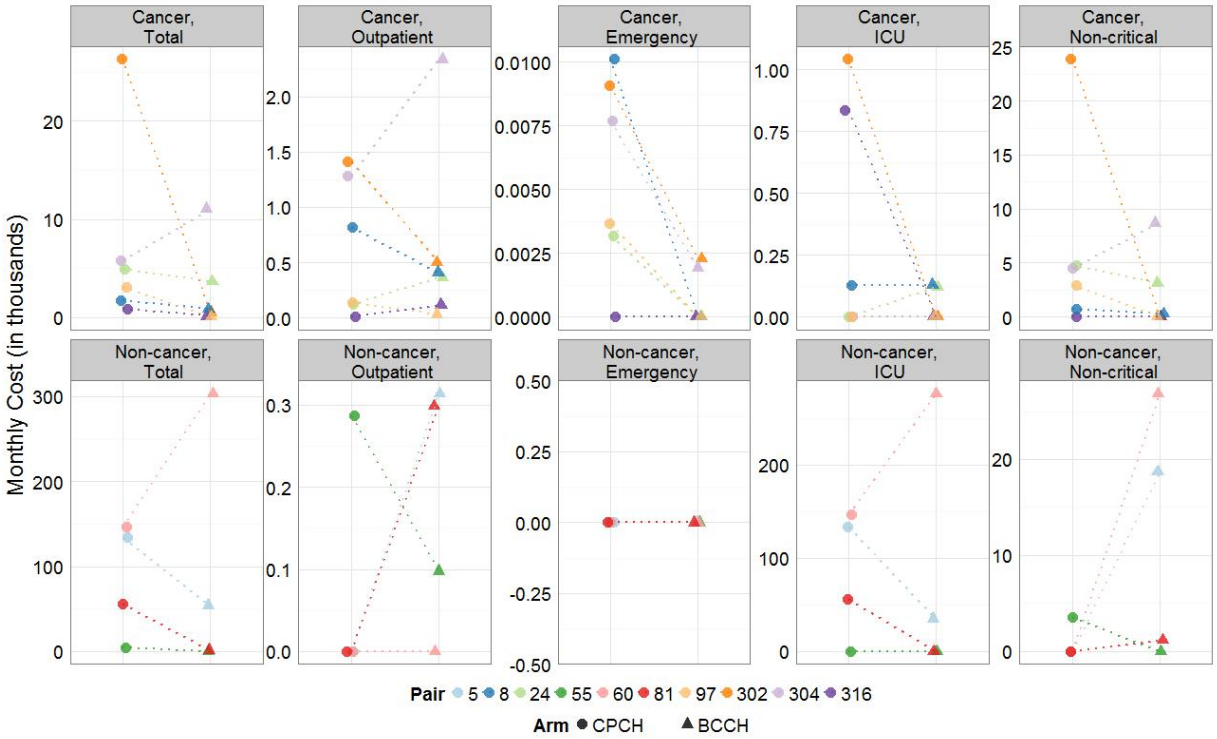


Figure 39. Pairwise Comparison of Monthly Costs – Post-Referral Period.

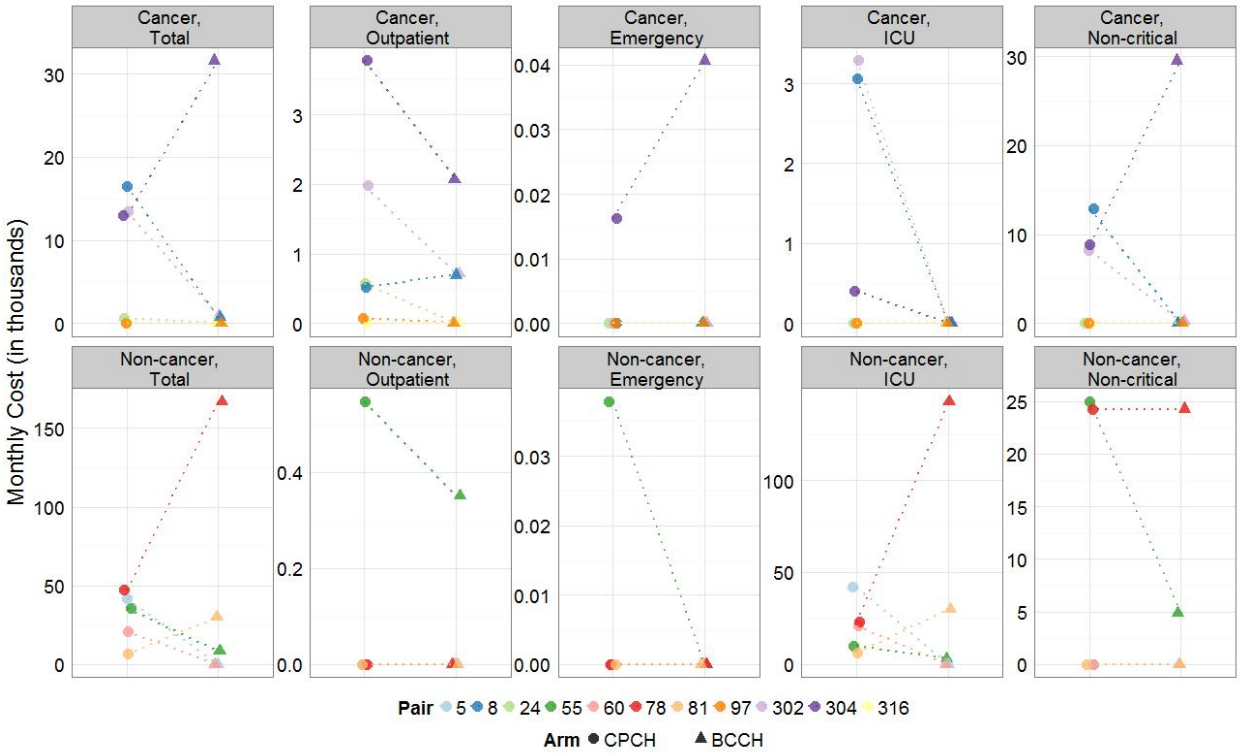


Figure 40. CPCH Arm – Changes in Monthly Costs – Pre- to Post-Referral Period.

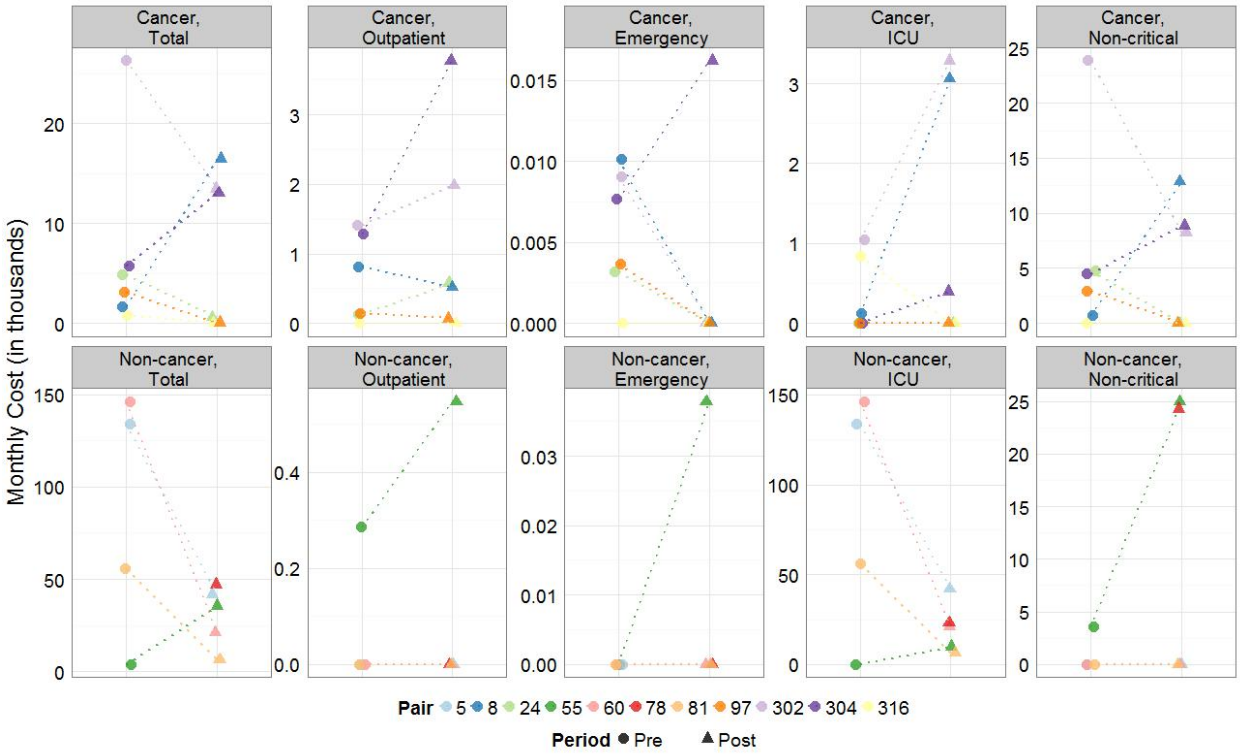


Figure 41. BCCH Arm – Changes in Monthly Costs – Pre- to Post-Referral Period.